

Feeling Normal? Long-Term Follow-up of Patients with a Cleft Lip–Palate after Rhinoplasty with the Derriford Appearance Scale (DAS-59)

Andreas E. Albers, MD, PhD¹ Andreas C. Reichelt, MD¹ Gilbert J. Nolst-Trenité, MD, PhD²

Dirk Jan Menger, MD, PhD³

¹Department of Otorhinolaryngology, Head and Neck Surgery, Charité Universitätsmedizin Berlin, Berlin, Germany

²Department of Otorhinolaryngology, Facial Plastic Surgery, Jan van Goyen Kliniek, Amsterdam, The Netherlands

³Department of Ear, Nose, Throat – Facial Plastic Surgery, University Medical Center Utrecht, Utrecht, The Netherlands

Address for correspondence Andreas E. Albers, MD, PhD, Department of Otorhinolaryngology, Head and Neck Surgery, Charité Universitätsmedizin Berlin, Hindenburgdamm 30, Berlin 10117, Germany (e-mail: andreas.albers@charite.de).

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Abstract

The stigma of nasal deformity due to a congenital cleft lip–palate has an undeniable influence on the affected patient's life. It is therefore of interest to investigate if efforts to reduce esthetic and functional impairments by rhinoplasty (single or multiple) can result in an increased satisfaction with appearance and a self-perception similar to the noncleft population. Retrospective scoring before and after rhinoplasty using the validated Derriford Appearance Scale (DAS-59) and subsequent statistical evaluation and comparison to datasets available in the literature for further classification was used. Of the 61 patients who underwent at least one rhinoplasty, 26 responded to all questions. The mean age of responders was approximately 30 years of age and the male:female ratio was 1:1.2. The scale showed a significant overall improvement after surgery. The full scale and all subscale scores of the DAS-59 were significantly reduced after surgery demonstrating an improvement in the respective categories. Most importantly, if postoperative results were compared with a population concerned and unconcerned about appearance, no difference “facial self-consciousness” of appearance was apparent. Also postoperative subscores for “general self-consciousness” (GSC) and “social self-consciousness” of appearance (SSC) showed no difference from those obtained from the population concerned about appearance. The postoperative subscore for “sexual and bodily self-consciousness” of appearance (SBSC) indicated improvement beyond the level found in the concerned control population. Due to only a low improvement in the difference compared with the subscore representing a “negative self-concept,” a statistically significant difference to the concerned population remained, possibly indicating that therapy beyond surgery is needed for improvement. After rhinoplasty, the investigated group of cleft lip–palate patients with nasal deformities showed an improvement in their self-conceived appearance as measured by the DAS-59. Their assessment of self-appearance was comparable to that of a group of noncleft persons with concern about their appearance. Taken together, rhinoplasties, primary and revision, add to the psychosocial well-being and an improved self-perception enhancing quality of life and enabling a more normal life. Further research is needed to clarify how the low reduction found in the “negative self-concept” may be addressed successfully.

Keywords

- revision rhinoplasty
- outcome evaluation
- cleft lip
- cleft palate
- self-perception

The congenital cleft lip with or without cleft palate belongs to one of the most common and important congenital malformations resulting in an anomaly of the lip, nose, and face. Its incidence varies according to geographical and ethical belonging. In Europe, between 1/500 and 1/700 babies are born with clefts of the lip or palate, or both.^{1,2} The proportion of male patients increases, as lesion and defects are more severe. Functional impairments include nutritional problems, speech disorders, recurrent middle-ear infection, misalignment of teeth, and nasal breathing problems. In addition to functional impairments, the aesthetical stigma of nasal and lip deformity has an undeniable influence on the affected patient's life.³ It can lead to social retraction and exclusion as well as primary and consecutive psychological problems.^{3,4}

Effective treatment is important and one key is surgery aiming to reconstruct an appearance as close to normal as possible.² The surgery should include functional improvements as well to support a normal self-perception.^{5,6} Examples for this are a reconstruction of the palate to enable normal eating and drinking, placement of middle-ear drainage tubes to improve hearing and to support normal speech development, and correction of nasal breathing problems by functional septo-rhinoplasty.

To evaluate the outcome, objective measures such as anatomic measurements and clinical photographs are used.^{7–9} Although important, these methods are not sufficient if used without any further references. Patient-reported outcome measures that include aesthetic results, speech, functionality, self-image, incorporation into society, and quality of life (QOL) provide surgeons with a more comprehensive assessment of surgical outcomes.¹⁰ The main consequences of a cleft lip and/or palate are facial and functional impairments that seem to be intimately related to difficulties in psychosocial functioning.⁴

It is of high interest to investigate if efforts to reduce aesthetic and functional impairments by rhinoplasty (single or multiple) can result in a reduction in interpersonal problems and an increased satisfaction with appearance and QOL. For measuring health-related QOL, a number of measures are available. Only a few of them are directed specifically toward cosmetic surgery to assess concerns about physical appearance and that are sensitive to the outcome of appearance-altering effects of surgical treatment.

The Derriford scales have been designed to meet this need and are extensively studied, demonstrating excellent validity, reliability, and internal consistency in both clinical and general populations.¹¹ There are two versions of the scale: the Derriford Appearance Scale 24 (DAS-24), a short form composed of 24 items, and the Derriford Appearance Scale 59 (DAS-59), a longer form containing 59 items.^{12,13} Both scales are designed to generate a comprehensive assessment of the disruption represented by the score obtained through the scales. A higher score is associated with a higher degree of image-related distress and dysfunction. The DAS-59 thus generates a full-scale score and five factorial subscale scores of which three are not feature specific (general self-consciousness of appearance [GSC], social self-consciousness of appearance [SSC], and negative self-concept [NSC]) and two

are feature specific (sexual and bodily self-consciousness of appearance [SBSC] and facial self-consciousness of appearance [FSC]).^{11,12}

The DAS-24 generates a single score without a factorial structure and its detail, but still maintains a strong relationship with the original factorial scale.¹³ As an acceptable short form of the original DAS-59, it also is widely applicable, psychometrically robust, and discriminates well between patient groups.¹³ Both scales meet the need for an objective measure of the spectrum of psychological distress and dysfunction that is characteristic of disfigurements, deformities, and aesthetic problems of appearance. The scores are highly sensitive as a measure of change following surgical treatment with large and significant preoperative–postoperative reductions in full scale and, for the DAS-59, the factorial scores of patients treated for facial features or bodily/sexual features.^{12,13} Furthermore, a patient's score can be compared with normative data available for a clinical and/or general populations.¹² Previous studies showed an increase of patient satisfaction after secondary rhinoplasty in cleft lip patients using the validated Rhinoplasty Outcome Evaluation (ROE) questionnaire.¹⁴ Results showed a significant improvement in both aesthetic appearance/self-perception and nasal breathing. Interestingly, the improvement in aesthetic appearance was judged more as successful.^{14,15} It was therefore of great concern to investigate the distress and dysfunction in relation to self-consciousness of appearance using DAS-59 and DAS-24 in patients with a cleft lip–palate before and after undergoing rhinoplasty.

Consequently, the objective of this study was to investigate if one or multiple rhinoplasties in patients with a cleft lip–palate result in an improved QOL as measured by the DAS-59 that ideally would be as close as possible to what a control group perceives as normal.

Materials and Methods

Patients

The surgical patient database of the Academisch Medisch Center (AMC), Amsterdam, the Netherlands, was searched for patients who were diagnosed with uni- or bilateral cleft lip–palate who had received one or several rhinoplasties between October 2004 and May 2011 with the aim to improve and at best to normalize facial appearance. Typically, prior to rhinoplasty, a closure of a cleft lip and palate had already been performed and was followed by one or several rhinoplasties according to a strategy proposed by us earlier.^{16,17} Sixty-one patients could be retrieved with complete contact information. Those patients who were not in active treatment or do not have any sort of dependency at the AMC were contacted via mail stating that participation in the study was completely voluntary and anonymous; the former surgeons were blinded for participation and responses and that data would only be used for the purpose of statistical analysis and subsequent publication in a scientific journal. They were asked to complete the DAS-59 questionnaire from the perspective before and after the last rhinoplasty. Nonrespondents were given reminders via telephone by a nurse. Of the 61 patients, 26

returned complete questionnaires (13 females and 13 males) with an average age of 31. The youngest patient was 12 years old and the oldest patient was 63 years old. The average time passed after the last surgery was 3.9 years with a minimum of 2 years and a maximum of 6 years after the surgery. Therefore in the view of the authors, in all cases sufficient time after surgery had passed to allow complete healing and to assume a concluded settling into the altered body image. Twenty-three of the respondents were of Caucasian origin, two were Dutch-Surinamese (a multi-ethnic state with African, Asian, and European influence), and one was of Asian background.

Statistical Analysis

Data management and statistical analysis were performed using Excel 2011 (Microsoft Corporation, Redmond, CA) and EpiCalc 2000 (version 1.02, freeware; Brixton Health, Brixton, UK).

The data were analyzed using within-group paired Student *t*-test. For comparison with data retrieved from the literature, mean values and standard deviation were statistically compared with data published by Carr et al¹¹ containing a sample from the general population (*n* = 1,001), the general population who are not concerned about appearance (*n* = 528), and who are concerned about appearance (473).

Results

Pre- and Postoperative DAS-59 Scores

Preoperative mean DAS-59 score of 98.2 was measured that decreased significantly to 70.1 postoperatively (►Table 1). Similarly, scores in all five subscales showed a significant decrease reflecting an improvement in the “general-, social, sexual, and bodily, as well as facial self-consciousness of appearance” and a reduction in “negative self-concept.”

The most prominent reduction in percentage was found in the categories of “self-consciousness of sexual and bodily appearance” (SBSC) (48%) and of “facial appearance” (52.5%).

Comparison of DAS-59 Scores of Patients with a Cleft to a Nonclinical Population

To put the acquired data into a wider perspective, we compared the scores determined for the patients with a cleft with previously published scores of a “nonclinical/

general population” divided into concerned and unconcerned about their appearance.¹¹

The purpose of this comparison was to investigate if differences between the populations had changed after surgery.

A comparison between the general population *unconcerned* about appearance and patients with a cleft preoperatively showed a highly significant difference (*p* < 0.001) in their distress level with full-scale DAS-59 scores of 29.2 and 98.2, respectively (►Table 2). Similarly, the comparison of the DAS-59 subscales resulted in all cases in highly significant differences.

Surgery resulted in patients with a cleft in a reduction in the full-scale and all subscale scores, compared with the general population *unconcerned* about appearance indicating lower distress level. However, only for the subscale of FSC, the reduction of the score was sufficiently high to diminish any significance.

The comparison of the general population *concerned* about appearance, displaying a higher level of distress, to patients with a cleft before surgery revealed a significant difference (DAS-59 score, 79.70 vs. 98.23; *p* < 0.05). Yet after the surgery, no significance was found for the full-scale score and the subscale scores of GSC, SCC, and FSC.

While for the subscale representing SBSC pre-operatively no difference in the score from the general population *concerned* about appearance was apparent, after surgery an improvement was observed that led to a significant difference (DAS-59 score, 11.45 vs. 5.04; *p* < 0.001).

Scores for the subscale of negative self-concept remained significantly different; however, a reduction in the postoperative score could be noted.

Discussion

Previous studies showed that secondary functional rhinoplasties in patients with a cleft resulted in improvement of nasal breathing and^{7,18} facial aesthetics^{7,19} and an increased postoperative satisfaction, self-esteem, and QOL.^{14,18} In a study of patients undergoing primary, secondary, functional, and cosmetic rhinoplasty, the level of distress was measured by using the DAS-24 scale before and after surgery. Although levels of increased distress could be determined before and early after surgery, later a reduction in distress matching baseline values became apparent.²⁰ Similarly, in a group of

Table 1 Comparison of pre- and postoperative DAS-59 scores

Factor	Score		
	Preoperative (SD)	Postoperative	Change
Full scale	98.23 (15.01)	70.08 (12.69)	28.15 ^a
Subscales			
1. General self-consciousness of appearance	37.62 (15.11)	29.15 (16.24)	8.46 ^a
2. Social self-consciousness of appearance	24.54 (13.63)	15.00 (11.22)	9.54 ^a
3. Self-consciousness of sexual and bodily appearance	10.04 (6.96)	5.04 (4.35)	5.00 ^a
4. Negative self-concept	17.65 (3.55)	15.65 (3.80)	2.00 ^a
5. Self-consciousness of facial appearance	3.96 (4.20)	1.88 (2.29)	2.08 ^a

^a*p* < 0.001, one tailed Student *t*-test.

Table 2 Preoperative to postoperative changes in mean DAS-59 full-scale

Patient group	n	Full scale (SD)	DAS-59 subscales				
			GSC (SD)	SSC (SD)	SBSC (SD)	NSC (SD)	FSC (SD)
General population unconcerned	528	29.20 (20.92)	5.57 (6.24)	6.41 (8.54)	2.92 (4.12)	10.02 (3.68)	1.25 (2.01)
Patients with a cleft preoperatively	26	98.23 (15.01)	37.62 (15.11)	24.54 (13.63)	10.04 (6.96)	17.65 (3.55)	3.96 (4.20)
Difference		p < 0.001	p < 0.001	p < 0.001	p < 0.001	p < 0.001	p < 0.001
General population unconcerned	528	29.20 (20.92)	5.57 (6.24)	6.41 (8.54)	2.92 (4.12)	10.02 (3.68)	1.25 (2.01)
Patients with a cleft postoperatively	26	70.08 (12.69)	29.15 (16.24)	15.00 (11.22)	5.04 (4.35)	15.65 (3.80)	1.88 (2.29)
Difference		p < 0.001	p < 0.001	p < 0.001	p < 0.05	p < 0.001	n.s.
General population concerned	473	79.70 (37.08)	29.10 (13.55)	17.81 (12.98)	11.45 (8.66)	13.57 (4.03)	2.65 (2.93)
Patients with a cleft preoperatively	26	98.23 (15.01)	37.62 (15.11)	24.54 (13.63)	10.04 (6.96)	17.65 (3.55)	3.96 (4.20)
Difference		p < 0.05	p < 0.05	p < 0.05	n.s.	p < 0.001	p < 0.05
General population concerned	473	79.70 (37.08)	29.10 (13.55)	17.81 (12.98)	11.45 (8.66)	13.57 (4.03)	2.65 (2.93)
Patients with a cleft postoperatively	26	70.08 (12.69)	29.15 (16.24)	15.00 (11.22)	5.04 (4.35)	15.65 (3.80)	1.88 (2.29)
Difference		n.s.	n.s.	n.s.	p < 0.001	p < 0.05	n.s.

Abbreviations: DAS, Derriford Appearance Scale; FSC, facial self-consciousness of appearance; GSC, general self-consciousness of appearance; n, number of patients included; NSC, negative self-concept; n.s., not significant; SD, standard deviation; SSC, social self-consciousness of appearance; SBSC, sexual and bodily self-consciousness of appearance.

^aData extracted from Carr et al¹¹ for comparison. Scores and factorial subscale scores of cleft patients were compared with scores of patients treated for abnormalities of facial features and of the general population.¹¹

patients prospectively enrolled in a study measuring the impact of cosmetic facial surgery including surgery for the aging face and rhinoplasty with the DAS-59, improvement in the postoperative QOL was found.²¹

In line with these results, we found for the DAS-59 full-scale score and the separate subscales a significant reduction in the scores after rhinoplasty in patients with a cleft indicating a reduction of distress in all categories. Along with this general improvement and confirming a specific effect of the operation, the most prominent reduction in the score was found for the FSC score. Thus, our results also confirm the DAS-59 feature-specific factorial structure.¹²

Beyond the comparison of perioperative results, our next aim was to put the study results into a broader perspective and to compare the results derived from a defined group of patients with a cleft requiring rhinoplasty with the general population. Such comparison would allow to evaluate how close the studied group of patients with a cleft achieved a feeling of "normality" following surgery as determined by the different scales of the DAS-59 and in comparison to two different groups of individuals. For this, we compared previously published results obtained from a large population of individuals who were either *concerned* or *unconcerned* about appearance with our pre- and postoperative results.¹¹ A significant difference between postoperative scores from patients with a cleft and the

nonclinical population *unconcerned* was still present indicating a generally increased psychological distress of the patients with a cleft. However, when looking at the subscales, the difference between the two groups for the FSC changed from significant to not significant postoperatively, again confirming also the feature-specific factorial structure of the DAS-59.

The comparison between the general population *concerned* about their appearance and the patients with a cleft showed even more pronounced results. No significant difference for the full-scale and the FSC, GSC, and SSC subscores was present postoperatively. In other words, the psychological distress of this clinical population was reduced postoperatively to a level found comparable in a nonclinical population. Moreover, a significantly lower level of distress was determined postoperatively by a reduction in the SBSC. These data indicate that even after years of stigmatization and psychological adaption to disfigurement, relatively small changes due to functional and cosmetic surgery can result in a significant reduction in distress and increase psychological well-being. However, surgery had no impact on the NSC subscale, possibly indicating that surgery alone is not enough to improve on the negative self-concept of these patients and that psychotherapy specifically targeting this issue would be helpful.

Taken together, our results demonstrate that from the patients' perspective, which is what ultimately counts, a clear improvement in QOL was achieved through primary, secondary, and sometimes multiple rhinoplasties. Moreover, we could demonstrate by comparing patients after rhinoplasty to a collective of patients concerned and unconcerned about appearance that the self-perception of patients with a cleft after surgery is more comparable to what may define as "normal" than before.

Conclusion

- The full scale and all subscale scores of the DAS-59 were significantly lower after surgery demonstrating an improvement in all categories.
- Postoperatively, no difference in FSC between the unconcerned population and concerned population was apparent.
- The postoperative subscores for FSC, SSC, and GSC showed no difference from those obtained from a population concerned about appearance.
- The postoperative subscore for SBSC indicated improvement beyond the level found in the concerned control population.
- Due to only a low improvement in the NSC subscore, a statistically significant difference to the concerned population remained, possibly indicating that therapy beyond surgery is needed.

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Guest Editor Comments

The authors should be congratulated for contributing to this important aspect of patient-centered outcomes research. The result of a rhinoplasty in a child with a cleft should be objectively assessed and graded, and technical pearls guided by this clinical outcomes research. On equal footing, but often neglected, is the perception of the patient, which this manuscript has made an attempt to gather with the Derriford scale.

The only major concern is that the authors were unable to give the patients a presurgical assessment prior to the procedure, instead relying on the patient to remember how they felt before the surgery. Future studies should include the quality-of-life assessment truly pre-procedural to limit recall bias.

— Travis T. Tollefson, MD, MPH, FACS