

Evaluating aesthetics of the nasolabial region in children with cleft lip and palate: professional analysis and patient satisfaction

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Abstract: Cleft lip and palate is one of the most common deformities of the craniofacial region, and treatment of this deformity is essential for social reintegration. One of the major goals of surgery and treatment of craniofacial deformities is to improve the aesthetic appearance of the face, and thereby improve the patient's social acceptability. Here, we present a critical review of the criteria for aesthetic evaluation of the nasolabial region in cleft patients by assessing publications with the highest level of evidence, including professional evaluation, and patient satisfaction. The findings indicate treatment of this condition represents a major challenge for multidisciplinary team care.

Keywords: cleft lip palate, craniofacial deformity, nasolabial appearance

Introduction

Cleft lip and palate is one of the most common deformities of the craniofacial region.¹⁻³ The main goal of surgery and dental treatment of craniofacial deformities is aesthetic improvement of the face, and consequent enhancement of the acceptability of the individual in society.⁴⁻⁶ The development of surgical techniques has improved the patients' quality of life in severe cases of cleft lip and palate.⁷ Currently, the most widely used surgical protocol involves lip repair at 3 months of age, and palatoplasty in a single procedure at 18 months of age. The alveolar bone graft is performed between ages 7 and 11 years, around the time of eruption of lateral incisors, if present, or the permanent canines adjacent to the cleft. Orthognathic surgery, when necessary, is performed at the age of skeletal maturity. The final surgery is cleft secondary rhinoplasty for correction of possible residual nasal deformity.⁸

The appearance of the nasolabial region is undoubtedly one of the most important measures for evaluating the success of a treatment.⁹ Although there are many reports of surgical correction of cleft lip and palate, the results are often analyzed in a subjective and nonstandardized manner and research in this field has encountered difficulties due to a lack of tools for assessing aesthetic conditions of the face.⁷ It is widely accepted that facial appearance is important for first impressions, and can reinforce or induce a social stereotype.¹⁰ Almeida¹¹ demonstrated that individuals behave differently in the presence of others who look good versus those with an unsightly appearance. Moreover, Raposo-do-Amaral et al⁸ report that congenital and acquired anatomic changes that affect the anatomy of the face can cause cognitive and psychological sequelae.

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In this work, we present a critical review of the criteria for aesthetic evaluation of the nasolabial region in cleft patients by professional evaluation and patient satisfaction.

Methods

We performed a literature search in the databases PubMed/MEDLINE, EMBASE, Biosis, SciSearch, and Scielo, covering the period from 1991 to 2012, for original studies using the terms “cleft lip and palate,” “facial deformity assessment,” “facial aesthetic conditions,” and “photographic assessment.” These terms were chosen to facilitate a critical review of the literature on evaluation criteria for aesthetic aspects in children and adolescents with cleft lip and palate. Summaries, reviews, and incomplete articles were excluded. Each item was individually evaluated to confirm that it was a primary source on the topic. Studies including patients with a diagnosis of cleft lip and palate who were treated surgically and evaluated by photographic parameters were included in this review.

Aesthetic evaluation

The concept of beauty involves a subjective and varied appraisal, depending on the particular view of the observer and ethnic, cultural, and age standards, and is therefore difficult to measure. However, we can say that, on balance, beauty is understood and judged as a whole. The well-established rule for aesthetics is that each third of the face – frontal, nasal, and oral – should be equal and unchanged. There must be a parallelism between the various planes (pupillary distances, labial commissures, and tragus and gonion distances), which are perpendicular to the median sagittal plane that divides the face into two almost symmetrical hemifaces (right and left).¹² The aesthetic assessment of professionals (eg, plastic surgeons, orthodontists, and maxillofacial surgeons) in this field is an even greater challenge in special cases, such as patients with cleft lip and palate.

Photography has been an important tool in the diagnosis of skeletal discrepancies, and in aesthetic appreciation and the study of facial harmony. Photographic facial analysis assists in assessing the balance of craniofacial structures, such as muscles and fat tissue, that may mask existing skeletal disharmonies.¹¹ Photographs, especially digital images, are already used in the medical field, for documentation, diagnosis, and planning, and are now also fundamental tools in the evaluation of therapeutic results.^{13–15}

Evaluation criteria

Asher-McDade et al¹⁶ developed a method for rating nasolabial appearance in patients with cleft lip and palate, which

became very popular. This classification scheme can be used in combination with both conventional cephalometric analysis and evaluation of the dental arch. The method was designed to reduce the influence of the surrounding facial nasolabial area, since it has been shown that judges are influenced by the overall attractiveness of the face. An ordinal scale was established based on evaluation by six judges (all orthodontists familiar with the conditions of the cleft) who analyzed the appearance of the nasolabial region of 75 patients by means of frontal and profile photographs. All patients had undergone repair of complete unilateral clefts. From this initial study, a classification system was established wherein 1 indicates excellent appearance and 7 indicates very poor appearance. Establishment of this scale facilitated evaluation of the appearance of the nasolabial region using a numerical scale. A second evaluation was conducted with five judges who evaluated four separate factors related to the nose and lips of 32 patients who had undergone complete unilateral cleft repair. The nasolabial profile, nasal shape and symmetry, and the vermilion border were rated on a 5-point scale (since a 7-point index is rarely used by evaluators, the scale was reduced to 5 points). Acceptable levels of reliability and reproducibility were obtained. This standardized grading system can be used to distinguish results from different therapeutic treatment centers.

Asher-McDade et al¹⁷ evaluated the nasolabial appearance of children with unilateral cleft lip and palate from six European centers. The authors studied color photographs of the front and profile of the nasolabial regions, masked to reduce the influence of appearance as a whole, and to obscure the origin of the patient. They assessed four components of the nasolabial region which had been scored by six members of the European Research Group, all of whom were orthodontists experienced and familiar with the treatment of cleft patients. The judges used the following 5-point scale: 1 = very good appearance, 2 = good, 3 = regular appearance, 4 = poor, and 5 = very poor. Scoring of nasal deviation was significantly different between centers, with center C showing the worst score (3.0) compared with centers A and E (2.3 and 2.2, respectively). The remaining components showed score variations between centers that were not statistically significant. By analyzing the components, no longer individually but as a whole, a significant difference was revealed between centers B and E compared with center C, with B and E exhibiting better scores. The limitations of image-based analysis are well known, but these scoring systems allow satisfactory assessment.

Becker et al¹³ conducted a study that aimed to compare clinical examination and morphometry from digital photographs, in the evaluation of repaired cleft lip. They used a predetermined protocol comprising 20 variables describing the nasolabial appearance, both for the clinical examination and for assessment of images acquired from a digital camera. Analysis of correlation between the two methods showed high consistency between the approaches. For metric variables, the average correlation coefficient was 0.73, and when all measures were combined, the coefficient reached 0.98. Variables related to muscle dynamics could not be judged by photographs, for obvious reasons. However, image scans enabled more precise detection of the minimum angular nose, and measurement of the areas of the nostrils. The authors concluded that morphometry of digital photographs is a valuable clinical tool for evaluation of patients with cleft lip and palate.

Johnson and Sandy⁹ developed an index to evaluate the aesthetics of surgical repair of bilateral cleft lip and palate. The study included evaluation of photographs of 50 patients by seven cleft orthodontists at two different times. Appearance was evaluated by assessing symmetry of the upper lip, scars, and continuity of the lip vermilion on a frontal view. Nose appearance was evaluated by assessing symmetry of the nostrils, the alar base, and the centralization of the columella. After determination of the level of agreement, a 5-point scale was established for clinical use. The authors subsequently conducted a field study in Australia with two trained examiners, evaluating patients directly, and indirectly with photographs taken at the same time. The direct and indirect assessments were compared, and agreement was moderate to good with no significant systematic bias. The authors concluded that the reproducibility of this index is comparable with that of other aesthetic indices, and is not affected by patient age. The index offers flexibility by allowing both direct and indirect assessment. The study did not specify whether plastic surgeons and orthodontists use the index in a similar way; further studies, using a joint committee of evaluators, might be interesting in this regard.

Kim et al⁷ proposed a tool for quantitative evaluation of the nasolabial deformity that compared four parameters established from images of cleft patients: the angular difference between the two nostrils; the center of each nostril and the distance between the two centers; the overlapping area of the nostrils; and the ratio of the overlap of the two nostrils. The analysis was standardized by three plastic surgeons, at three different times. The method was found to be reliable, and allowed for ease of use in determining the degree of deformity.

Nollet et al¹⁰ conducted a study to evaluate nasal appearance in patients with unilateral cleft lip and palate treated in The Netherlands, and to compare these individually, with the results of the six-center Eurocleft study. Relationships between nasolabial aesthetics, dental occlusion, and slit width were investigated in 42 patients in their center. Nasolabial appearance was evaluated by applying an aesthetic index and then subsequently compared with the findings of the six Eurocleft centers. The slit width was measured at birth using a plaster model of the maxillary arch. The authors found that 90% of patients presented scores from 2.0 to 3.7 for all aesthetic relations. Results in their patients were similar to those at centers A, D, E and F; were significantly better than results for patients at center C; and significantly worse than at center B. No correlation between aesthetics, dental occlusion, and width of the slit was noted. The authors concluded that the treatment protocol could not explain the differences in results.

Fudalej et al¹⁸ compared nasolabial aesthetics after different treatment protocols. They used four observers to assess four components of nasolabial appearance (shape, nasal deviation, nasal mucocutaneous junction, and profile) in 60 children who underwent surgical repair of the lip in one stage, and in 48 children who underwent lip repair in three stages (with group mean ages of 10.8 years and 8.9 years, respectively). The researchers used the aesthetic index developed by Asher-McDade et al,¹⁶ in which a low score indicates better aesthetics. The shape of the nose was judged aesthetically less favorable in both groups (scores of 3.1 and 3.2, respectively). The nasal deviation, the mucocutaneous junction, and the profile were scored in the range of 2.1 to 2.3 for both groups. Because there was no difference between the results of the two protocols, the authors concluded that nasal appearance after one- or three-stage repair is similar. Moreover, lip repair technique did not seem to affect the aesthetics of the nasolabial area.

He et al¹⁹ developed a method for assessing nasal appearance after lip repair, in patients with unilateral cleft lip and palate. The study included children with complete and incomplete clefts, with a mean age of 13.75 and 12.6 years, respectively. All patients underwent primary repair without nasal correction in several hospitals before 1 year of age. The authors used frontal, submental, and profile photographs for evaluation by seven examiners who rated appearance from very good to very poor. The reliability and reproducibility of the method were acceptable.

Kuijpers-Jagtman et al²⁰ sought to identify photographic illustrations related to the rating scale of the nasolabial region

Table 1 Distribution of studies on the aesthetic evaluation of the nasolabial region of children and adolescents with cleft lip and palate

Study	Number of patients	Number of parameters evaluated	Number of raters	Criteria	Age of patient	Opinion of the patient
Asher-McDade et al ¹⁶	75	4	6 (orthodontists familiar with cleft conditions)	Photography	9–13 years	No
Asher-McDade et al ¹⁶	32	4	5	Photography	9–13 years	No
Becker et al ¹³	24	20	2 (one investigator performed clinical examinations and another evaluated the photographs)	Clinical examination and photography	29.3 years	No
Johnson and Sandy ⁹	50	6	7 orthodontists	Photography	5–12 years	No
Kim et al ¹⁷	86	4	6 (3 plastic surgeons and 3 laypersons)	Photography	Not available	No
Nollet et al ¹⁰	42 (14 duplicated cases were randomly ordered among the 42 patients)	4	4	Photography	7.9–10.3 years	No
Fudalej et al ¹⁸	108 (60 underwent one-stage repair of all cleft structures and 48 underwent three-stage repair)	4	4	Photography	10.8 years (group of one-stage repair) and 8.9 years (group of three-stage repair)	No
He et al ¹⁹	45 (26 completed UCLP patients and 19 incomplete UCLP patients)	5	7	Photography	13.75 years (26 complete UCLP patients) and 12.26 years (19 incomplete UCLP patients)	No
Kuijpers-Jagtman et al ²⁰	42	4	4	Photography	9 years old	No
Raposo-do-Amaral ²¹	34	2	1	Photography and software program Adobe Photoshop® CS4 Extended	3 months–25 years old	No
Fudalej et al ²⁴	60	5	5 (1 rater analyzed the Nasolabial symmetry and 4 orthodontists evaluated nasolabial aesthetics)	Photography and the software program CorelDraw® v.11	11.2 years	No
Feragen et al ²⁵	268	4	Patients and their parents	The Child Experience Questionnaire	10 years	Yes
Mani et al ²⁸	83	5	3 professionals (2 medical doctors and 1 orthodontist) and patients	Photography and appearance questionnaire	20–47 years	Yes
Schwenzer-Zimmerer et al ²⁶	11	6	Evaluation by software	3D laser scanner and the software Polygon Editing Tool® 2.0	13.8 years	No
Tziavaras et al ²⁷	29	13 osseous landmarks	Evaluation by software	3D-CT and the software Persona	3.8 years	No

Abbreviations: UCLP, unilateral cleft lip and palate; CT, computed tomography.

proposed by Asher-McDade, in order to facilitate its use. Four observers evaluated the appearance of nasolabial photographs, in frontal and profile views, of 42 children (aged 9 years) with unilateral cleft lip and palate. Subsequently, they selected photographs that showed the highest degree of agreement between observers. For each of four components (nasal form, nasal deviation, profile shape of the nose, and redness), five photographs were selected to illustrate scores on the 5-point scale, resulting in the selection of 20 pictures. The authors concluded that nasolabial appearance can be reliably assessed using average scores of a panel of judges. The reference photographs developed in this study may facilitate the classification process.

Raposo-do-Amaral²¹ assessed the percentage of asymmetry of the lips and nose of patients with complete unilateral cleft lip palate who underwent incomplete primary lip repair using the technique of Mohler²² and Cutting and Dayan²³ (associated with modifications to the nasal region). Another objective was to compare the percentage of lip asymmetry pre- and postoperatively. They obtained measurements of area and distance between anatomical elements identified in photographs. The results showed a mean preoperative lip asymmetry index of approximately 43.16%, decreasing to 4.04% postoperatively (this difference was statistically significant). The average nasal asymmetry on postoperative follow-up was 19.5%, ranging from 4% to 45%. The authors concluded that the primary lip repair technique is effective for the correction of asymmetries inherent in the lip and nasal deformity of cleft patients.

Fudalej et al²⁴ evaluated the association between symmetry and nasolabial aesthetics in children with unilateral complete cleft lip and palate. These authors studied basal and frontal photographs of children with unilateral clefts and of a control group of unaffected children (with a mean age of 11 years and 2 months). By means of anthropometric measures and a coefficient of asymmetry, a single examiner established the nasolabial symmetry. The authors found a statistically significant difference between the cleft group and the control group. The authors described study limitations that may have affected the outcome, including the use of a handheld camera (implying that rotation of the head can influence symmetry), a very wide age range, and use of a single examiner. They concluded that symmetry and nasolabial aesthetics seem to exhibit a weak association in patients with unilateral cleft lip and palate.

Aesthetics and psychosocial impact

Feragen et al²⁵ conducted a study involving 268 children (aged 10 years) divided into five groups based on treatment type.

They evaluated psychosocial condition, and cognitive and emotional functions through a questionnaire and a clinical interview involving the children and their parents. Appearance was evaluated using a 10-point scale reflecting the degree of satisfaction with the cleft side, and overall appearance and visibility of the cleft. The different types and subtypes were a difficulty encountered in this study. The results indicated that contrary to expectations, the visibility of the cleft had no association with psychosocial condition. This may have been due to the age of the children in the study. Nevertheless, the cleft proved to be a negative factor in children with emotional difficulties and low self-acceptance.

Evaluation using three-dimensional imaging

Schwenzer-Zimmerer et al²⁶ prospectively evaluated the clinical application of three dimensional (3D) imaging for morphological analysis, planning, and preoperative assessment of symmetry. They also used this 3D imaging procedure to compare the postoperative features of treated patients with those of a control group that included nonfissured individuals. The authors of this study selected eleven patients (three females and eight males; mean age 13.8 years) with nonoperated unilateral cleft lip, unilateral cleft alveolus and lip, complete cleft lip, or complete cleft alveolus. The control group included 25 individuals without fissures (8 children and 17 adults). Preoperative and postoperative facial profile data were obtained using a 3D laser scanner for rapid measurement (VI 910®; Konica Minolta Holdings Inc, Tokyo, Japan). The photographs (Nikon D 70 70®; Nikon, Tokyo, Japan) and videos (Camcorder DCR-HC17E Handycam®; Sony, Tokyo, Japan) were taken using a standardized procedure, and these data were obtained for all patients. The 3D anthropometric data of the control group were collected, and these data were treated as the gold standard of symmetry in this ethnic group. Results were analyzed using the Polygon Editing Tool® software (v2.0; Konica Minolta Holdings Inc, Tokyo, Japan). Schwenzer-Zimmerer et al²⁶ evaluated six clinically relevant characteristics: length of the lip, crack width/mouth width, length of the columella, width of the nostrils, pouting effect, and shape of the filter. Postoperative 3D images of the cleft group showed symmetrical values within the range of the normal cohort. The 3D image facilitated a deeper understanding of the complex morphology to be treated, and could have contributed to the customization of surgical procedures.

Tziavaras et al²⁷ used 3D computed tomography to compare the craniofacial morphology of nonoperated

children with cleft palate with a control group of unaffected children. The study included 29 randomly selected ethnic Malay individuals with cleft palate (12 females and 17 males): ten with unilateral cleft palate, five with bilateral cleft palate, seven with primary cleft lip and palate, and seven with isolated cleft palate. The control group included 12 patients (four females and eight males) with no craniofacial abnormalities. The mean age in the cleft group was 3.8 years (range, 1.1–12.2 years), and in the control group was 4.8 years (range, 0.4–11.9 years). The software developed by Persona Australian Craniofacial Unit software (ACFU) (Adelaide Women's and Children's Hospital, Adelaide, Australia) was used for the 3D reconstruction of craniofacial imaging and determination of the coordinates of bony landmarks on the computer graphic display. The amplitudes of the midface and the dimensions of the nasal bones showed some differences between the cleft group and the control group. Nasal bone dimensions and facial width were larger in the unilateral cleft lip palate group than in the control group. Researchers concluded that a cleft palate affects the size and orientation of the nasal bone, and is associated with changes in the morphology of other facial bones located at some distance from the cleft.

Professional versus nonprofessional evaluation

Mani et al²⁸ evaluated the relationship between classification by professionals and lay workers, and patient satisfaction with nasolabial appearance in adults with bilateral cleft lip and palate. They formed two groups: a study group of 83 patients, and 65 control subjects who were evaluated in the same way. Assessments were done by committees of professionals and lay people, and involved classification from photographs, using a 5-point scale for four factors related to nasolabial appearance; and a survey of patients' satisfaction with their appearance. There was no significant correlation between professional and lay evaluation of nasolabial appearance in patients or control subjects. Lay judgment and the evaluation of patients and controls showed a low correlation, as did that of professionals and patients and controls. The authors suggested that the judgment of nasolabial appearance differs among professionals, lay people, and patients, and that this should be considered in the decision to perform surgical refinishing of unsightly cleft signs. In another study²⁹ about smile and degree of satisfaction in individuals with cleft lip and palate, frontal facial photographs were evaluated in 135 patients at rest and in natural and forced smile. Here,

results showed 84.4% of individuals considered their smile as aesthetically pleasant.

Perspectives

In recent years, systems with high-definition cameras and high-speed microprocessors capable of handling large data streams have become more readily available. However, the cost of creating a system with 3D imaging is likely to be prohibitive for the diffusion of these technologies in the postoperative evaluation of fissured patients.³⁰ Different types of 3D technologies can be applied for a more accurate postoperative assessment of the aesthetic aspects of cleft patients, including 3D cephalometry,³¹ morph analysis, moire topography,^{32,33} 3D magnetic resonance imaging, 3D ultrasonography, laser scanning,³⁴ digital stereophotogrammetry, and 3D videography.³⁵ However, studies need to be conducted with proven cost-effective methods to confirm the applicability of these technologies.

Conclusion

Facial deformities generated by cleft lip and palate are crucial in the socialization and quality of life of these patients. The aesthetic evaluation of results of surgical procedures in these patients represents a major challenge to the care team. However, several groups have sought to establish some standard measures that can facilitate the evaluation of results, and systematization of the literature.

Disclosure

The authors report no conflicts of interest in this work.

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