Evaluation of Facial Appearance among Patients with Repaired Unilateral Cleft Lip and Palate: Comparison of Patient- and Clinician-Ratings of Satisfaction

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The objective of this study was to determine the levels of patient-satisfaction on facial and dental appearance compared with clinician ratings. Participants included 61 patients with repaired unilateral cleft lip and palate (UCLP), aged 14-25 years. Raters comprised three cleft team clinicians. A Likert scale was used to assess the levels of satisfaction of the patients themselves and the clinicians. The results revealed that the patients were moderately satisfied with their appearance. Nose was the least satisfactory feature, followed by lip appearance. When compared to the clinician ratings, the patients were less satisfied with their own nose and lip, but more satisfied with teeth. Concerning age, self-assessment did not differ between adolescents and young adults. Females were less likely to be satisfied compared to males, but the difference was not statistically significant. In conclusion, patients with repaired UCLP were moderately satisfied with their facial and dental appearance. Clinician- and patient-opinions were different in some aspects. This study highlights the importance of patient satisfaction as a meaningful treatment outcome assessment, which could lead to an improvement in cleft care to meet the patient expectations.

Keywords: *Patient satisfaction, Clinician rating, Appearance, Young adult, Adolescent, Cleft lip and palate*

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Facing a multitude of problems, cleft patient requires several therapeutic procedures of rehabilitation starting immediately at birth and continuing to adulthood. The optimal procedures as well as their timing remain controversial[1,2], and different treatment protocols have been adopted by various cleft centers worldwide. Comprehensive examination of the multidimensional treatment outcome has been focused in many cleft centers[3-6]. Among multiple aspects concerning treatment outcome audits, facial esthetic assessment is one of the most relevant measurements of treatment success[7-10]. This is because a major goal of the surgical and dental treatment in craniofacial anomalies is to improve facial esthetics and social acceptability[11,12].

Facial appearances of cleft patients could be determined by objective measurement[13,14] or subjective assessment of esthetic perception among individuals involved in treatment process, namely clinicians, patients and their parents. The patients' satisfaction with their own appearance could be considered as most important because it is regarded as a crucial requirement for healthy psychosocial development especially in adolescents, when facial esthetics is important for their self-perception and self-esteem[15]. Even though the literature suggests that an individual's psychosocial well-being is not affected greatly by cleft lip and palate (CLP)[16-19], some problems appear to be related to dissatisfaction with facial appearance[18,20]. Moreover, earlier studies did show that facial esthetics is an important aspect of quality of life (QoL) in adults with repaired CLP[21-23]. In a cross-sectional study, Mani et al[23] found an association

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between lower mental health, QoL and dissatisfaction with nasal appearance. Furthermore, dissatisfaction with facial appearance was reported to be a significant predictor of depression[18].

Even after receiving surgical and dental rehabilitations to normalize their appearance, patients with clefts still seem to have concerns about their appearances, especially related to the cleft deformity[18,21]. Patients affected by a visible cleft type including UCLP, bilateral cleft lip and palate (BCLP) and cleft lip were more dissatisfied with their appearance than patients with invisible impairments, such as cleft palate and submucous cleft[15,24]. Gender has been reported to be associated with self-assessed appearance[18,21,23]. In regard to age, Thomas et al[15] found 20-year-old subjects with cleft were significantly more satisfied with their appearance than younger counterparts although some older subjects remained greatly dissatisfied with some aspects of their facial appearance.

Clinician's opinion about facial appearance may have important implications for management. They may influence patients' and parents' perceptions of the need for treatment. The clinician's opinion is influenced by gender, type of training and familiarity with cleft condition[25-28]. Eliason, Hardin and Olin[25] reported that judges familiar with cleft condition assigned more negative ratings of facial appearance than did judges unfamiliar with the condition. In addition, male clinicians commented more negatively than did female clinicians.

To understand the perspectives of adolescent and young adult patients about facial appearance better, the present study was designed with two objectives:

1) To determine and compare the level of satisfaction with facial and dental esthetics among UCLP patients and clinicians.

2) To examine whether psychosocial developmental stages (adolescent versus adult) and gender have an important impact on the satisfaction with facial and dental esthetics of UCLP patients.

Material and Method

Patients

Potential participants had repaired complete UCLP, aged between 14 and 25 years old, who attended routine or follow-up orthodontic visit at Orthodontic clinic, KKU Cleft Center during 3 months period of data collection. Patients with syndromes, other congenital anomalies or intellectual disability were excluded. Sixty-one complete UCLP were matched with inclusion criteria. Of all subjects, 57 were undergoing various stages of orthodontic treatment with fixed appliances while four subjects had completed their orthodontic treatment. To determine the effect of age on the level of satisfaction, subjects were grouped to adolescent (aged between 14 to 17 years) and young adult groups (aged between 18 to 25 years). The participant demographics are summarized in Table 1.

*Rater*s

The assessors comprised three cleft team clinicians: a plastic surgeon, a maxillofacial surgeon and an orthodontist all experienced in cleft lip and palate care.

Procedures

Patient satisfaction

The subjects were invited to participate in the study by a research assistant who was not a part of the cleft team. Confidentiality was assured for every subject, and it was also made clear at the start that the data would not be connected with the subject's cleft care specialists. Patients' level of satisfaction was collected by a self-administered questionnaire. With the aid of a face mirror, the subjects rated five features: 1) overall facial appearance, 2) nose, 3) upper lip, 4) profile, and 5) anterior teeth. Patients rated each feature based on 5-point Likert scale which scores: 1 = not at all satisfied, 2 = slightly satisfied, 3 = moderately satisfied, 4 = very satisfied, and 5 = completely satisfied. Higher scores represent greater satisfaction with appearance.

Clinician rating

A set of photographs for each subject was taken, consisting of extra-oral photographs comprising a full face view with lips at rest, full face smiling view, profile and oblique view of an affected side, close-up

Table 1. Participant demographics

Demographic data	Adolescent group (n = 31)	Adult group (n = 30)	Total (n = 61)
Age			
Mean ± SD	15.9±1.1	20.3±2.0	18.1±2.7
Gender, n (%)			
Male	13 (41.9)	15 (50)	28
Female	18 (58.1)	15 (50)	33

view of lip and nose area, inferior view of nose and a frontal intra-oral view with the teeth in occlusion. The subjects stood 0.5 m in front of a standardized cobalt blue non-reflective background. All the photographs were taken with a 60-mm lens. The 4x6" color photographs were mounted in photo albums, one patient to each page.

The indirect assessments with color photographs were separately performed by three clinicians. They were instructed to rate facial and dental esthetics based on a 5-point Likert scale. Each clinician had a practice session before the start of data collection to become familiar with the assessment procedure using the clinician assessment guide (Appendix 1). The assessment for each subject took about 1 minute.

Ethical approval was granted for the study by the Khon Kaen University Ethics Committee for Human Research.

Reliability of clinician ratings

In order to assess intra-observer reliability of clinician rating, replicate ratings were made on 25 randomly selected subjects within 1-month interval. Weighted Kappa test was used to evaluate reliability using STATA version 10 (STATA Corp, LP Station TX, USA). According to Altman[29], intra-observer reliability ranged from moderate to good (Kappa value 0.52 to 0.78) in most features except profile which ranged from moderate to very good (Kappa value 0.58 to 0.86).

Statistical analysis

Clinician rating of appearance was derived as an average of three clinicians' scores. Shapiro-Wilk test showed that differences between patient and clinician ratings were normally distributed. Thus, paired t-test was used to determine the differences between clinician and patient satisfaction levels. Student's t-test was used to compare the patient satisfaction levels between different genders, and age groups. The significance level was set at 0.05 for all tests. Statistical analyses were performed using the Statistical Package for the SPSS Version 17.0 (SPSS Inc., Chicago, IL, USA).

Results

Patient- versus clinician ratings

For the patients, the highest mean score achieved was for overall face (mean 3.57 out of 5) followed by anterior teeth (mean 3.52), and facial profile (mean 3.30). Nose was found to be the lowest-rated feature (mean 2.87) (Table 2).

Comparative data indicate that there were significant differences in patient satisfaction levels and clinician esthetic ratings with respect to nose (mean difference -0.55, 95% confidence interval (CI) = -0.80 to -0.31, $p<0.001$), lip (mean difference -0.40, 95% CI = -0.64 to -0.17, $p = 0.001$) and dental appearance (mean difference 0.29, 95% CI = 0.04 to 0.53, $p = 0.02$). The mean scores for the ratings by the patients and clinicians are shown in Table 2.

Adolescents versus young adults

Comparisons were made of how adolescents rated their appearance compared to young adults. No significant difference in the levels of satisfaction was evident considering to age group (Table 3).

Males versus females

Comparison of the satisfaction levels between male and female subjects was shown in Table 4. There was no significant difference between the ratings of males and females, although the females tended to be less satisfied with every feature of their facial appearance.

Discussion

Human judgment is accepted to be a reliable

Table 2. Comparisons of levels of satisfaction between the cleft patients and by the clinicians

Features	Levels of satisfaction mean ± SD		Mean difference (95% confidence interval)	*p*-value
	Patient	Clinician		
Overall face	3.57±0.83	3.66±0.37	-0.08 (-0.30, 0.14)	0.45
Nose	2.87±0.96	3.42±0.39	-0.55 (-0.80, -0.31)	<0.001
Upper lip	3.03±0.93	3.44±0.47	-0.40 (-0.64, -0.17)	0.001
Facial profile	3.30±1.02	3.53±0.64	-0.24 (-0.48, 0.01)	0.06
Anterior teeth	3.52±0.98	3.24±0.59	0.29 (0.04, 0.53)	0.02

Table 3. Patient satisfaction with facial and dental appearances by age group

Features	Levels of satisfaction mean ± SD		Mean difference (95% confidence interval)	*p*-value
	Adolescents	Adults		
Overall face	3.39±0.80	3.77±0.82	-0.38 (-0.80, 0.04)	0.07
Nose	2.87±0.96	2.87±0.97	0.00 (-0.49, 0.50)	0.99
Upper lip	2.87±0.88	3.20±0.96	-0.33 (-0.80, 0.14)	0.17
Facial profile	3.16±1.00	3.43±1.04	-0.27 (-0.80, 0.25)	0.30
Anterior teeth	3.48±0.89	3.57±1.07	-0.08 (-0.59, 0.42)	0.74

Table 4. Patient satisfaction with facial and dental appearances by gender

Features	Levels of satisfaction mean ± SD		Mean difference (95% confidence interval)	*p*-value
	Males	Females		
Overall face	3.61±0.83	3.55±0.83	0.06 (-0.37, 0.49)	0.77
Nose	2.93±1.02	2.82±0.92	0.11 (-0.39, 0.61)	0.66
Upper lip	3.14±0.89	2.94±0.97	0.20 (-0.28, 0.68)	0.40
Facial profile	3.50±1.04	3.12±0.99	0.38 (-0.14, 0.90)	0.15
Anterior teeth	3.57±1.00	3.48±0.97	0.09 (-0.42, 0.60)	0.73

tool in the assessment of cleft impairment and ratings of facial attractiveness decrease as the severity of impairment increases[30]. To find out if cleft subjects currently under treatment and cleft team clinicians have the same opinion about facial and dental appearance, the self-assessment of the subjects was compared with the assessment of color-printed photographs by a group of cleft team clinicians. Methodological approaches, stimulus type and professional experience have been reported to have a potential influence on such a subjective assessment[28]. Although direct clinical assessment might be the standard evaluation method, use of indirect photographic assessment was more convenient for both subjects and clinicians. Moreover, photographic assessments can avoid stimulation of patient concerns and unnecessary referral for further treatment urged during clinical assessment. To provide the assessors as good as possible view of the patient's features as the patient had with the mirror, close-ups of nasolabial area, submento-vertex and full face oblique view were included. From the points of view of the three clinicians participating in this study, standardized photography provides more uniform viewing of a subject for all clinicians than with a live subject assessment.

A personal bias has been reported to be a reason for poor agreement on lip evaluation among surgeons[31]. The use of a panel of representative judges to generate a single mean score for each case has been recommended to remove the inter-observer bias and also improve reliability[8,18,32]. For this reason, the averages of three clinician ratings were used in this study.

Intra-observer reliability of the clinicians obtained from two assessments ranged from moderate to very good. This corresponds to previous studies[28,33] in which the clinicians consistently rated the esthetic appearance of cleft impairments. In contrast, reliability of self-rating of subjects with cleft was considered to be unjustified because self-perceived satisfaction is subjective by nature and could be influenced by what an individual has experienced such as being teased[16]. Bias of introspection about their esthetic appearances by the subjects may occur if they are required to repeat their assessments.

Although the mean scores of patient-ratings showed a moderate satisfaction in this study, the subjects tended to be satisfied with their overall face

(mean 3.57) and dental esthetics (mean 3.52). They also accepted their facial profile (mean 3.30) and lip (mean 3.03), while the nose was the lowest-rated feature (mean 2.87).

When compared with the clinicians, the subjects were inclined to be less satisfied with their facial appearances, especially with nose and upper lip that reached a statistically significant difference. This is in accordance with previous studies[18,21] which reported a trend towards more unfavorable ratings of appearance by the patients themselves compared with the clinician-ratings. This was different from a previous study on non-cleft adolescents, which found that they tended to more satisfied with the attractiveness of their own faces than ratings by others[34]. The discrepancy between patient ratings of satisfaction and clinician ratings may be explained by the fact that patient satisfaction seems not to be dependent on actual appearance alone. Realistic self-perception and expectation could have an effect on patient satisfaction. Richman, Holmes and Eliason[35] found that realistic self-perceptions of facial appearance in adolescents with CLP depend on how psychologically adjusted they were. The well adjusted CLP patients tended to have a realistic perception of appearance compared to teacher ratings of facial appearance. A literature review[36,37] suggests that a vital factor influencing patient satisfaction is patient expectation. Satisfaction of patients is related to how their perceptions from treatment benefits meet their expectations. Patients with lower expectation tend to be more satisfied[36]. Unrealistic expectations regarding the outcome of the cosmetic surgery may also predict a poor psychosocial outcome[38]. However, information about subjects' psychosocial conditions and their expectation with treatment was not an objective of this study. Further studies with a specific design are needed to assess the effect of these factors on patient satisfaction. Most subjects in this study were currently undergoing orthodontic treatment. It was possible that they might defer their judgements of esthetic outcome while waiting on completion of treatment. On the other hand, the esthetic judgements of the clinicians are also a subjective perception. The clinicians tended to have favorable esthetic outcome judgements because their clinical experiences may tell them that those patients could not gain significant improvement from further revision. Discrepancies of perception among patients and clinicians highlight the need of careful evaluation of patients' concern during determining treatment outcome and planning for revision of secondary deformity.

Concerns about the nose appearance were the commonest reported by both normal subjects[39] and cleft subjects[18,40]. Dissatisfaction with the nose can cause a psychological problem[23] as well as an esthetic issue for patients with clefts. Adults with repaired clefts generally expect better results from secondary rhinoplasty[18]. Finding the nasal deformity is the most appearance concern among patients with repaired anterior oral cleft, this study may encourage continuing search for improvement in nose surgery and in adjunctive procedure including nasoalvolar molding (NAM).

The dental esthetics obviously contributes to facial appearance. Teeth were the first feature laypeople noticed when looking at photographs of BCLP who finished their treatment[41]. Only a few studies reported patient satisfaction with dental esthetics. In a study from Malaysia[42], CLP patients aged 12 to 17 years and their parents were least satisfied with teeth because the subjects had not yet undergone orthodontic treatment. A follow-up study of Swedish adults with repaired clefts[18] reported no difference in satisfaction with teeth between cleft and non-cleft groups. In the current study, although most of the subjects were still undergoing treatment in the Orthodontic Clinic, they were quite satisfied with their teeth (mean 3.48). Regarding a comparison of dental esthetics, although a statistical test suggests significant difference, the means for patient (3.52) and clinicians (3.24), and the CI of the differences between those means (0.04-0.53) suggests that the significant difference may be unimportant from an everyday clinical practice. No previous studies have reported such a comparison and further investigations are required to gain insight into different perceptions among patients and clinicians.

The present study found no differences between males and females in self-assessment of esthetics. This was consistent with a study from Northern Ireland[20], which also reported no differences between males and females when comparing the level of satisfaction in children and young adults aged 8 to 21 years old. In BCLP, levels of satisfaction with appearance did not differ between males and females[22]. But this is in contrast to a study from Sweden[18] which found that, in adults with repaired cleft, female ratings of their mouth and profile were significantly poorer than those of males. In another study from Sweden by Mani et al[23], females were less satisfied with nasal appearance than male counterparts. The authors

concluded that gender is a potential predictive factor for satisfaction of nasolabial appearance. In patients treated according to the Vienna concept[21], females also were more likely to be dissatisfied with esthetic outcomes than males. The conflicting result in our study can be attributed to different study population. It may also reflect a current cultural trend that young people in this generation tend to similarly concern over appearance regardless of gender[24].

There appeared to be no difference in the level of satisfaction between adolescents and young adults in the current study. This may indicate that the level of satisfaction does not increase over time. This is in line with a study conducted among 5 to 18 year-old subjects with CL and/or CP that also found no age differences in patient satisfaction[24]. However, few studies have reported an influence of age on satisfaction. A study from United Kingdom, Thomas et al[15], found that children and young adolescents aged 10 to 15 years were more dissatisfied with their appearance than subjects aged 20 years. On the other hand, another study from United Kingdom[20], revealed that children became less happy with their facial appearance as they got older. However, there is a lack of longitudinal cohort study on how satisfaction with facial appearance alters with age among subjects with a cleft. Such a study is needed to identify the effect of age change over satisfaction and to determine whether specific developmental ages are influential in the occurrence of dissatisfaction with appearance.

Satisfaction with appearance may depend on treatment protocol[43] and number of secondary rhinoplasties[23,44]. In this study, effect of these factors was not evaluated because the subjects had their surgery from various hospitals and treatment record concerning operation method and number of revision was not available. These subjects may have been anticipating further lip/nose revisions, orthognathic surgery and dental correction during their later years. Future studies of the patient satisfaction with their appearance would allow for an assessment of final treatment outcome.

Conclusion

Adolescents and young adults with repaired UCLP were moderately satisfied with their appearance and were least satisfied with nose. Compared with clinician ratings, the patients were less satisfied with their nose and lip appearance, but more satisfied with dental esthetics. There was no difference in self-assessment between different age groups and genders.

What is already known on this topic?

Medical professionals usually judge facial esthetics more optimistically, while cleft patients are less satisfied with their cleft-related facial features.

No studies have reported a comparison of patient's and professional's perception on dental esthetics.

What this study adds?

The nasal deformity is the most appearance concern among repaired cleft patients.

Although patients are less happy with their nose and lip appearance, they are more satisfied with their dental esthetics than clinicians are.

The discrepancy between patient and clinician found in this study highlights the importance of patient satisfaction as a meaningful outcome assessment of treatment outcome, which could lead to an improvement in cleft care to meet the patient expectations.

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Potential conflicts of interest

None.

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Appendix 1. Clinician assessment guide
Criteria for facial and dental assessment, based upon the characteristic stigmata of repaired UCLP

Features	Criteria
Overall face	Overall esthetic of total face
Nose	Nostrils symmetrical in size, shape and height
	Alar of nose depression
	Centrality of columella and nose tip
	Alar bases symmetrical
	Nasal tip projection
Upper lip	Symmetry of the cupid's bow
	Symmetry of lateral lip
	Scarring, notching
	Whistle deformity
	Vermilion continuity, vermilion/white-roll mismatch
Facial profile	Balance facial profile
	Relationships of soft tissue of the lower facial and nasolabial profile
	Upper and lower lips relationship
Anterior teeth	Incisal show
	Smile line, smile arch
	Size, shape, number and proportion of anterior teeth
	Relationship of dental and facial midline

การประเมินความสวยงามของใบหน้าผู้ป่วยปากแหว่งเพดานโหว่ข้างเดียว: เปรียบเทียบความพึงพอใจระหว่างผู้ป่วยและทีมแพทย์

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วัตถุประสงค์ของการศึกษานี้เพื่อประเมินระดับความพึงพอใจของผู้ป่วยต่อความสวยงามของใบหน้าและฟัน เปรียบเทียบกับการประเมินโดยทีมแพทย์ กลุ่มตัวอย่างประกอบด้วย ผู้ป่วยปากแหว่งเพดานโหว่ข้างเดียว 61 คน อายุระหว่าง 14-25 ปี และทีมแพทย์ 3 คน ประเมินความพึงพอใจต่อความสวยงามของผู้ป่วยโดยใช้มาตรวัดของลิเคิร์ท พบว่าผู้ป่วยยังมีความพึงพอใจต่อความสวยงามของใบหน้าและฟันของตนระดับปานกลาง จมูกเป็นส่วนที่ผู้ป่วยพึงพอใจน้อยที่สุดและริมฝีปากเป็นอันดับต่อมา เมื่อเปรียบเทียบกับการประเมินของทีมแพทย์ พบว่าผู้ป่วยพึงพอใจต่อจมูกและริมฝีปากน้อยกว่าอย่างมีนัยสำคัญทางสถิติแต่พึงพอใจต่อความสวยงามของฟันมากกว่า ผลการเปรียบเทียบระหว่างกลุ่มอายุไม่พบความแตกต่างของระดับความพึงพอใจของวัยรุ่นและผู้ใหญ่ตอนต้น เมื่อเปรียบเทียบระหว่างเพศ พบว่าผู้ป่วยหญิงมีระดับความพึงพอใจน้อยกว่าผู้ป่วยชาย แต่ไม่แตกต่างอย่างมีนัยสำคัญทางสถิติ สรุปผลการศึกษาผู้ป่วยมีความพอใจระดับปานกลางต่อความสวยงามของใบหน้าและฟัน ความพึงพอใจมีความแตกต่างกันระหว่างผู้ป่วยและทีมแพทย์ในบางด้าน ซึ่งชี้ให้เห็นความสำคัญของการประเมินความพึงพอใจของผู้ป่วยปากแหว่งเพดานโหว่ต่อการพัฒนาการรักษาเพื่อให้ตอบสนองต่อความคาดหวังของผู้ป่วย