



การประเมินรูปลักษณ์ใบหน้าและคุณภาพชีวิตในผู้ป่วยอายุ 4-6 ปี ที่มีภาวะปากแหว่งเพดานโหว่: การศึกษาในศูนย์ตะวันฉาย ประเทศไทย

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Evaluation of Facial Appearance and Quality of Life in 4-6 Year Old Patients with Cleft Lip and Palate: A Study at the Tawanchai Center, Thailand

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บทคัดย่อ

หลักการและวัตถุประสงค์: ภาวลักษณ์และคุณภาพชีวิตของผู้ป่วยปากแหว่งเพดานโหว่เป็นสิ่งสำคัญ ดังนั้นการประเมินผลลัพธ์การรักษาและเปรียบเทียบผลลัพธ์ที่ได้พร้อมนำมาพัฒนาการดูแลและคุณภาพชีวิตของผู้ป่วยจึงเป็นสิ่งจำเป็น

วิธีการศึกษา: การศึกษาเชิงพรรณนาภาคตัดขวางในผู้ป่วยปากแหว่งเพดานโหว่อายุ 4-6 ปี ที่ศูนย์ตะวันฉาย ประเมินความสวยงามของจมูก ริมฝีปาก และภาพรวมของจมูก โดยใช้ภาพถ่ายดิจิทัลตามมาตรฐาน ให้ค่าคะแนน 5 ระดับ โดยผู้เชี่ยวชาญทางการแพทย์ที่มีและไม่มีประสบการณ์ ประเมินคุณภาพชีวิตจากผู้ดูแล โดยใช้แบบสอบถาม THAICLEFT QoL ใช้สถิติเชิงพรรณนาในการวิเคราะห์ข้อมูล ใช้สัมประสิทธิ์อัลฟ่าครอนบาช ในการตรวจสอบความเชื่อมั่นระหว่างผู้ประเมิน และใช้ Wilcoxon signed-rank test ในการเปรียบเทียบผลการประเมิน

ผลการศึกษา: พบว่า มีผู้ป่วยปากแหว่งเพดานโหว่ข้างเดียวและสองข้างชนิดละ 6 ราย ไม่พบความแตกต่างของความสอดคล้องระหว่างกลุ่มผู้ประเมิน ผลการประเมินด้านความสวยงามของริมฝีปากและภาพรวมของจมูก จากผู้เชี่ยวชาญทางการแพทย์ที่มีประสบการณ์มีคะแนนสูงกว่าผู้ไม่มีประสบการณ์อย่างมีนัยสำคัญทางสถิติ ($p = 0.04$ และ 0.02 ตามลำดับ) ผู้เชี่ยวชาญที่ไม่มีประสบการณ์มีความเชื่อมั่นสูงในการประเมินริมฝีปากและจมูก (Cronbach's alpha = $0.76 - 0.88$) การประเมินคุณภาพชีวิตด้านการรักษา การให้บริการ และการรักษา มีความพึงพอใจในระดับมาก ส่วนผลกระทบต่อครอบครัวอยู่ในระดับต่ำ

สรุป: การประเมินรูปลักษณ์ของใบหน้าโดยแพทย์ผู้ไม่มีประสบการณ์มีมุมมองทางด้านความสวยงามแตกต่างกัน ส่วนคุณภาพชีวิตผู้ดูแลประเมินผลค่อนข้างดี ผลกระทบต่อครอบครัวมีน้อย แสดงให้เห็นว่ามีคุณภาพชีวิตค่อนข้างดี อย่างไรก็ตาม ยังต้องนำผลการศึกษานี้มาพัฒนาการดูแลผู้ป่วยต่อไป

คำสำคัญ: ปากแหว่ง เพดานโหว่, คุณภาพชีวิต, ความพึงพอใจของผู้ป่วย, ภาวลักษณ์ของใบหน้า, การประเมินด้วยภาพถ่าย

Abstract

Background and objective: Facial Appearance and quality of life of patients with cleft lip and palate are essential, so the evaluation and comparison of treatment outcome to improve care and quality of life (QoL) of cleft patients are required.

Methods: We conducted a cross-sectional descriptive study with cleft lip and cleft palate (CLP) patients aged 4-6 years at the Cleft Center. Aesthetic outcomes for the nose, lip, and nasolabial region (NR) were evaluated using standardized digital photographs rated on a five-point scale by both experienced and inexperienced evaluators. QoL was assessed with THAICLEFT QoL Questionnaires. Descriptive statistics are used to analyze data. Inter-rater reliability is evaluated using Cronbach's alpha coefficient. The Wilcoxon signed-rank test is employed to compare esthetic evaluations between two groups of evaluators.

Results: The study included 6 unilateral and 6 bilateral CLP cases. There were no significant differences in overall agreement between professional groups. Experienced rated the aesthetic outcomes for the lip and nasolabial region significantly higher than inexperienced ($p = 0.04$ and 0.02 , respectively). Inexperienced demonstrated high reliability in assessing the lip and nose (Cronbach's alpha = $0.76 - 0.88$). QoL scores were high for medical treatment, service, and costs; moderate for social psychology; and low for family impact.

Conclusions: The evaluation of facial appearance by experienced and inexperienced evaluators reveals different perspectives on aesthetics. In terms of QoL evaluation, caregivers indicated a good score, and the impact on the family appears to be minimal, indicating a relatively good overall quality of life. However, the findings from this study should be used to further improve patient care in the future.

Keywords: cleft lip and palate, quality of life, patient satisfaction, facial appearance, photo-graphic evaluation

Introduction

Cleft lip and/or palate (CLP) is the most common congenital craniofacial abnormality. In Thailand, the incidence is reported to be about 2.14:1000 live births and 2.28:1000 live births in Northeast of Thailand¹. The cause happens with multiple factors such as genetic and environment². The CLP are associated with many problems, such as speech disorder, hearing deficit, chronic ear infection, dental deformities, and psychosocial problem including affecting the quality of life (QoL) in terms of physical, mental, social and economic aspects of the patient, family and the nation as well.^{3,4}

Providing care by interdisciplinary team with standard protocol for treatment is required to cleft patients. In the early phase, from birth to 5 years old is the most important periods for primary surgery including cheiloplasty and palatoplasty. In the stage of speech and language development of children allows them to face the external social environment, including peers in the school. Therefore, evaluating treatment outcomes during age range 4-6 years is essential for both of interdisciplinary team and treatment assessment⁵.

According to the field literature related to equality of life, three parts have been identified: (1) the physical health; (2) psychological health; and (3) social health⁶. To assess the QoL of patients, we employed the THAICLEFT QoL Questionnaire developed by the World Health Organization Quality of Life (WHOQoL) group (1998), consisting of five dimensions⁷. The five parts are as follows: 1) healthcare; 2) provision of counsel; 3) financial treatment; 4) social psychology aspect; and 5) the impact on the family⁷. All five parts have an impact on the QoL of both patients and their families.

The facial appearance is also an important factor related to QoL. Surgical procedures on the cleft lip and nose are significant for patients with CLP⁸. The nose, being a body part feature in facial aesthetics, contributes to the overall facial appearance. The attractiveness of the face is not solely dependent on

the mouth but also on the nose, which occupies a central position on the face⁹. The previous study for patients aged 4-6 years found that they gave esthetic score in low level. Increased surgical interventions and lower socioeconomic and social status may contribute to significant behavioral problems¹⁰. Satisfaction with facial appearance and organ function significantly correlate with the QoL in terms of mental health and emotional well-being among patients^{11,12}. Cleft children aged 2 to 6 years have struggled with their social interaction, eating, habits, daily activities, and overall QoL¹³. The assessment of facial appearance in unilateral CLP aged between 6-12 years conducted by inexperienced and experienced medical professionals, shows high interrater agreement and satisfaction levels in the assessment of lip and nose aesthetics¹⁴.

The aim of this study was to determine the facial appearance and QoL outcomes in children with CLP aged between 4-6 years who have severe condition and the critical age for entering school and society, including peer groups and adaptation. Evaluating treatment outcomes will aid in planning more effective surgical care for patients.

Methods

Setting

This is a cross-sectional descriptive study of CLP patients aged 4-6 years' old who received primary surgery at the Tawanchai Center, Srinagarind Hospital, Faculty of Medicine, and Faculty of Dentistry, Khon Kaen University, from October 2020 to September 2021. Patients without comorbidities and whose parents agreed to participate in the study were included. The population was calculated as 108 with a confidence level of 95% using a formula¹⁵. This resulted in a sample group of 12 participants.

$$n = \frac{NZ^2 \frac{\sigma^2}{d^2}}{d^2(N-1) + Z^2 \frac{\sigma^2}{d^2}}$$

Using the overall mean score of treatment outcomes in terms of quality of life and self-image among 10-year-old patients with cleft lip and palate,

with a standard deviation (σ) of 1.39 ($\sigma^2 = 1.39$), $Z\alpha/2 = 1.96$, $d = 0.45$, the population (N) is 108.

Inclusion criteria

Patients with complete unilateral and bilateral lip and cleft palate, without any associated syndromes, who are willing to participate in the study.

Exclusion criteria

Patients who have only a cleft lip or only a cleft palate.

The tool employed in this study

- Standard 2D photographs were taken from three positions — frontal, left lateral, and right lateral — with the child's face inclined at a 45-degree angle in each view.
- The evaluators were medical professionals with over 10 years of experience in surgery and treatment of children with cleft lip and cleft palate, including two plastic surgeons, one orthodontist, and one pediatric dentist. In addition, there were four inexperienced medical professionals, including two general surgeons and two general dentists.
- Evaluation parameters were used to assess the facial appearance including lip, nose, and NR. This assessment involved cropping the 2D photograph according to standardized position, with a blue background to enhance facial visibility. A total of four images were captured for evaluation (Figure 1). The lip and nose were evaluated individually, and NR was evaluated as a whole. The esthetic aspects analyzed were symmetry and lip volume, continuity of the vermilion of the upper lip, scar, symmetry of the nose tip and columella, alar base insertion, nasolabial angle, and maxillomandibular relationship¹⁶. Each picture was assessed for one minute, followed by a five-second break.

The evaluators would be informed the detailed assessment and perform a practice test before conducting the real evaluation.

The scoring criteria were based on five rating scale; scores ranging from 1.0 – 2.5 indicated a “good level”, scores ranging from 2.6 – 3.5 indicated a “fair level”, and scores ranging from 3.6 – 5.0 indicated a “poor level”. To ensure consistency in the assessments, the criteria were also tested on three non-sample cleft palate patients.

- THAICLEFT QoL Questionnaires. The questionnaires were used to assess the satisfaction of parents with the treatment of their child, consisting of 41 items that were divided into five parts, including 1) medical treatment (8 items), 2) service (3 items), 3) treatment cost (4 items), 4) social psychology (6 items), and 5) the impact on family (20 items)⁷.

Evaluation and Interpretation Criteria: 1.00–1.50 indicates the lowest level of satisfaction, 1.51–2.50 indicates a low level of satisfaction, 2.51–3.50 indicates a moderate level of satisfaction, 3.51–4.50 indicates a high level of satisfaction, 4.51–5.00 indicates the highest level of satisfaction¹⁵.

Medical record for each patient

All participants, parents, evaluators received both oral and written information about this study.

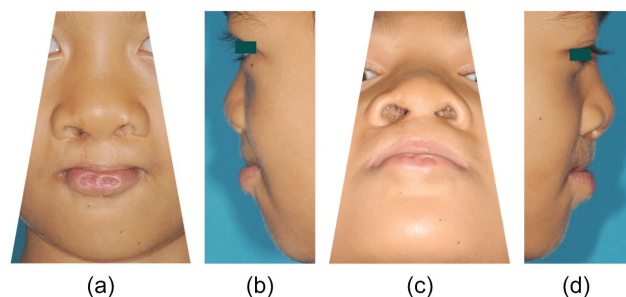


Figure 1: Representative standardized photo taken of each patient (a) front view; (b) left lateral view; (c) basal view and (d) right lateral view.

Statistical Analysis

The statistical analysis of data was performed descriptive to analyze percentage, mean, and standard deviation. The inter-rater reliability of two groups of evaluators was evaluated using the Cronbach's alpha coefficient. To analyze the differences between the two groups of experienced and inexperienced evaluators in terms of satisfaction ratings across the four dimensions, the Wilcoxon signed-rank test was used with a confidence level of 95% (p value < 0.05).

The study was approved by the Center for Ethics in Human Research Committee, Khon Kaen University (KKU) (HE651269).

Results

There were 12 patients with 6 having unilateral and 6 having bilateral CLP (50%). The average age of patients is 5.18 ± 0.80 years. The majority of patients received treatment through the use of universal coverage (66.67%) and were primarily cared for by their parents (83.33%). The average income is $19,313.92 \pm 21,571.5$ Baht. (Table 1)

Table 1 The general information and medical treatment of cleft lip-palate patient aged between 4 to 6 years (n=12)

General information and medical treatment	number/percentage (%)
Genders (cases)	
male	5 (41.67)
female	7 (58.33)
average age (years)	5.18 ± 0.80
Types of cleft lip-cleft palate (cases)	
unilateral cleft lip and cleft palate	6 (50)
bilateral cleft lip and cleft palate	6 (50)
Palate surgery (cases)	
completed palate surgery	12 (100)
Caregivers (people)	
parents	10 (83.33)
grandfather/grandmother	2 (16.67)
Health benefit schemes	
universal Coverage	8 (66.67)
civil servant medical benefit scheme/others	4 (33.33)
average income (baht)	$19,313.92 \pm 21,571.5$
Medical care provided by	
pediatrician (cases)	9 (75)
breastfeeding	4 (33.33)
otolaryngologist	12 (100)
speech specialist	12 (100)
dental treatment	12 (100)

There are no differences in degree of agreement in the nose evaluation using Cronbach's alpha given by the experienced and inexperienced professional.

Regarding the esthetic of nose, the experienced professional showed a high degree of reliability (0.88) and it is significant ($p < 0.01$). The inexperienced professional showed a high degree of reliability in the lip and nose assessment (0.76 - 0.88).

Experienced evaluators gave a good score in the lip, nose, and NR and inexperienced evaluators gave a good score only in the nose and indicated fair score in the lip and nasolabial region. (Table 2)

Table 2 Cronbachs' alpha test: verification of the degree of agreement and esthetic evaluation by experienced and inexperienced evaluators

region	mean±SD	Cronbach's alpha coefficient	interpretation	p-value
lip ex	2.40±0.91	0.63	Good	0.11
lip inex	2.77±0.81	0.88	Fair	<0.01*
nose ex	2.50±0.83	0.88	Good	0.07
nose inex	2.47±0.85	0.76	Good	0.18
nasolabial ex	2.39±0.79	0.54	Good	0.43
nasolabial inex	2.85±0.80	0.68	Fair	0.08

remark: "ex" refers to experienced evaluators

"inex" refers to inexperienced evaluators

For the esthetic of lip and nasolabial region, it was found that there are statistically significant differences ($p = 0.04$ and 0.02 , respectively) and the

experienced evaluators gave a high score more than inexperienced evaluators. (Table 3).

Table 3 Verification of the differences between experienced and inexperienced professional evaluators for the lip, nose, and nasolabial region.

variable pair	n	mean±SD	minimum	maximum	interquartile	p-value
lip ex	48	2.39±1.12	1	5	2-3	0.04*
lip inex	48	2.77±0.80	2	5	2-3	
nose ex	48	2.5±0.92	1	4	2-3	0.60
nose inex	48	2.46±0.90	1	4	2-3	
NR ex	48	2.39±0.91	1	4	2-3	0.02*
NR inex	48	2.85±0.89	1	5	2-3	

remark: "ex" refers to experienced evaluators

"inex" refers to inexperienced evaluators

In terms of parental satisfaction with QoL of children with CLP, five aspects were investigated. The results showed that the overall satisfaction score of medical treatment was high. Similarly, satisfaction with services and medical costs was also high.

However, satisfaction regarding social and psychological aspects was at moderate level. Regarding the impact on families, the results showed a high level of agreement across all items. (Table 4).

Table 4 The satisfaction of parents based on 4 aspects and the impact on patient's families (n=12)

quality of life	mean±SD	interpretation
1. medical treatment	4.07 ± 0.89	high
2. service at the center	4.45 ± 0.50	high
3. medical and additional costs	3.72 ± 0.94	high
4. parental satisfaction	3.34 ± 0.92	moderate
5. the effects of CLP on the Family	3.76 ± 1.21	high

Discussion

The present study demonstrates the facial appearance in 3 parts which are lip, nose, and NR by experienced and inexperienced medical professionals. It was found that there were no differences in degree of agreement in the ratings of two groups. For the esthetic of lip and NR, the experienced professionals showed higher rating scale than inexperienced professionals but there are no differences in term of nose. Additionally, experienced evaluators indicated good scores in the lip, nose, and NR while inexperienced evaluators indicated good scores only in the nose and fair scores in the lip and NR.

In our study of facial esthetics in patients with CLP, the results demonstrated high reliability in terms of lip and nose evaluation and experienced or inexperienced evaluators showed high agreement in lip and NR evaluation. However, according to Alley and Hildebrandt¹⁷, human judgement can serve as a reliable tool in the assessment of facial attractive-ness.

We adopted the rating scale of Asher-McDade et al¹⁵, for the analysis of standardized clinical photographs, and the high interrater agreement confirmed the applicability of this classical scale. However, Power and Matic¹⁸, using the same scale as Asher-McDade et al¹⁶, reported poor agreement

among experienced surgeons in their esthetic evaluations of the lip. This suggests that discrepancies in relation to other published studies may reflect personal bias when a surgeon evaluates the postoperative results of another surgeon.

Nollet et al.¹⁹ stated that the average results may be used if the coherence between the components of the individual groups is substantial. For this reason, in this comparative study between groups of experienced and inexperienced professional evaluators, we used the average of each group's esthetic analysis of the lip, nose, and nasolabial region.

In our study, we found no differences in the esthetic analysis of nose between professionals experienced and inexperienced in the treatment of cleft lip and palate, however, in assessing the lip and NR the experienced professionals showed higher satisfaction with the esthetic results than the inexperienced professionals.

Some authors²⁰⁻²² also reported that experienced practitioners showed higher satisfaction with the appearance of the NR than lay assessors. However, there are studies that have reported opposite results, with the lay assessors giving higher attractiveness ratings than the experienced professionals^{23,24}. Furthermore, some studies have shown no differences in ratings between professionals and lay assessors^{25,26}.

Mosmuller et al²⁷. compared the Asher-McDade Aesthetic Index to a numerical scale (0 to 200) developed by Prahl et al²⁸., and its simplified 5-point scale version. Although three or more observers could use all three-scoring systems reliably, the Asher-McDade system was superior with an overall estimated reliability, with the average score of three observers of 0.8927. This is similar to the overall estimate reliability of our scoring system for both experienced and inexperienced evaluators.

The inconsistencies of these findings could be a result of methodological differences. Experienced evaluators might be more critical as they are trained to critically examine faces and notice fine differences in the degree of impairment. Many inexperienced are not familiar with CLP, and their own perception of residual deformity may influence their assessment negatively²⁹. In the present study, no significant difference was found in the way experienced versus inexperienced assessed the lip and NR.

In terms of socio-emotional well-being, it was found that the parents are satisfied with a low level of anxiety regarding illness and speech outcome. Feeling worried or anxious about illness or appearance may lead to the interruption of social activities. Concerns about others' perceptions, experiences of teasing, bullying, or being called names by other children are additional factors contributing to anxiety.

Similarly, another study revealed that, besides the functional aspect, socio-emotional factors play a key role in promoting QoL in these children³⁰. Additionally, these psychological problems may be interconnected. Anxiety and depression were reported about twice as often by subjects with CLP compared to controls. These psychological issues were strongly associated with concerns about appearance, dentition, speech, and the desire for further treatment³¹. Speech problems is one of the serious problems in CLP patients that could be described with hypernasality and unintelligible voice, which invariably results in a fear of speaking and communicating. As vocal resonance is indispensable for the comprehensive

rehabilitation of patients with CLP, its impairment contributes to social isolation²⁰. According to a study by Gkantidis et al.,³² as reported by patients and their parents, speech problem was one of the key factors that decreased satisfaction and was correlated with the increased effect of the cleft in everyday life³³. Furthermore, according to an investigation by de Buys Roessingh et al³⁰, the assessment of speech after palate surgery resulted in less than 50% of socially acceptable speech. Therefore, speech therapy is a common and necessary treatment for patients with cleft palate. The lack of clarity in their speech may discourage them from engaging in communication with others.

Strength and limitation

This study demonstrates the concept of facial appearance evaluation conducted by both experienced and inexperienced medical professionals. The results are proved by the reliability of medical professionals who are intensive in the field of cleft. The reliable outcome we received from the medical professionals will lead to improvement and develop the cleft care treatment. In the term of QoL, parents are concerned about anxiety regarding illness, especially speech outcomes can affect living society for cleft patients. Additionally, the study conducted on QoL in five domains is rarely observed, so it is beneficial to study in the field of cleft patients aged 4-6 years.

However, the sample size for this study is limited to only 12 patients which might not represent the whole population, but the selected sample is also applicable as they are homogeneous group.

Conclusions

The evaluation of facial appearance by experienced and inexperienced evaluators reveals different perspectives on aesthetics. In terms of QoL evaluation, caregivers indicated a good score, and the impact on the family appears to be minimal, indicating a relatively good overall quality of life. However, the

findings from this study should be used to further improve patient care in the future.

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