OPEN

ORIGINAL ARTICLE
Pediatric/Craniofacial

Photographic Measurements Partially Correlate to Nasal Function and Appearance among Adult Cleft Patients

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Background: Unilateral cleft lip and palate (UCLP) affects nasal function and appearance. There is a lack of objective measurements to evaluate these features. This study analyzes whether objective measurements on photographs correlate with nasal function and/or appearance among adults treated for UCLP.
Methods: All patients with UCLP born from 1960 to 1987 treated at the Uppsala University Hospital were invited (n = 109). Participation rate was 68% (n = 74); mean follow-up was 35 years. An age-matched control group (n = 61) underwent the same tests. Nostril area, nasal tip deviation angle, and width of the nostril were measured on photographs and were compared with functional tests and with appearance as assessed by self-assessment questionnaire, professional panel, or laymen panel.
Results: The photographically measured nostril area correlated with nasal volume (acoustic rhinometry) among UCLP patients, both cleft side and noncleft side, and controls (0.331, $P = 0.005$; 0.338, $P = 0.004$; and 0.420, $P < 0.001$, respectively). For the patients' noncleft side and controls, the area correlated inversely with airflow resistance at inspiration (noncleft side: −0.245, $P = 0.043$; controls: −0.226, $P = 0.013$). Laymen assessment of nasal appearance correlated with width ratio of the patients (0.27, $P = 0.022$) and with nasal tip deviation angle and area ratio of the controls (0.26, $P = 0.041$, and 0.31, $P = 0.015$, respectively).
Conclusions: Photographic measurements correlate partially with both functional tests of the nose and panel ratings of appearance. No correlation was found with self-assessment of appearance. Evaluation of photographs needs to be combined with patient-reported outcome measures to be a valuable endpoint of nasal appearance. *(Plast Reconstr Surg Glob Open 2016;4:e720; doi: 10.1097/GOX.0000000000000728; Published online 26 May 2016.)*

Unilateral cleft lip and palate (UCLP) is a complex facial malformation that may affect nasal function and aesthetics. Adults treated for UCLP experience problems related to form and function to a varying extent. Many patients request secondary surgical treatment, and several studies have indicated that the greatest concern for the patient is correction of appearance and function of the nose.[1–5] Earlier studies on nasal airway function among patients treated for UCLP indicate that objectively measured nasal function is extensively impaired; the patients have lower values regarding

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Received for publication August 1, 2015; accepted March 23, 2016.

DOI: 10.1097/GOX.0000000000000728

Disclosure: *The study was funded by Uppsala University and Uppsala University Hospital research funds. The authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the authors based on funding from the Thureus' Foundation.*

nasal volume, higher resistance to breathing, and lower results regarding olfaction compared with controls.[6,7]

Assessment of appearance is multifactorial and subjective. In the decision-making process for secondary surgery, it is important to understand how cleft stigmata are judged differently depending on the perspective of the observer. For example, self-assessment of nasal appearance among patients with clefts does not correlate with laymen nor professional ratings.[8] Patients rate their own appearance significantly lower than experts.[8,9]

There is still no widely accepted standard rating method to assess facial aesthetics in cleft lip and palate.[10] Nearly all cleft surgery outcome measures contain a subjective component.[11] To standardize evaluation of treatment outcome, Asher-McDade et al[12] elaborated a method of rating nasolabial appearance, developing a numerical scoring system using cropped photographs so that only the nose and upper lip were shown and assessed. This method has been used to assess outcome of cleft treatment in earlier studies, such as, the Eurocleft study[1] and the Clinical Standards Advisory Group study.[13]

It has earlier been discussed that nasolabial appearance should be evaluated in accordance with other outcomes of cleft care, including satisfaction with treatment, psychosocial adjustment, and quality of life.[10] The present study aims to investigate whether objective measurements on photographs (nostril area, nasal tip deviation angle, and nostril width ratio) correlate with nasal airway function among patients treated for UCLP or with satisfaction with appearance rated by patients, laymen, and professionals, to find a simple, noninvasive way of objective evaluation of form and function of the nose.

MATERIALS AND METHODS

Subjects

Uppsala University (UU) Hospital, Uppsala, Sweden, covers tertiary health care services for a population of approximately 1,500,000 people, with no private or alternatively funded hospitals treating patients with cleft lip and palate. All consecutive, nonsyndromic patients with complete UCLP (excluding patients with the Simonart band or partial clefts) in this population, born from 1960 to 1987, were considered for the study (n = 128). Of these, 19 individuals were not included on account of the following reasons: death (n = 6); major mental or physical incapacity (n = 5); living abroad (n = 5), or not found in the Swedish national population registry (n = 3). Of the 109 patients who were invited, through an information letter and a follow-up telephone call, 83 patients (76%) answered the questionnaires and performed the functional and clinical analyses. The nonparticipating group did not differ from the participating group regarding age and sex. The reasons given for nonparticipation were long travel distance (n = 11), lack of time (n = 7), and not wanting to be reminded of the treatment period (n = 3). Five patients did not specify a reason. The mean follow-up time from the first operation (ie, lip closure at 3 months of age) to the participation date in the current study was 35 years (20–47 years). Nine patients had missing photographs; thus, in total data from 74 patients were included in the current study (68%). Fifty-eight percent of the patients were men (n = 43) and 42% were women (n = 31). An age- and sex-matched control group (n = 61) underwent the same examinations, answered the same questionnaires, and was photographed according to the same protocol as the patients. Forty-one percent of the controls were men (n = 25) and 59% were women (n = 36). Mean age was 32 years (20–53 years).

Functional Tests and Examinations

The nasal patency and function were analyzed with a series of examinations. The tests were performed on all patients and controls, during an outpatient visit especially arranged for the study, at the Department of Otorhinolaryngology, UU Hospital.

Acoustic rhinometry (AR) was used to measure the volume of each nasal passage separately by sonographic signaling into each nostril. Rhinomanometry (RM) was used to evaluate nasal function by measuring airflow and pressure during nasal respiration. All measurements were performed before and after decongestion with 2 nasal puffs of 0.5 mg/mL oxymetazoline on each side. The tests were performed out of the allergy season, that is, if the patient had a common cold, the tests were postponed 4 weeks. In the control group, all measurements were performed bilaterally. No side differences were detected with AR or RM, and thus, the mean of the right and left side was calculated for the controls and used for the analysis. All tests have been described in more detail in earlier studies by the authors.[7]

Evaluation of Nasal Appearance by Panels and Photographic Measurements

The photographs used in the present study were all taken by a professional photographer at the UU Hospital under standardized and reproducible conditions. A yardstick with a color palette was used to allow for color and size calibration. The yardstick was placed in a holder at the level of the base of the nose (Fig. 1). The frontal and profile photographs were

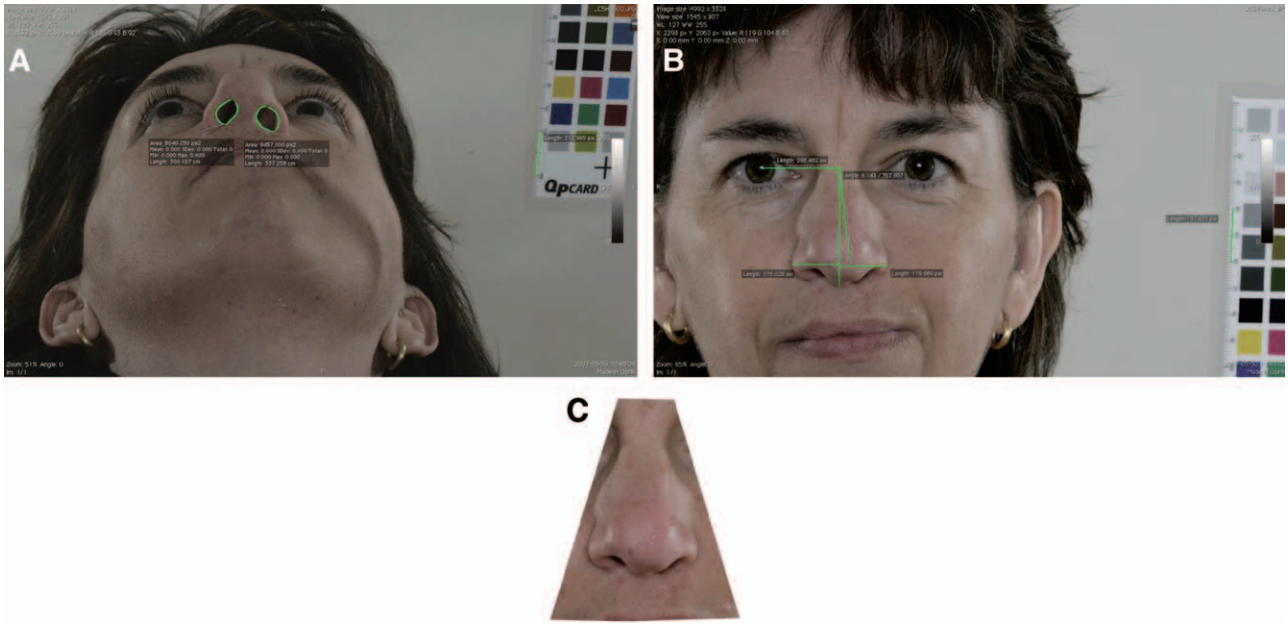

Fig. 1. A, Photograph of subject from snake view with the measures marked in the picture and a yardstick with color palette. B, Photograph of subject from frontal view with the measurements marked and a yardstick with color palette. C, Photograph cropped according to Asher-McDade.

then cropped and rated according to Asher-McDade et al.[12] The mean of the 4 parameters was calculated. Ratings were performed by a laymen panel and a professional panel. The professional panel consisted of 2 medical doctors and 1 orthodontist, none of whom had been involved in the treatment of the included patients nor in the current study, but with knowledge and experience of cleft care. The laymen panel consisted of 3 acquaintances of the authors with no medical experience and with no connection to the present study. The procedure and specific data outcome from the panel judgments have been described earlier.[7] All data files of the patients were encoded and blinded.

Three measurements were analyzed for evaluation of asymmetry and nasal appearance on the photographs: nasal tip deviation angle, nostril width, and nostril area. The nasal tip deviation angle was measured at the frontal view photographs and was defined as the angle between the midpupil sagittal line and the tip of the nose. Nostril width was defined as the distance from the sagittal midline of the nose to the most distal part of the nostril on each side (Fig. 1). To evaluate asymmetry of the nose, the difference between the distances from the midsagittal line to the lateral part of the nostril of each side was calculated. To compensate for differences in nose size between subjects, the ratio of the differences between distances from midsagittal line to each side and sum of these distances was calculated (width ratio) (Table 1).

The area of the nostril was analyzed on the snake view photographs (Fig. 1). The OsiriX MD software (Pixmeo, Geneva, Switzerland) was used for all measurements of the digital photographs, and pixels were converted to square centimeter, according to the yardstick. Similar to the width of the nostril, a ratio of the differences between areas of the nostrils and the sum of the areas of the nostrils was used in the analysis (area ratio) (Table 1).

Self-Assessment of Nasal Appearance

For self-evaluation of appearance, the Satisfaction with Appearance (SWA) scale was used.[14] The SWA reflects satisfaction with cleft-related and non-cleft-related parts of the face, speech, and overall appearance and extra oral visibility of the cleft. The questions of the form were answered with markings on a visual analogue scale, from 0 to 10, where a score of 0 was a very high level of satisfaction and 10 was very low. In the current study, the question specifically asking for level of satisfaction with appearance of the nose was used for the analysis. The SWA questionnaire was sent to the patients and the controls to be answered at home.

For patients and controls, travel expenses and partial salary loss were reimbursed, to diminish dropouts due to financial reasons. None of the persons involved in the evaluation of the current results had been involved in the treatment of the patients. The study was approved by the Research Ethics Committee of Uppsala University Hospital (reference num-

Table 1. Definition of Width Ratio and Area Ratio

$$\text{Width ratio} = \frac{\text{Midsagittal line to right nostril (mm)} - \text{Midsagittal line to left nostril (mm)}}{\text{Midsagittal line to right nostril (mm)} + \text{Midsagittal line to left nostril (mm)}}$$

$$\text{Area ratio} = \frac{\text{Area of the right nostril (mm}^2\text{)} - \text{Area of the left nostril (mm}^2\text{)}}{\text{Area of the right nostril (mm}^2\text{)} + \text{Area of the left nostril (mm}^2\text{)}}$$

A negative result was multiplied with −1.

ber 2005:245), and informed consent was obtained from each subject participating in the study.

Statistical Analysis

Data were presented as median and interquartile range (IQR). The data were tested for normality with histograms. Nonparametric analyses were used because not all data were normally distributed. Correlations were tested with scatter plots and the Spearman rho rank correlation test (2-tailed). Wilcoxon signed-rank test and Mann-Whitney *U* test were performed to analyze differences between professional and laymen ratings and patient satisfaction questionnaire, for differences between patient and control self-assessments, and for analysis of differences in nostril area between patients and controls and between cleft- and noncleft sides. A *P* value of <0.05 was regarded as significant.

For the panel assessments, to reduce variability, the scores of the 3 observers of each panel were averaged for each nasolabial component and for the sum of the 4 subscores. The intrarater agreement of the panel scorings showed good to very good agreement among professionals and fair to good agreement among layman panel members as presented in earlier studies. Consequently, the mean scores of the 3 observers were used for comparison between panels, and when comparing the results of the panels to the SWA self-assessed scores.[8] Statistical evaluations were carried out with a computer software package (SPSS PC version 22.0; SPSS, Chicago, Ill).

RESULTS

Patients were found to have a larger nostril area as measured on the photographs both on the cleft and the noncleft side compared with controls: patients' cleft side median 105.8 mm² (IQR, 36.9 mm²), patients' noncleft side 85.7 (39.9), and controls 69.8 (28.0); $P < 0.001$ for both comparisons. Furthermore, the cleft side was significantly larger than the patients' noncleft side ($P < 0.001$). The nostril area for all subgroups (patients' cleft and noncleft side and controls) correlated with the anterior volume of the same nostril as measured by AR before and after decongestion (Table 2). The airflow resistance at inspiration measured with RM correlated inversely with the patients' noncleft side and with the controls, after decongestion, $P = 0.043$ and $P = 0.013$, respectively. No significant correlation could be found for the patients' cleft side and airflow resistance at inspiration (Table 2).

When comparing measurements from photographs representing asymmetry (nasal tip deviation angle, width ratio, and area ratio) between UCLP patients and controls, patients presented with a larger nasal tip deviation angle; median 4.2° (IQR, 4.0°) compared with controls; 2.5 (3.3) ($P = 0.001$). Similarly, the area ratio was larger for the patients, 0.98 (0.12), compared with controls 0.05 (0.07) ($P < 0.001$). No difference was found for width ratio between patients and controls (Table 3).

Analysis of correlation between measurements of nasal asymmetry on photographs and nasal appear-

Table 2. Correlation of Measurements From Photographs, Representing Asymmetry, With Functional Test Outcomes

	Photographic Variables					
	Cleft Patient Nostril Area; Cleft Side		Cleft Patient Nostril Area; Noncleft Side		Control Subject Nostril Area	
	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*
Functional variables						
Anterior volume before decongestion	0.33	**0.005**	0.34	**0.004**	0.42	**<0.001**
Anterior volume after decongestion	0.29	**0.015**	−0.03	**0.016**	0.37	**<0.001**
Airflow at inspiration before decongestion	0.03	0.818	−0.01	0.924	−0.20	**0.025**
Airflow at inspiration after decongestion	−0.64	0.603	−0.25	0.043	−0.23	**0.013**

*The Spearman rho test. Bold indicates a *P* value < 0.05.

Table 3. Comparison of Photographic Measurements for Asymmetry Between Patients and Controls

	Patients		Controls		
	Median	IQR	Median	IQR	*P**
Nasal tip deviation angle (°)	4.24	3.98	2.47	3.28	**0.001**
Width ratio	0.08	0.12	0.07	0.08	0.243
Area ratio	0.98	0.12	0.051	0.06	**<0.001**

*Mann -Whitney *U* test.
IQR, interquartile range. Bold indicates a *P* value < 0.05.

ance assessed by layman, professional, and patients showed correlation only with layman panel judgment (Table 4). The correlation was significant for patients' width ratio (0.27, $P = 0.022$) and for controls' nasal tip deviation angle (0.26, $P = 0.041$)) and area ratio (0.31, $P = 0.015$). No further correlation was found.

DISCUSSION

Photographs are widely used to assess outcome of cleft care. The current study hypothesized that measurements from digital photographs, representative of nasal asymmetry, can be used to analyze and compare nasal form and function between patients and over time. Data indicate that parameters measured on photographs are affected in a cleft population and correlate with some variables of nasal function.

A key end point of cleft surgery is reconstruction of nasal form and function. Assessment of end points is a continuous debate in cleft surgery—what aspects, at what time point, and how should they be evaluated? Objective measures are sought for as these can easily be compared between patients and over time. The evaluation method needs to be simple enough to allow application in different clinical settings. Two-dimensional photography can easily be performed and is a noninvasive method from which objective measurements can be made. The current study analyzes whether measurements from photographs can be used to assess these aspects, that is, changes over time or differences between different populations, and whether it could be used as an easily evaluated, indirect end point, for nasal function and satisfaction with appearance.

The treatment of clefts aims at normalization of facial appearance and improved quality of life, which has been shown to be associated with satisfaction of appearance.[15] Patients are less satisfied with their appearance than controls.[4,8,9] Professional opinions are not always the same as the patients' opinion of appearance and successful treatment; self-assessment and panel judgment of appearance differ.[8] A series of studies have presented that the majority of cleft patients express a wish for further surgical treatment, both aesthetically and functionally.[3,4,9]

In the current study, cropped photographs were evaluated by laymen and professional panels, to reduce rating based on background facial attractiveness rather than nasolabial appearance. This 4-featured assessment allows individual features of the nasolabial area to be assessed independently or added together.[1] Panel evaluations and cropped versus noncropped photographs have been extensively studied in cleft treatment, and the current method with cropped photographs has been thought to give the most adequate evaluation of nasal features after cleft treatment and has been internationally chosen as a validated way of evaluating cleft treatment outcome.[12] Still, it is known from earlier studies that photographs are limited in their 2-dimensional representation.[1,16] Yildirim et al[17] performed an analysis of morphed photographs to improve evaluation of photographs as an outcome tool, concluding that morphing of pictures is a suitable method for creation of standard cleft faces to eliminate other facial appearance aspects that may affect assessment outcome.

The measurements from photographs correlated with laymen panel judgment, but not with professional panel judgment or with self-assessment (Table 4). Laymen are nontrained judges and consequently do not look for surgically associated outcome nor do they have a personal history with cleft deformity, which may affect judgment of appearance. This may explain why laymen may have a more nontrained/nonaffected and thus nonbiased judgment and thus correlating with photographic measurements. Similarly, Vegter and Hage[18] found that patient satisfaction with appearance and wish to perform secondary corrective surgery of the nose did not correlate with anthropometric measurements. Appearance is multifactorial and needs to be evaluated in coherence with patients' experience.

The present study demonstrated that the nasal tip deviation angle was significantly larger among patients compared with the control group. The size of the nasal tip deviation angle showed no correlation with any self-assessment of appearance made by patients or controls. Similar results were found

Table 4. Correlation of Measurements from Photographs Representing Asymmetry with Panel and Self-assessment of Appearance

	Photographic Variables											
	Patients						Controls					
	Nasal Tip Deviation Angle		Width Ratio		Area Ratio		Nasal Tip Deviation Angle		Width Ratio		Area Ratio	
	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*	Correlation Coefficient*	*P*
Panels and self-assessment of nasal appearance												
Professional panel assessment	0.10	0.393	0.06	0.569	0.14	0.256	0.16	0.238	0.16	0.231	0.24	0.063
Layman panel assessment	0.17	0.159	0.27	**0.022**	0.06	0.618	0.26	**0.041**	0.21	0.115	0.31	**0.015**
Self-assessment	−0.00	0.980	0.02	0.856	0.11	0.373	−0.11	0.421	0.10	0.494	−0.17	0.214

*The Spearman rho test.

when comparing the ratio of the distance from the midsagittal line to the lateral nostrils with the panel ratings and the self-assessments. There was a correlation between layman ratings and the width ratio, whereas there was no correlation with the self-assessments. The results of the current study suggest that both professionals and laymen consider different measurable features in the face to be important factors for facial appearance, whereas satisfaction with appearance among patients is not directly associated with actual facial features. Vegter and Hage[18] concluded this in a very clear statement, “Patients do not seem to have an anthropometric interest in their appearance.”

People treated for UCLP demonstrate a wide range of nasal function impairments.[6,7,19,20] The volume and minimum cross-sectional area of the cleft side are smaller in people treated for UCLP compared with controls, when measured with RM.[7] In the current study, the nostril area was larger on the cleft side for UCLP patients than for controls. This is explained by differences in method of analysis; area of nostril as measured on photographs is measured at the level of the nasal opening, whereas the minimum cross-sectional area (as measured by RM) is the area at the level of narrowing in the anterior part of the nose. The anterior part is defined from the entrance to 2.2 cm into the nose.[21] The cleft deformity or the surgical treatment of the malformation can over time affect the nasal septum, the turbinates, and the cartilages leading to changes in size of the nasal conduits anywhere along this level.[2]

A potential drawback of the current study is the large age range among the study group (20–47 years). However, all the patients were full-grown adults and the degenerative process of the nose has theoretically not begun at the age of 47 years.[22] Furthermore, the control group was age-matched to limit any shortcomings due to age differences. The limited number of patients is always a challenge in long-term follow-up studies. On the contrary, no other study with a follow-up time of mean 35 years have been found in the field with a similarly large population or participation rate (n = 74; 68%). A further limitation of the current study is that different scales were used for self-assessment and panel judgment of nasolabial appearance, making comparisons of results more difficult. Therefore, nonparametric statistics based on ranking, and not numeric values, were used. The patients who chose not to participate in the study might have differed from the patients participating in the study. However, telephone communication with the nonparticipants showed that the reason for not participating mainly was due to time limits or long travel distance.

CONCLUSIONS

Photographic measurements correlate partially with both functional tests of the nose and panel ratings of appearance. Acknowledging the limitations stated in the current study, these measurements are not specific enough to suggest indication for treatment on an individual level; however, they may serve as a simple method to follow treatment over time. No correlation was found with self-assessment of appearance. Evaluation of photographs needs to be combined with patient-reported outcome measures to be a valuable end point of nasal appearance. It is essential to continue the development of more standardized outcome measurements in cleft care as this can ensure quality control and be helpful in comparison of results between centers and over time.

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PATIENT CONSENT

The patient provided written consent for the use of her image.

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