Shared Surgical Decision-Making and Youth Resilience:
Correlates of Satisfaction with Clinical Outcomes

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Abstract

The aim of this study was to identify factors associated with youth satisfaction with surgeries performed to address oral cleft or craniofacial conditions (CFC). It was hypothesized that youth mental health, participation in decision-making, perceived consequences of living with a CFC, and coping strategies would be associated with satisfaction with past surgeries.

Two hundred and three youth between the ages of 11 and 18 (Mean age = 14.5. SD = 2.0; 61% male; 78% oral cleft) completed a series of questionnaires measuring depression, self-esteem, participation in decision-making, condition severity, negative and positive consequences of having a CFC, coping, and satisfaction with past surgeries.

Multiple Regression Analysis using boot-strapping techniques found that youth participation in decision making, youth perception of positive consequences of having a CFC, and coping accounted for 32% of the variance in satisfaction with past surgeries ($p < .001$). Youth age, sex, and assessment of condition severity were not significantly associated with satisfaction with surgical outcome. Depression, self-esteem, and negative consequences of having a CFC were not associated with satisfaction with past surgeries.

Youth should be actively involved in the decision for craniofacial surgery. Youth who were more satisfied with their surgical outcomes also viewed themselves as having gained from the experience of living with a CFC. They felt that having a CFC made them stronger people and they believed that they were more accepting of others and more in touch with others' feelings because of what they had been through.

Oral Cleft; Craniofacial Conditions; Resilience; Decision-Making; Surgery Satisfaction

A major goal of treatment for oral clefts and craniofacial conditions in adolescence is improved quality of life. Reconstructive surgery and other medical therapies (dental, orthodontic, hearing and speech) are the primary intervention tools used to meet this broad goal, while individual counseling/therapy and school interventions are used less frequently.[1] When facial reconstructive surgery is the treatment of choice for patients with craniofacial conditions, improved physical appearance is the goal with the spoken or unspoken hope that this change in appearance will result in an improved quality of life.[2-4] Research has demonstrated that a technically well-performed surgery does not necessarily result in a satisfied patient.[4, 5] However we do not really understand how a patient develops their sense of satisfaction or disappointment. The disconnect between the assumed benefit of improved appearance on quality of life with evidence that a successful surgical experience does not necessarily lead to a satisfied patient points to the need for a closer look at these issues.

Children with facial disfigurement due to oral clefts or other craniofacial conditions (CFCs) are unique in the plastic surgeon's patient population. Children born with CFCs are typically involved with a team of providers, including a plastic/craniofacial surgeon, from infancy. This ongoing involvement both normalizes the contact with a surgeon and promotes a relationship with the family and child. It also psychologically connects appearance related concerns with functional problems and the underlying diagnosis. This "medicalizes" the issue of appearance dissatisfaction or difference. Conversations intertwining appearance and function happen with multiple providers over years, reinforcing this interplay. Thus discussions about how appearance concerns affect an individual's quality of life are seldom separated from other physical concerns and surgical options are sometimes promoted to solve social problems.

When a youth comes to a cleft/craniofacial team consultation there is often an assumption that the youth and his/her family will be interested in learning about and pursuing the next step in treatment, including a surgery to address appearance issues, if it is indicated or is part of a team protocol. In many instances the youth and/or family will have clearly articulated goals for appearance changes due to the many previous discussions, while in other situations the youth and family will not have considered surgery. Especially in the latter case, when a decision for surgery is actually made, it is not always clear whether parent, youth, or surgeon has been the primary impetus for that decision.[4, 6, 7]

There has been little direct research investigating how decisions for surgery within cleft/craniofacial teams are made.[7] Turner et al[6] reported that 23% (7/30) of the 15 year-olds and 15% of the parents of all ages of children (19/130) felt excluded from the treatment planning and decision making process. In that study adolescents reported significantly less satisfaction with their overall appearance than their parents; however, satisfaction with appearance was not linked to the adolescents' perceptions of their participation in the decision-making process. On the other hand, Lefebvre and Munro[8] and Phillips and Whitaker[9] noted that adolescent patients who were ambivalent about surgery or who had felt pressured into surgery were more likely to express dissatisfaction with the outcome. More recently, Bemmels et al[4], based on clinical interviews, concluded that child involvement in surgical decision making was critical whenever child age and cognitive capacity supported such participation. Noor and Musa,[10] emphasizing the importance of child involvement in treatment decisions, reported that 38% of adolescents with CLP felt that they were 'usually involved' while 8% reported they were 'never involved'.

More research has focused on satisfaction of adolescents and families with treatment outcomes. Strauss, Broder, and Helms[11] found that nearly 40% of adolescents with clefts were not fully satisfied with their appearance. Satisfaction with appearance did not differ between younger and older subjects.[12] Pope and Ward[13] reported that negative self-perceived facial appearance related to several aspects of social adjustment including greater loneliness, fewer same-sex close friends, and parents' reports of withdrawal and being disliked by peers in youth aged 11 to 13. Feragen, Borge & Rumsey[14] reported that better emotional functioning, high satisfaction with appearance, and a lower frequency of reported teasing were more important than cleft visibility, child gender or presence of an additional diagnosis in predicting child resilience. These studies did not directly investigate the relationship between adolescent participation in surgical decision-making and satisfaction with surgery or psychological adjustment of the adolescent after surgery; nor did they determine if there were events that occurred during the surgical encounter, hospitalization or recuperation that might have impacted their assessment of the impact of surgical intervention

The purpose of this paper is to investigate the relative impact of psychological adjustment issues and adolescent participation in surgical decision making on satisfaction with surgical outcome. We have intentionally included children with a wide range of craniofacial conditions and severity in this study in order to allow results to generalize to the broader population of youth with craniofacial conditions. Previous research has found that it is the *fact* of a facial difference and the *youth's perceptions and judgements* about those differences rather than the type or severity of difference that influences self-concept and psychological adjustment (Marik and Hoag, 2012). Specific hypotheses are: 1) Adolescent psychological health including positive self-concept and freedom from depression is associated with satisfaction with surgical outcome; 2) Active participation in surgical decision making is associated with increased satisfaction with surgical outcome; and 3) Coping abilities and attitudes about the impact of CFCs are associated with positive surgical outcome.

Materials and Methods

Sample

All participants were recruited as part of a multisite observational study examining the impact of congenital and acquired CFCs on the health and quality of life of adolescents. Participants were enrolled after informed consent had been obtained. The study was approved by the institutional review boards (IRB) from each participating center: the University of Washington (Seattle), Northwestern University (Chicago), the University of North Carolina (Chapel Hill), and University of the West of England (Bristol). The sites partnered with cleft-craniofacial centers at Children's Hospital and Regional Medical Center in Seattle (now Seattle Children's), Shriners Hospitals for Children in Chicago, the UNC Craniofacial Center at the University of North Carolina at Chapel Hill, Great Ormond Street Hospitals (London), and the South West UK Cleft Team in Bristol, England.

Patients

Youth between ages 11 to 18 years who presented to one of the craniofacial clinics with a visible facial difference and who were able to speak and read English at a fifth-grade level were recruited. Of the 284 potential subjects contacted, 203 (71%) were deemed eligible and agreed to participate. Youth were considered ineligible if their primary caregiver indicated that they had a co-morbid mental or physical condition that had a greater impact on their life than their facial difference. Across four sites, only five youth were excluded based on that criteria: three because of coexisting conditions the parent estimated had a greater impact than the facial difference (learning disability, overweight, attention-deficit hyperactivity disorder), one who had cleft palate only (not visible), and one who was not able to comprehend language at the fifth-grade level based on parent report. All enrolled participants gave consent/assent to be in the study following the protocol of their respective IRB. (See Table 1 for sample characteristics)

Measures

Data for this study comes from a larger investigation of quality of life for youth with craniofacial conditions. Measures included in this study were: *Facial Differences Module of Youth Quality of Life Instruments* (*YQOL-FD*),[15] the *Craniofacial Surgery Attitudes Measure* (*CSAM*) (Patrick, Edwards, Topoloski, Kapp-Simon, Strauss, unpublished), the *Children's Depression Inventory* (*CDI*),[16] and the *Child Health Questionnaire* (*CHQ*).[17]

Facial Differences Module of the Youth Quality of Life Instruments (*YQOL-FD*)**.** The YQOL-FD is a validated craniofacial specific quality of life (QoL) instrument for youth ages 11 to 18 years with a range of congenital and acquired craniofacial conditions.[15] Forty-eight items make up five domains: Stigma, Negative Self-Image, Positive Consequences, Negative Consequences, and Coping. Cronbach alphas and intraclass correlation coefficients for all domains exceed .70.[15] Scales of the *YQOL-FD* were found to correlate highly with scales of the *Youth Quality of Life Instrument Research Version- Adolescent,*[18, 19] a validated generic QoL instrument previously used with adolescents with facial differences.[15] The Positive Consequences, Negative Consequences and Coping Domain total scores were used in analyses.

Craniofacial Surgery Attitudes Measure (*CSAM*)**.** The CSAM is a new instrument constructed for use with youth aged 11 to 18 years in order to evaluate self-perceptions specifically in relationship to past or future surgeries. Past surgery domains include: Negative Feeling, Positive Outcomes, Negative Consequences, and Negative Self-Image. Future surgery domains include the Need for More Surgery, Aesthetic; the Need More Surgery, Function; Confidence in Surgery; and Worry About Surgery. For the purpose of this study, the Past Surgery Positive Outcomes domain (CSAM, Past Surgery-Positive Outcomes) was used as a measure of satisfaction with past surgeries. This domain has been found to have good internal consistency with a Cronbach's alpha of .75 (Patrick, et al. unpublished).

Participation in Decision Making: Two questions, both focused on youth involvement in decision making within their own family, were totaled in order to yield a measure of youth decision-making: "My parents involve me in deciding if and when I have surgery on my face or head," and "I feel my parents or guardians allow me to participate in important decisions which affect me". Ratings were made on a scale of 0 to 10 with 0 indicating no participation ("not at all") and 10 indicating a "great deal" of participation in the decision making process.

Children's Depression Inventory (*CDI*). The CDI provides a direct assessment of the presence and severity of depressive symptoms in children and adolescents ages 7 through 17 years. The CDI provides a total score and factor scores on five domains of depression are calculated from 27 items. Internal consistency reliability estimate for the normative sample on the CDI is .86 for the total score. The CDI correlates significantly with other assessment of depressive symptoms including the *Reynolds Adolescent Depression Scale* (*RADS*) and corresponding subscales of the *Behavior Problem Checklist.*[20] The CDI total score was used in this study. A higher score indicates a greater number of depressive symptoms.

Child Health Questionnaire (*CHQ*). The CHQ measures health-related quality of life from the patient's perspective. It is a paper-and-pencil self-report measure with 12 scales measuring health concerns, illnesses/health conditions, and sociodemographics. All questions are based on a retrospective recall of health in the preceding 4 weeks with the exception of one single-item scale (Change of Health Scale). It was developed for use with youth ages 10 through 18 years. For the purpose of this study, only the Self-Esteem Scale was analyzed. It is a 13 item scale with Cronbach alpha coefficient of .92.[21] A higher score on this scale indicates poorer self-esteem.

Severity Index: All youth participants, a parent (generally mother), and a professional (generally the surgeon) rated the severity of the medical condition affecting the face on a scale from 1 (not at all severe) to 7 (extremely severe). Youth, parent, and professional severity indices are reported separately.

Procedure

Potential research subjects were identified from clinic lists and physician referral. At least one parent was present at the time of the initial interview. Informed consent and assent was obtained from each subject prior to participation in the research following approved IRB protocols. The research instruments were administered at clinical or research locations at each of the sites. The interviewer determined reading ability by administering the first 8 sentence comprehension items of the American Guidance Service (AGS) Reading Level Indicator. The items reflected a 1st to 5th grade reading level. If less than 6 out of the 8 items were completed correctly, the interviewer read the questions on the research questionnaires to the adolescent. Once questionnaires were completed, the interviewer checked though them to make sure questions were not missed. Adolescents received $20 as compensation for time required completing the entire research packet, which took approximately 1 hour.

Data Analysis

Descriptive statistics were developed for study variables and differences by sex and age group were determined using analysis of variance (ANOVA). Age groups were defined as 11-14 and 15-18 years with males and females forming separate groups within each age. Two tailed Pearson correlational analysis was used to examine the relationships among the study variables. Preliminary analysis showed that self, parent, and professional ratings of condition severity were significantly correlated (See Table 2). Self-ratings of severity were judged most salient[22] and used in the final analyses to conserve power and maximize the number of subjects included in the analyses

Multiple regression analysis (MRA) was used to evaluate the relative contributions of youth age, sex, self-perception of condition severity, self-esteem (CHQ Self Esteem scale), the degree to which youth endorsed positive (YQOL-FD Positive Consequences) and Negative (YQOL-FD Negative Consequences) consequences of having a facial difference, youth use of positive coping strategies (YQOL-FD Coping), youth Participation in Decision-Making and youth depression (CDI Total score) on youth perception of outcomes from previous surgeries (CSAM Past Surgery-Positive Outcomes scale). Age (continuous), Sex, and Self-reported Severity of Medical Condition affecting the youth's face (Severity) were entered first as control variables; Participation in Decision-Making was entered in the second block; Positive and Negative Consequences of having a facial condition and Coping were entered in the third block; and Depression and Self-Esteem were entered in the final block. Bootstrapping techniques based on 1,000 random samples were used to derive robust estimates of standard errors and confidence intervals for the regression analysis. Those variables contributing to an R^2 change with a probability greater than 1.0 and control variables were retained and the MRA was rerun. Data analysis was completed using the SPSS 20 statistical package.

RESULTS

Preliminary Analysis

Table 1 provides demographic information on the 203 subjects who made up the subject pool available for analysis. The majority of the subjects were male (61%), white (76%) and had a diagnosis of cleft lip and palate (64%). Younger subjects, ages 11-14, made up 54% of the sample and older subjects, ages 15-18, made up 46% of the sample.

Correlations among the variables are provided in Table 2. Note that higher scores indicate poorer self-esteem and more symptoms of depression. Significant correlations were found between CSAM Past Surgery-Positive Outcomes and Youth Participation in Decision-Making, YQOL-FD Positive Consequences, YQOL-FD Coping, and CHQ Self-Esteem. CDI Total Depression correlated with CSAM Past Surgery-Positive Outcomes in the expected direction but was not significant. Youth, Parent and Professional Severity ratings demonstrated low but significant correlations with each other, but only Youth Severity correlated significantly with other variables including CDI Total, CHQ Self-Esteem and YQOL-FD Negative Consequences. YQOL-FD Positive Consequences and YQOL-FD Coping correlated at .54 ($p < .001$) and CHQ Self-Esteem and CDI Total Depression correlated at .69 ($p < .001$).

Table 3 presents means and standard deviations as function of sex and age and the results of one-way ANOVAs for each study variable. There were no differences by age or sex for the majority of variables. Parents rated younger females as having greater condition severity than males at either age but not more severe than older females. Older subjects of both sexes reported that they were more involved in decision-making than younger subjects with older females significantly more involved than younger males. Females regardless of age reported more positive consequences from having a craniofacial condition than did males though differences were significant only for older females.

Multiple Regression Analysis

Initial regression analysis did not support a contributing role for YQOL-FD Negative Consequences, CDI Total Depression, or CHQ Self-Esteem as predictors of CSAM Past Surgery-Positive Outcomes and these variables were dropped from further analyses. Table 4 presents the final regression analysis including standardized regression coefficients and confidence intervals, and R^2 change for each block for the prediction of CSAM Past Surgery-Positive Outcomes. Control variables including youth age, sex and Severity did not contribute significantly to perceptions of outcome at step 1. Adding Participation in Decision-Making at step 2 resulted in a significant R^2 change of .08 (F change = 16.32, $p < .001$). At step 3 adding YQOL-FD, Positive Consequences and Coping resulted in an R^2 change of .24 (F Change = 31.04, $p < .001$) while Participation in Decision-Making remained a significant contributor.

DISCUSSION

The goal of this paper was to determine what factors influence perceptions of positive surgical outcome for youth with visible craniofacial conditions. In particular we were interested in learning to what extent psychological health, attitudes about the craniofacial condition, adaptive coping strategies, and youth participation in decision making were associated with perceptions of positive surgical outcome.

Our first hypothesis that youth mental health, as indexed by level of depression and self-esteem, would be associated with perceptions of positive outcomes of previous surgeries was not upheld. Subjects reporting greater depression also reported poorer self-esteem, more negative consequences of having a CFC and greater condition severity (see Table 2), but these variables were not associated with the perceptions that surgery improved their lives.

Our second hypothesis that active participation in surgical decision making would be associated with positive perceptions of surgical outcome was upheld with Participation in Decision-Making accounting for 8% of the variance in CSAM Past Surgery-Positive Outcomes. Age was correlated with decision-making such that older youth were more likely to report that parents involved them in the decision-making process than younger youth; however, in the regression analysis Participation in Decision-Making was significantly associated with youth perceptions of positive outcomes even after controlling for age. Thus giving youth a voice in the decision-making process increased their perceptions that the surgery was beneficial regardless of the youth's age.

There is growing recognition in the craniofacial literature that youth participation in surgical decision making is critical. In this study youth involved in the decision to have surgery were more likely to report that they were happy with the results of surgery, looked better after surgery, that surgery improved their lives, and that they got along better with others since surgery. Unfortunately, our data does not provide insight into how the youth participated in the decision-making process. Youth were reflecting retrospectively on the extent to which they perceived they were involved in the decision for surgery and the extent to which their opinions were ‘heard’ within their family. This is a very limited measure of the decision-making process. It does not take into account what the child was actually told about the surgery or who provided the information.

As was described earlier, youth with CFCs will be seen by a variety of providers throughout their childhood. It is probable that surgical options have been described over time and by many different medical professionals. Youth and parents/guardians may have had discussions about the procedures and the youth may well know their parents/guardians’ position. The cumulative effect of these interactions and discussions provide the backdrop that influences the atmosphere on the day a specific procedure is discussed. Additionally, the context of that conversation must be considered. For example, did the surgeon speak directly to the youth about the proposed surgery and explain the benefits and risks? Or was information provided through an adult-filter, giving the young person only what the adult thought the youth could handle?

Youth need to be involved in the discussions about their bodies, whether surgery is elective, as in a lip revision for a child with cleft lip; or necessary to support function, as in a midface advancement to improve breathing and protect the eyes in a child with Crouzon’s Syndrome. Shared decision making means getting the child and family’s impressions up front, before we give our professional opinion. While as medical practitioners we may feel that the presence of significant cleft lip scar or cleft-related nasal deformity should “convince” the youth and family of a problem and always lead to surgery, it is important to first determine the perspective of the youth and family.

The third hypothesis investigated the extent to which coping abilities and attitudes about the impact of CFCs are associated with youth perception of positive surgical outcome. The final analysis demonstrated that 24% of the variance in satisfaction with surgery was accounted for by youth perceptions of positive consequences of having a CFC. The items that make up the YQOL-FD Positive Consequences scale fall under the category of “Personal Growth” identified by Egan, Harcourt, & Rumsey[23] as one of the characteristics of adults who felt that they were coping well with their physical differences. Essentially the youth who were more satisfied with their surgical outcomes also viewed themselves as having gained from the experience of living with a CFC. They felt that having a CFC made them stronger people. And they believed that they were more accepting of others and more in touch with others’ feelings because of what they had been through.

The items that make up the YQOL-FD Positive Consequences scale (like all of the YQOL-FD items) were generated through face to face discussions with youth who were living with a CFC. During these interviews youth identified ways in which having a CFC improved their outlooks and made them, in their minds, stronger and better people. In the current study youth who endorse these positive consequences demonstrated better mental health. They were less likely to be depressed and they reported more positive self-esteem. But more than that they seem to have embraced their facial differences such they believed having a CFC was of benefit to them. Treatment of children with CFCs needs to focus on fostering this type of resiliency. It may well be that engaging youth in the decision-making process from an early age would allow them to develop confidence in their own abilities and subsequently increase their sensitivity to the feelings and needs of others. It may also deliver the message from the clinicians that a decision to change to a person’s body needs to be a shared decision. The team of clinicians is responsible to make a recommendation based on their technical assessment, training and experience. This must be balanced with the patient’s perspective since they bear the direct benefits and risk of any surgical treatment. Through this process the clinicians will foster an ongoing message to the parents of young children with CFCs about the impact their attitudes toward the CFC will have on their child's adjustment and their willingness to talk about their hopes and fears, both in general and specifically related to proposed surgery. Parents are the child's first teachers. How they talk about the child's CFC to each other and outsiders and how they respond to others reactions to the CFC will teach the child how to respond. These interactions teach the child what meaning to attribute to the CFC and will ultimately influence how the child thinks about the CFC. The interplay between how a child feels internally about their situation, and what people tell them about what they see happening externally, may help to reduce the worry children have about having these discussions. The reduction in worry could then directly impact the child's feelings about him/herself, which in turn affects how the child behaves.

Taken as a whole, this data underlines the importance of the child's general psychological status and specific personal adaptation to his or her craniofacial condition. Consistent with tenets of cognitive psychology, the meaning the children attribute to the presence of their condition significantly impacts their ability to effectively cope with their condition.[24, 25] Further research is required to understand the complexity of the interaction of child psychological status and appraisal of surgical outcomes. The data from this study point to the importance of directly assisting parents and children in attaining a positive adaptation to their craniofacial condition with an emphasis on capabilities and potential. Currently, it is not entirely clear how these positive adaptations are made but encouraging conversations about the child's feelings and coping strategies is critical. In addition, our findings indicate that the direct involvement of children in the informational and decision making processes related to surgery appears to have a positive impact on their perceptions of the benefits of surgery and its outcomes. Because our goal is to foster improved patient reported quality of life, we have an obligation to continue studying these linkages to better understand what truly helps people born with craniofacial conditions.

References

1. Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial anomalies. American Cleft Palate-Craniofacial Association. *Cleft Palate Craniofac J.* 1993;30(Suppl 1 [Latest revision 2009]):http://www.acpa-cpf.org/uploads/site/Parameters_Rev_2009.pdf.

2. Pruzinsky T, Sarwer DB, Cash TF, et al. Multiple perspectives on the psychology of plastic surgery. In: Sarwer DB, Pruzinsky T, Cash TF, et al., eds. *Psychological Aspects of Reconstructive and Cosmetic Plastic Surgery: Clinical, Empirical, and Ethical Perspectives* Philadelphia: Lippincott Williams & Wilkins; 2005:3-11.

3. Marsh JL. To cut or not to cut? A surgeon's perspective on surgically shaping children. In: Parens E, ed. *Surgically Shaping Children: Technology, Ethics, and the Pursuit of Normality*. Baltimore: The Johns Hopkins University Press; 2006:113-124

4. Bemmels H, Biesecker B, Schmidt JL, et al. Psychological and social factors in undergoing reconstructive surgery among individuals with craniofacial conditions: An exploratory study. *Cleft Palate Craniofac J.* 2013;50(2):158-167.

5. Rumsey N, Hardcourt D. Visible difference amongst children and adolescents: Issues and interventions. *Dev Neurorehabil.* 2007;10(2):113-123.

6. Turner SR, Thomas WN, Dowell T, et al. Psychological outcomes amongst cleft patients and their families. *Br J Plast Surg.* 1997;50:1-9.

7. Alderson P. Who should decide and how? In: Parens E, ed. *Surgically Shaping Children: Technology, Ethics, and the Pursuit of Normality*. Baltimore: The John Hopkins University Press; 2006:157-175

8. Lefebvre A, Munro IR. Psychological adjustment of patients with craniofacial deformities before and after surgery. In: Herman P, Zannna MP, Torry Higgins E, eds. *The Ontario Symposium*. Hillsdale NJ: Lawrence Earlbaum Associates; 1986:53-62.

9. Phillips J, Whitaker LA. The social effects of craniofacial deformity and its correction. *Cleft Palate J.* 1979;16:7-15.

10. Noor SN, Musa S. Assessment of patients' level of satisfaction with cleft treatment using the Cleft Evaluation Profile. *Cleft Palate Craniofac J.* 2007;44(3):292-303.

11. Strauss RP, Broder H, Helms RW. Perceptions of appearance and speech by adolescent patients with cleft lip and palate and by their parents. *Cleft Palate J.* 1988;25(4):335-342.

12. Broder HL, Smith FB, Strauss RP. Habilitation of patients with clefts: parent and child ratings of satisfaction with appearance and speech. *Cleft Palate Craniofac J.* May 1992;29(3):262-267.

13. Pope AW, Ward J. Self-perceived facial appearance and psychosocial adjustment in preadolescents with craniofacial anomalies. *Cleft Palate Craniofac J.* 1997;34(5):396-401.

14. Feragen KB, Borge AI, Rumsey N. Social experience in 10-year-old children born with a cleft: Exploring psychosocial resilience. *Cleft Palate Craniofac J.* Jan 2009;46:65-74.

15. Patrick DL, Topolski TD, Edwards TC, et al. Measuring the quality of life of youth with facial differences. *Cleft Palate Craniofac J.* 2007;44(5):538-546.

16. Kovacs M. *Children's Depression Inventory (CDI)*. North Tonawanda, NY: Multi-Health Systems; 1992.

17. Langraf JM, Abetz L, Ware JE. *The Child Health Questionnaire (CHQ): A User's Manual 2nd Printing*. Boston, MA: HealthAct; 1999.

18. Edwards TC, Huebner CE, Connell FA, et al. Adolescent quality of life, part I: Conceptual and measurement model. *J of Adolesc.* 2002;25:275-286.

19. Patrick DL, Edwards TC, Topolski TD. Adolescent quality of life, part II: Initial validation of a new instrument. *J of Adolesc.* 2002;25:287-3000.

20. Kovacs M. *Children's Depression Inventory (CDI) Technical Manual Update*. Tonawanda, NY: Multi-Health Systems; 2003.

21. Raat H, Bonsel GJ, Essink-Bot ML, et al. Reliability and validity of comprehensive health status measures in children: The Child Health Questionnaire in relation to the Health Utilities Index. *J Clin Epidemiol.* 2002;55:67-76.

22. Moss TP. The relationships between objective and subjective ratings of disfigurement severity, and psychological adjustment. *Body Image.* 2005;2(2):151-159.

23. Egan K, Harcourt D, Rumsey N. A qualitative study of the experiences of people who identify themselves as having adjusted positively to a visible difference. *J Health Psychol.* 2011;16(5):739-749.

24. Cochrane VM, Slade P. Appraisal and coping in adults with cleft lip: Associations with well-being and social anxiety. *Br J Med Psychol.* Dec 1999;72 (Pt 4):485-503.

25. Kendall PC. Guiding theory for therapy with children and adolescents. In: Kendall PC, ed. *Child and Adolescent Therapy, Fourth Edition*. New York: Guilford Press; 2012:3-24.