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Facial attractiveness of patients with unilateral cleft lip and palate and of controls assessed by laypersons and professionals

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SUMMARY

OBJECTIVES: The aim of the study was to identify differences in the aesthetic evaluation of profile and frontal photographs of (1) patients treated for complete left-sided cleft lip and palate and (2) control patients by laypeople and professionals.

MATERIALS, SUBJECTS, AND METHODS: Left-side profile and frontal photographs of 20 adult patients treated for complete left-sided cleft lip and palate (10 men, 10 women, mean age: 20.5 years) and of 10 control patients with a class I occlusion (five men, five women, mean age: 22.1 years) were included in the study. The post-treatment photographs were evaluated by 15 adult laypeople, 14 orthodontists, and 10 maxillofacial surgeons. Each photograph was judged on a modified visual analogue scale (VAS, 0–10; 0 'very unattractive' to 10 'very attractive'). A four-level mixed model was fitted in which the VAS score was the dependent variable; cases, profession, view, and rater were independent variables.

RESULTS: Compared with laypersons, orthodontists gave higher VAS scores (+0.69, 95% confidence interval (CI) [0.53, 0.84]; $P < 0.001$), followed by surgeons (+0.21, 95% CI [0.03, 0.38], $P = 0.02$). Controls were given significantly higher scores than patients with clefts for profile and frontal photographs (+1.97, 95% CI [1.60; 2.35], $P < 0.001$). No significant difference was found between the scores for the frontal and lateral views ($P = 0.46$).

CONCLUSIONS: All the different rater panels were less satisfied with the facial aesthetics of patients with clefts compared with that of control patients. Further research should evaluate whether these findings correlate with patients' self-perception and to what extent it affects the patients' psychosocial well-being.

Introduction

Cleft lip and palate comprise one of the most common birth defects occurring in one of every 500–1000 live births worldwide (Murray, 1995). Highly specialized treatment is necessary from the early periods of life until adulthood to improve function, facial appearance, and psychosocial development. Although surgical closure of the cleft, orthodontic treatment, and orthognathic surgery improve facial symmetry, the treatment often does not result in an average facial appearance (Pruzinsky, 1992). Scars formed after surgical interventions and an asymmetry in the nose and mouth region are left behind. This impaired facial appearance can also affect the patients' psychosocial well-being (Berk *et al.*, 2001).

Facial appearance is an important measure of success in treatment of patients with cleft lip and palate. Treated patients with clefts have been compared with controls without clefts and have been rated as less attractive (Meyer-Marcotty *et al.* 2010, 2011b). Although the association between nasolabial asymmetry and aesthetic evaluation was found to be weak in children (Fudalej *et al.* 2012), increased nasolabial asymmetry in adults with clefts (Meyer-Marcotty *et al.* 2010, 2011b) might have contributed to these findings. However, in these studies, facial aesthetics had been rated exclusively by laypeople. It has been described in the literature, that raters with different backgrounds differ in their evaluation of facial aesthetics in patients with clefts: professionals tended to rate patients with clefts better (Marcusson *et al.* 2002; Sinko *et al.* 2005) or worse (Gkantidis *et al.* 2012) than did the patients themselves and better (Gkantidis *et al.* 2012) or worse (Foo *et al.* 2013) than laypeople. The patients themselves and their parents were more satisfied with the facial appearance than laypeople (Gkantidis *et al.* 2012). The inconsistency of the rating by the professionals may be partly due to the low number of raters (Marcusson *et al.* 2002; Sinko *et al.* 2005; Foo *et al.* 2013) and the inhomogeneity of specialization within the group (Marcusson *et al.* 2002; Sinko *et al.* 2005; Foo *et al.* 2013; Gkantidis *et al.* 2012). The study with the highest number of raters (12 orthodontists, 12 surgeons, and 12 laypeople; Papamanou *et al.* 2012) disposed off a relatively low number of patients with clefts (n = 12) who had

followed various surgical protocols. In the largest sample of patients (Foo *et al.* 2013), many different cleft types had been pooled.

Due to the described inconclusive results in the literature, further research is needed to assess the overall facial aesthetics achieved with a stable treatment protocol in patients with clefts, when compared with controls and rated by larger homogenous panels of laypersons and professionals.

Therefore, the aims of the present study were (1) to identify differences in the valuation of facial aesthetics of patients treated for complete unilateral left-sided cleft lip and palate and of controls and (2) to identify differences in the assessment of facial attractiveness of the two samples by three observer panels, i.e. maxillofacial surgeons, orthodontists, and laypeople. The null hypotheses of this study were that (1) no difference exists in the aesthetic evaluation of patients with unilateral clefts and controls and (2) the evaluation of facial attractiveness was not dependent on the professional background of the observers.

Materials, subjects, and methods

Ethical approval was granted for the study by the Ethics Committee of the University of Zurich, Switzerland (EK: KEK-StV-Nr. 28/2012).

Subjects

Group I (complete unilateral left-sided cleft lip and palate). The files of 20 adults (10 men, 10 women, mean age: 20.5 years, years of birth: 1978–90) treated for left-sided cleft lip and palate (left-sided cUCLP) were randomly selected from the archives of the Department of Orthodontics, University of Zurich, Switzerland. All patients had been treated according to the Zurich protocol. A passive orthopaedic plate was inserted after birth in all patients. Primary surgery included lip repair according to the protocol of Millard-Perko at 7 ± 1 months, soft palate repair according to the method of Widmaier-Perko at 18 ± 1.5 months, and hard palate closure with a mucoperiostal flap at 4.5 ± 0.5 years. During the primary and early mixed dentition, no removable or fixed orthodontic treatment was undertaken. Alveolar bone grafting with cancellous bone from the iliac crest was either performed before permanent canine eruption at the age of 10 ± 1.5 years (eight patients), or after permanent canine eruption at the age of about 17 ± 2.5 years (nine patients), or twice (three patients). In the permanent dentition, fixed orthodontic appliances were used for arch alignment. Seventeen patients had orthognathic surgery at the age of 18.5 ± 1.5 years (Le Fort I: 3 patients; surgically assisted rapid palatal expansion: 1 patient; Le Fort I and bilateral sagittal split osteotomy: 12 patients; Le Fort I and genioplasty: 1 patient). The indication for surgery to improve the facial profile was only given in five patients. In 12 patients, surgical space closure was performed in order to reduce orthodontic treatment time and to avoid bridgework or implants. However, this intervention could also have influenced the facial appearance, especially when additional surgeries for profile improvement were performed in the same run. Rhinoplasty (14 patients) and minor lip revision (9 patients) were also performed at the young adult stage.

Group II (controls). The files of 10 orthodontically treated adults (5 men, 5 women, mean age: 22.1 years, years of birth: 1987–92) with pretreatment Angle class I, minor dental problems, and no major skeletal problems were randomly selected from the post-retention files of the Department of Orthodontics of the University of Zurich, Switzerland. None of these had undergone surgical treatment.

Exclusion criteria for both patient groups were the following: patients with syndromes, other congenital facial anomalies, or outstanding facial characteristics such as piercings or tattoos. Patients with incomplete records and patients who refused the recommended treatment procedures were also excluded.

A convenience sample was selected in order not to fatigue or discourage the observer panels by presenting too many photographs for evaluation.

Methods

Existing standardized frontal and left-side profile photographs of each patient, taken 0.5–2 years post-treatment in patients with clefts and 3–5 years post-treatment in control patients, were used. All photographs had been taken in natural head position, with a neutral facial expression, and without glasses in front of the same dark blue background.

Each photograph was presented on one printed page with a modified visual analogue scale (VAS, 100 mm) from 0 to 10 underneath.

A total of 60 photographs were anonymized and presented on 60 pages in random order so that the observer was unaware of the fact that there were pairs of frontal and profile photographs.

Raters

The photographs were sent to 20 randomly selected maxillofacial surgeons (20 men, members of the Swiss Society of Oral and Maxillofacial Surgery), 20 randomly selected orthodontists (5 women and 15 men, members of the Swiss Orthodontic Society), and 20 laypersons (5 women and 15 men). Maxillofacial surgeons and orthodontists involved in the treatment were excluded from the random selection. The laypersons were of mixed socioeconomic backgrounds and were selected from incidental contacts; none of them was trained in dentistry, surgery, or aesthetics. The observers were instructed in writing to evaluate the photographs with regard to facial attractiveness as objectively as possible, without being influenced

by factors such as hairstyle, make-up, or others. The appropriate VAS score was to be circled underneath each photograph, 0 meaning 'very unattractive' and 10 meaning 'very attractive'. The observers were not informed about the patients' previous treatment and history.

Statistical analysis

Descriptive statistics were calculated and plotted to determine mean scores and standard deviations (SDs) for cleft and control patients as assessed by the different observer groups. A four-level random-intercepts mixed model was fitted, in which the VAS score given to the patient was the dependent variable and the case (control or cleft), profession (maxillofacial surgeons, orthodontists, or laypersons), view (frontal or lateral), and rater were the independent variables. The VAS scores were nested within cases, profession, and views in order to account for the clustering effects and the consequent data correlations (Rabe-Hesketh and Skrondal, 2008). All statistical analyses were conducted using Stata 12.1 (Stata Corp, College Station, TX, USA).

Results

The response rates for maxillofacial surgeons, orthodontists, and laypersons were 50, 70, and 75 per cent, respectively. Mean age and gender distribution are given in Table 1.

Data were normally distributed. The mean VAS scores, SDs, and 95% confidence intervals (95% CIs) for cleft and control patients resulting from the judgements of the different observer panels are given in Table 2.

The null hypotheses that (1) there is no difference in the aesthetic evaluation of cleft and control patients and (2) the assessment of facial attractiveness is independent from the professional background of the observers could be rejected:

In the adjusted model, compared with laypersons, the orthodontists gave higher scores (VAS: +0.69; 95% CI [0.53, 0.84]; $P < 0.001$), followed by surgeons (VAS: +0.21; 95% CI [0.03, 0.38]; $P = 0.02$). Significantly higher VAS scores for both profile and frontal photographs were found in controls (VAS for profile view: +1.97; 95% CI [1.60, 2.35]; $P < 0.001$) compared with patients with clefts. There was a high variation among raters ($P < 0.001$). No statistically significant difference was found in the scoring between frontal and lateral views (VAS for profile view: +0.13; 95% CI [−0.22, 0.49]; $P = 0.46$; Figure 1; Table 3).

The inclusion of the rater in the random effects part did not allow for the model to converge and therefore the rater was entered as a fixed effect only and the statistical model with and without the rater as a predictor was assessed for overall statistical significance using the likelihood ratio test (Table 4). The raters' profession was found to be a significant predictor, with orthodontists giving the highest scores and laypersons, the lowest ones. The high precision of the estimates indicating good study power is reflected in the associated CIs. A high variation in scores given by the individual raters was revealed ($P < 0.001$; Table 3).

Table 1 Mean age, responders, and gender distribution of the rating panels.

Rater group	Mean age, years (minimum/maximum)	Responders (female/male)
Laypersons	52.2 (34/65)	15 (5/10)
Maxillofacial surgeons	56.6 (43/65)	10 (0/10)
Orthodontists	51.7 (38/64)	14 (5/9)

Table 2 Mean visual analogue scale (VAS), scores, standard deviations (SDs), and 95% confidence intervals (CIs) for patients treated for clefts and for control patients, as assessed by the different rating panels.

Subgroups	Mean VAS scores	SDs	95% CIs
Laypersons, cleft	4.24	1.75	4.10, 4.38
Laypersons, control	6.04	1.68	5.85, 6.23
Orthodontists, cleft	4.82	1.76	4.68, 4.97
Orthodontists, control	7.14	1.74	6.94, 7.35
Surgeons, cleft	4.74	2.05	4.54, 4.94
Surgeons, control	6.48	1.91	6.22, 6.75

Discussion

We hypothesized that there are differences in the appreciation of facial aesthetics of patients with unilateral clefts and control patients and that the evaluation of facial attractiveness was dependent on the professional background of the observers. Our hypotheses were confirmed. After adjusting for view, profession, and rater, the VAS score for a random individual in the control group was 1.97 VAS score points higher than that for a random individual from the cleft group. This is in accordance with the findings of Meyer-Marcotty *et al.* (2011b), who evaluated face perception in patients with unilateral clefts compared with orthognathic class III patients and a control group. The ratings of the laypersons was compared with an objective three-dimensional analysis of the facial asymmetry. The unfavourable face perception in patients with clefts might be due to the nasolabial asymmetry that is common in these patients despite surgical interventions (Komori *et al.*, 2009, Meyer-Marcotty *et al.*, 2011b). In contrast, Fudalej *et al.* (2012) found only a weak relation between nasolabial aesthetics and symmetry in children with clefts on cropped photographs. Meyer-Marcotty *et al.* (2011a) found that patients with clefts are looked at differently; the nose

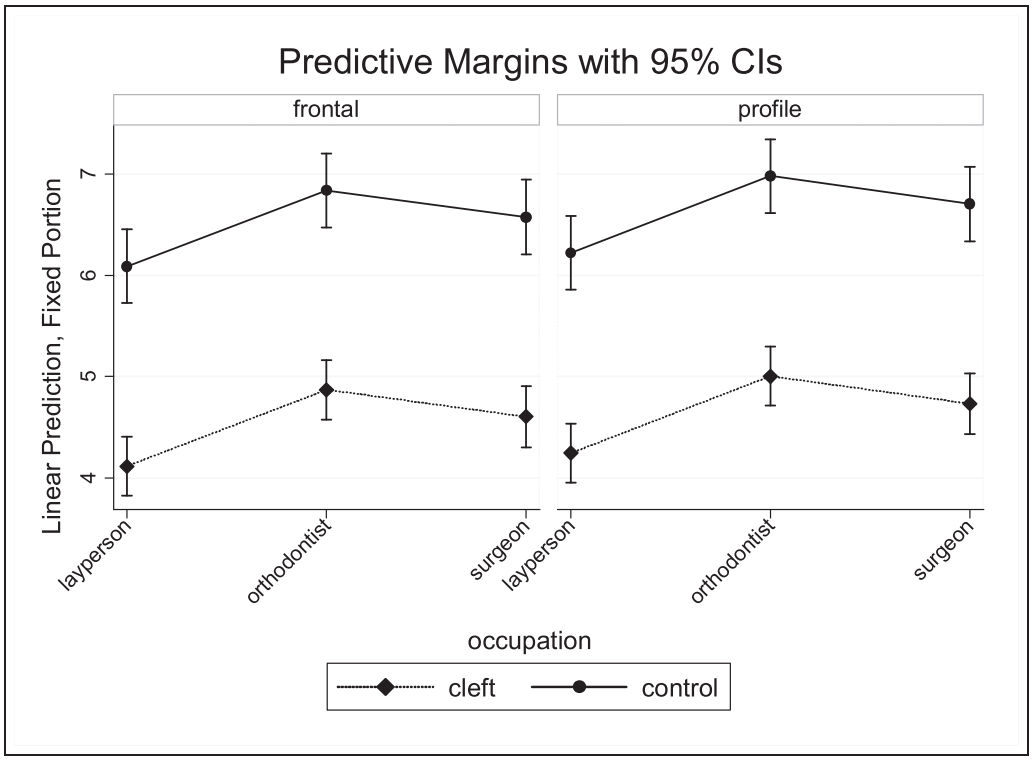

Figure 1 Mean visual analogue scale scores by occupation, case [patients with clefts (closed diamonds), or control patients (closed circles)] and view (frontal or profile); the bars show the 95% confidence intervals (CIs) for predictive margins.

in particular is observed for a longer period compared with the noses of unaffected faces. This might also have led to the lower scores of the patients with clefts in the present study.

In the present study, profession was found to be a significant predictor, with orthodontists giving the highest scores and laypersons giving the lowest ones. On average, an orthodontist gave a score of 0.69 VAS points higher and a surgeon 0.21 VAS points higher compared with the layperson after the results were adjusted for case, rater, and view. This is in contrast with the study of Foo *et al.* (2013), who found a significantly better rating of patients with clefts in the laypeople rating group compared with the professional rating group. However, their rating panels comprised five professionals (one dentist, one orthodontist, one psychologist, and two plastic surgeons) and four laypeople (two judges with clefts and two without). The small number and the difference in rating groups may have influenced the aesthetic ratings. In other studies, no significant differences in the ratings by laypeople and professionals could

Table 3 Adjusted coefficients and 95% confidence intervals (CIs) for the effect of case, view, and profession on visual analogue scale scores.

	Category	β (95% CIs)	*P*-value
Predictor			
Case	Cleft	Reference	—
	Control	1.97 [1.60, 2.35]	<0.001**
Profession	Laypersons	Reference	—
	Orthodontists	0.69 [0.53, 0.84]	<0.001**
	Surgeons	0.21 [0.03; 0.38]	0.02**
View	Frontal	Reference	—
	Profile	0.13 [−0.22, 0.49]	0.46**
Rater	—	—	<0.001*

*log-likelihood test; **Wald test

Table 4 Random effects parameters, estimated standard error and 95% confidence intervals (CIs) to provide the random effect for the independent variables.

Random effects parameters	Estimated standard error	95% CIs
Case: identity		
sd(_cons)	1.01e-09, 9.64e-09	7.59e-18; 0.13
Profession: identity		
sd(_cons)	5.03e-10, 5.73e-09	1.01e-19; 2.51
View: identity		
sd(_cons)	1.49e-09, 1.57e-08	1.62e-18; 1.38
Patient: identity		
sd(_cons)	0.62, 0.055	0.52; 0.74
sd(Residual)	1.56, 0.042	1.48; 1.64
LR test versus linear regression	$\chi^2_{(4)} = 100.67$	Probability > $\chi^2 = 0.0000$

LR, likelihood ratio; CI, confidence interval.

be found (Tobiasen, 1987, Lo *et al.*, 2002). In the studies of Meyer-Marcotty *et al.* (2011a), Gkantidis *et al.* (2012), and Papamanou *et al.* (2012), laypeople were significantly less satisfied than orthodontists and surgeons. They concluded that specialists were more familiar with the difficulties and the aesthetic consequences of treating patients with clefts. This is in accordance with the present study. But the question regarding whether the significant difference of 0.69 and 0.21 VAS points is of clinical importance remains open.

Different studies used cropped photographs to evaluate the nasolabial aesthetics, as recommended by Asher-McDade *et al.* (1991). As the purpose of the present study was to analyse the final treatment outcome at an adult age, full-face photographs were used. Full-face photographs were also used in the studies of Fabré *et al.* (2009, 2010), where the aesthetic assessment of class III subjects was analysed and also in studies evaluating the aesthetic appearance of the face after cleft treatment (Marcusson *et al.* 2002; Foo *et al.* 2013; Meyer-Marcotty *et al.* 2011b; Gkantidis *et al.* 2012; Papamanou *et al.* 2012). In order to reduce subjectivity in the present study, the raters were instructed to evaluate the photographs in the most objective way without letting them be influenced by distracting variables such as hairstyles and others.

Each observer rated the photographs on a modified VAS in order to transform the qualitative evaluation into a quantitative measurement. VAS scores are reported to be more objective, reliable, and sensitive than verbal descriptions (Grossman *et al.*, 1992; Grant *et al.*, 1999) and the evaluation procedure is less time consuming in order not to fatigue the raters.

The randomly selected cleft and control patients were of similar age and gender and all of the patients with clefts were treated with the same treatment protocol. Therefore, selection bias should be minimized. In the cleft sample, orthognathic surgery was performed if indicated, whereas the class I control patients had only minor orthodontic treatment, which is in accordance with the study of Fabré *et al.* (2009). In order to exclude further influencing factors, the control group comprised patients with no major dental or skeletal discrepancies. The sample size was 20 cleft and 10 control patients in order not to discourage the raters by presenting too many photographs. The study power is reflected in the precision of the CIs of the estimates.

The rating panels of the present study were laypeople and professionals, namely orthodontists and maxillofacial surgeons. The laypeople of this study represent the most objective group because they did not have previous experience related to clefts. Both orthodontists and maxillofacial surgeons routinely evaluate facial aesthetics in their daily practice and they had previous experience related to clefts in their careers. Therefore, they may have some type of professional bias. The orthodontists and the laypersons represented a similar wide range of age and consisted of 9 men–5 women and 10 men–5 women, respectively. The surgeons were approximately 4.5 years older and all of them were men. The intention of the present study was to represent the profession. Thus, the raters were randomly selected without taking the gender into account. To evaluate the difference between ratings by men and women, there should have been more female raters. As a study of De Smit and Dermaut (1984) did not find any significant difference between male and female judges, gender bias was not considered essential in the present study. The response rates were 70, 75, and 50 per cent for the orthodontists, laypersons, and maxillofacial surgeons. Fabré *et al.* (2009) reported similar response rates for their rating panels. In the present study, evaluation by a cleft-rating panel and self-assessment by the cleft cohort were not considered. Foo *et al.* (2013) showed that laypeople with a cleft rated significantly better the facial aesthetics of patients treated for orofacial clefts compared with laypeople without a cleft. Rating by a cleft-rating panel and comparison of the self-assessment of the cleft cohort with the ratings of different rating panels will be investigated in a further study.

Conclusions

The results of the present study revealed that, compared with laypeople, orthodontists and maxillofacial surgeons are more satisfied with the facial appearance of cleft and control patients. All observer panels reported greater satisfaction with the facial aesthetics of control patients compared with patients with clefts. Further research should evaluate whether the patients with clefts themselves perceive this difference similarly and to what extent it affects their psychosocial well-being.

References

Asher-McDade C, Roberts C, Shaw W C, Gallager C 1991 Development of a method for rating nasolabial appearance in patients with clefts of the lip and palate. The Cleft Palate-Craniofacial Journal 28: 385–90

Berk N W, Cooper M E, Liu Y E, Marazita M L 2001 Social anxiety in Chinese adults with oral-facial clefts. The Cleft Palate-Craniofacial Journal 38: 126–133

De Smit A, Dermaut L 1984 Soft-tissue profile preference. American Journal of Orthodontics 86: 67–73

Fabré M, Mossaz C, Christou P, Kiliaridis S 2009 Orthodontists' and laypersons' aesthetic assessment of class III subjects referred for orthognathic surgery. European Journal of Orthodontics 31: 443–448

Fabré M, Mossaz C, Christou P, Kiliaridis S 2010 Professionals' and laypersons' appreciation of various options for class III surgical correction. European Journal of Orthodontics 32: 395–402

Foo P, Sampson W, Roberts R, Jamieson L, David D 2013 Facial aesthetics and perceived need for further treatment among adults with repaired cleft as assessed by cleft team professionals and laypersons. European Journal of Orthodontics 35: 341–346

Fudalej P, Katsaros C, Hozyasz K, Borstlap W A, Kuijpers-Jagtman A M 2012 Nasolabial symmetry and aesthetics in children with complete unilateral cleft lip and palate. The British Journal of Oral & Maxillofacial Surgery 50: 621–625

Gkantidis N, Papamanou D A, Christou P, Topouzelis N 2012 Aesthetic outcome of cleft lip and palate treatment. Perceptions of patients, families, and health professionals compared to the general public. Journal of Cranio-Maxillo-Facial Surgery, doi:10.1016/j.jcms.2012.11.034

Grant S *et al.* 1999 A comparison of the reproducibility and the sensitivity to change of visual analogue scales, Borg scales, and Likert scales in normal subjects during submaximal exercise. Chest 116: 1208–1217

Grossman S A, Sheidler V R, McGuire D B, Geer C, Santor D, Piantadosi S 1992 A comparison of the Hopkins Pain Rating Instrument with standard visual analogue and verbal descriptor scales in patients with cancer pain. Journal of Pain and Symptom Management 7: 196–203

Komori M, Kawamura S, Ishihara S 2009 Averageness or symmetry: which is more important for facial attractiveness? Acta Psychologica 131: 136–142

Lo L J, Wong F H, Mardini S, Chen Y R, Noordhoff M S 2002 Assessment of bilateral cleft lip nose deformity: a comparison of results as judged by cleft surgeons and laypersons. Plastic and Reconstructive Surgery 110: 733–8; discussion 739

Marcusson A, Paulin G, Ostrup L 2002 Facial appearance in adults who had cleft lip and palate treated in childhood. Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery 36: 16–23

Meyer-Marcotty P, Alpers G W, Gerdes A B, Stellzig-Eisenhauer A 2010 Impact of facial asymmetry in visual perception: a 3-dimensional data analysis. American Journal of Orthodontics and Dentofacial Orthopedics 137: 168.e1–8; discussion 168

Meyer-Marcotty P, Gerdes A B, Stellzig-Eisenhauer A, Alpers G W 2011a Visual face perception of adults with unilateral cleft lip and palate in comparison to controls-an eye-tracking study. The Cleft Palate-Craniofacial Journal: 48: 210–216

Meyer-Marcotty P, Kochel J, Boehm H, Linz C, Klammert U, Stellzig-Eisenhauer A 2011b Face perception in patients with unilateral cleft lip and palate and patients with severe class III malocclusion compared to controls. Journal of Cranio-Maxillo-Facial Surgery 39: 158–163

Murray J C 1995 Face facts: genes, environment, and clefts. American Journal of Human Genetics 57: 227–232

Papamanou D A, Gkantidis N, Topouzelis N, Christou P 2012 Appreciation of cleft lip and palate treatment outcome by professionals and laypeople. European Journal of Orthodontics 34: 553–560

Pruzinsky T 1992 Social and psychological effects of major craniofacial deformity. The Cleft Palate-Craniofacial Journal 29: 578–84

Rabe-Hesketh S, Skrondal A 2008 Multilevel and Longitudinal Modeling Using Stata, 2nd edn. Stata Press, College Station

Sinko K, Jagsch R, Prechtl V, Watzinger F, Hollmann K, Baumann A 2005 Evaluation of esthetic, functional, and quality-of-life outcome in adult cleft lip and palate patients. The Cleft Palate-Craniofacial Journal 42: 355–361

Tobiasen J M 1987 Social judgments of facial deformity. The Cleft Palate Journal 24: 323–327