Direct Assessment of Nose and Upper Lip Morphology after Presurgical Nasoalveolar Molding in Complete Unilateral Cleft Lip and Palate Patients

Montian Manosudprasit DDS, MDS, FRCDT*,
Aggasit Manosudprasit DDS, DScD*, Siriya Pattarasiriporn DDS*

** Department of Orthodontics, Faculty of Dentistry, Khon Kaen University, Khon Kaen, Thailand*

Objective: *To evaluate the changes of the nose and upper lip morphology at two weeks after using presurgical nasoalveolar molding (PNAM) in unilateral cleft lip and palate (UCLP) patients.*
Material and Method: *Twelve infants with UCLP were treated with Khon Kaen University PNAM (KKU PNAM). Direct measurements of the nose and lip were recorded at initial visit (T1) and two weeks after KKU PNAM (T2). Paired t-test was used to compare statistically between T1 and T2.*
Results: *Two weeks after the treatment, significant changes were observed in the measurements at T2 compared to T1 ($p<0.05$). There was a significant increase in the columella length and the height of the nostril. Columella deviation and nostril width were significantly reduced. Moreover, the soft tissue gap, vermillion gap and lip protrusion were also significantly decreased.*
Conclusion: *These nose and lip morphology of the patients with complete unilateral cleft lips and palate were improved by the lip strapping and nasal molding device of KKU PNAM which possibly could help towards improvement of the surgical outcomes of cheiloplasty and nasal surgery.*

Keywords: *Unilateral cleft lip and palate, Presurgical nasoalveolar molding, Direct assessment*

J Med Assoc Thai 2017; 100 (Suppl. 6): S61-S67
Full text. e-Journal: *http://www.jmatonline.com*

The clinical features in unilateral cleft lip and palate are nasal asymmetry and deviation of the alveolar segments with the greater segment deviating laterally and the lesser segment deviating medially. The nasal asymmetry presents as a deformity of lower alar cartilage, missing nasal floor and shortened columella, flat and long alar, depressed alar base, deviation of the nasal septum and downward the tip of nostril[1,2].

Presurgical infant orthopedic therapy was first described by Hoffman in 1686, in which an extraoral anchored headcap extended arms to the face, was used to retract the premaxilla and narrow the cleft[3]. Grayson et al[4] introduced the definition of presurgical nasoalveolar molding (PNAM), which is a nonsurgical method to reshape the alveolar segments and nasal tissues prior to surgery. In 1993, he developed the Grayson's PNAM, consisting of a nasal stent extending from the anterior flange of an intraoral molding appliance[5]. However, this appliance is bulky when inserting in the mouth and it may cause feeding difficulty in a breast-fed baby. Monasterio et al[6] utilized the commercially available PNAM, including DynaCleft® lip-strapping device and nasal elevator. They claimed that this device was able to produce results similar to those of Grayson's PNAM without the need to take an impression from the infant. In 2012, Khon Kaen University Cleft Lip and Palate Center modified PNAM from Monasterio et al[6] and Doruk and Kilic[7] to create the Khon Kaen University PNAM appliance (KKU PNAM). To date, no evaluation has been made regarding the efficacy of this device. The purpose of this study was to evaluate the effectiveness of KKU PNAM in treating unilateral cleft lip and palate (UCLP) patients by measuring the changes of nose and upper lip morphology.

Correspondence to:
Manosudprasit M, Department of Orthodontics, Faculty of Dentistry, Khon Kaen University, Muang, Khon Kaen, Thailand, 40002.
Phone: +66-94-2914715, Fax: +66-43-202863
E-mail: monman@kku.ac.th

Material and Method

Study population

Twelve infants (three boys and nine girls) with complete UCLP attending the Tawanchai Cleft Center

of the Faculty of Medicine, Khon Kaen University were included in this present study. Patients with systemic diseases, general disabilities, craniofacial or other syndromes were excluded from the study. Patients who had received cheiloplasty and closed to normal position of lip, nose and alveolar cleft gap ≤2 mm were also neglected. The average age of the patients when starting KKU PNAM therapy was 27 days (ranged 5 to 60 days) (Table 1).

Treatment protocol of the study

This present study followed the treatment protocol of Manosudprasit et al[8], comprising:

Visit 1

History taking, clinical examination, diagnosis and treatment plan.

Advising treatment process to parents and signed the consent form.

Taking records at initial visit including:

1) Measurement the morphology of the nose and upper lip and re-measurements at least 30 minutes after the first examination. The two repeated measurements were averaged (T1), and

2) Taking impression for study model.

Applying lip strapping and forehead type of nasal molding device (Fig. 1).

Providing a lip strapping kit to parents, giving an advice on oral hygiene care, and how to use and take care of devices.

Visit 2

After two weeks, assessing treatment outcomes.

Taking records, including:

1) Measurements the morphology of the nose and upper lip and re-measurements at least 30 minutes after the first examination. The two repeated measurements were averaged (T2).

2) Taking impression for the study model.

Data collection and measurements

Direct measurements of the nose and upper lip were done with Boley gauge (the minimum length measurement recorded is 0.5 millimeters) at the initial visit (T1) and two weeks after using the KKU PNAM (T2) in order to evaluate the changes of the nose and upper lip morphology. Of note, the patients were not awoken while under examination. Re-measurements were performed at least 30 minutes after the first examination by the same examiner, in the same visit.

Table 1. Demographics data

Demographic data	Complete UCLP patients (n = 12)
Gender, n (%)	
Males	3 (25)
Females	9 (75)
Age at the start of treatment, range (mean ± SD) (days)	5 to 60 (27.4±18.4)

Fig. 1 Neonate with the KKU PNAM in position.

The measurements included the alar base width (ABW), alar base deviation (ABD), nostril width on the cleft side (NWC), nostril height on the cleft side (NHC), columella length (CL), columella deviation (CD), nose protrusion from subnasale (NPS), soft tissue gap (SG), vermillion gap (VG) and upper lip protrusion from subnasale (UPS) (Table 2, Fig. 2).

Statistical analysis

SPSS Version 16® (Statistical Package for Social Sciences for Windows) was used in data analysis. The changes of the nose and upper lip morphology between T1 and T2 were presented as means and

Table 2. The definitions of the measurements of the nose and upper lip

Measurement (mm)	Definition
ABW	The measurement between the most infero-lateral points of the alar.
ABD	The differences between vertical levels of the right and left alars.
NWC	The direct distance between the most medial nostril margin and the most lateral nostril margin on the cleft side.
NHC	The distance from the midpoint on the inner rim of the nostril perpendicular to the nostril width line on the cleft side.
CL	The distance between the midline base of columella and the most antero-superior of the columella.
CD	The distance from the subnasale perpendicular to the facial midline.
NPS	The measurement from the subnasale perpendicular to the vertical line that passes through the pronasale.
SG	The distance of the most medial point to the most lateral point on the cleft side.
VG	The distance of the most medial point on the vermilion border to the most lateral point on the vermilion border on the cleft side.
UPS	The measurement from the subnasale perpendicular to the vertical line that passes through the labrale anterius.

Fig. 2 Location of the nose and upper lip landmarks and measurement lines.

standard deviations (SD). The paired t-test was used to evaluate changes in the measurement lines. All *p*-values were two-tailed with 95% confidence intervals. Intraclass correlation coefficient (ICC) was calculated to assess the intra-examiner reliability that was done for all patients with a re-measurement at least 30 minutes after the first measurement by the same examiner, at the same visit. The intraclass correlation coefficient value in direct measurement must be at least 0.8.

This research project was approved by the Ethics Committee on Human Research, Khon Kaen University (HE592130).

Results

Each measurement was made directly on the infant's lip and nose at the initial visit (T1) and at two weeks of the treatment (T2) with the KKU PNAM (Fig. 3). Substantial reproducibility was found with ICC value in a range of 0.8 to 1.

The changes of nose morphology

Changes in the nose morphology measurements from T1 to T2 are shown in Table 3.

The changes of lip morphology

Changes in upper lip morphology measurements from T1 to T2 are displayed in Table 4.

Discussion

The primary purpose of the PNAM appliance is to reduce the width of the cleft deformity. In turn, the lip and nasal segments become better aligned and improved in symmetry, with an increase in tissue, and reduction in the severity of the lip, nose and alveolar

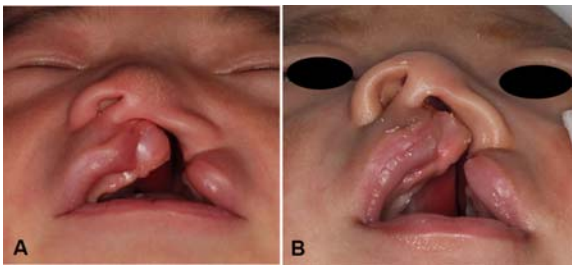

Fig. 3 Example of a patient treated with the KKU NAM (A) Before the treatment at the age of 7 days, (B) At 2 weeks after the KKU PNAM of the age 21 days.

clefts[3]. Berggen et al[9] and followed-up by Abdiu et al[10] developed the nasal alar elevator with lip tape, which they have used since 1996 and Monasterio et al[6], who reported the use of nasal elevator with elastic connection to a plastic base taped to the baby's forehead together with DynaCleft® labial strapping and without intra-oral plate. In a clinical trial, Monasterio et al. found that their treatment results compared closely with their use of the Grayson PNAM. They claimed the simplicity of their appliance with benefits over Grayson's PNAM, including being less invasive because of no need to produce an expensive custom-made molding appliance, easy handling for parents and no need for a dental specialist. Inspiring by these appliances, the KKU PNAM was created. The difference is that the KKU PNAM was applied to both nostrils to better control of columella shape and reduced over correction of septal deviation from a one-sided nasal elevator.

Our study included 12 patients with unilateral cleft lip and palate (UCLP). The average age at which the PNAM therapy was started was 27 days (ranged 5 to 60 days). This is still in the ideal cartilage molding period of the first 6 weeks of life, as determined by Matsuo and Hirose[11]. The range of starting age was variable since the patient referrals to Khon Kaen University Cleft Lip and Palate Center from other centers was often slow.

Each measurement was made directly on the infant's lips and nose at the initial visit (T1) and at the end of treatment (T2) with the KKU PNAM. The change of alar base width and alar base deviation between T1 and T2 was maintained which could be attributed to natural growth. This agreed with Ezzat et al[12] who analyzed 12 casts before and after using Grayson PNAM for patients with UCLP using stereoscopic imaging. They found a slight increase in columella base width but the result, although indicating statistical

Table 3. Measurements of differences of the nose morphology between pre- (T1) and 2 weeks after-(T2) KKU PNAM therapy for the 12 participating subjects

Nose	Mean±SD(mm)	Range(mm)	95% CI(mm)	Change:Increased/ Decreased	*p*-value
Alar base width (ABW)	0.00±0.21	-0.5 – 0.5	-0.14 – 0.14	No change	1.000
Alar base deviation (ABD)	-0.17±0.27	-0.75 – 0.0	-0.34 – 0.00	No change	0.06
Nostril width on the cleft side (NWC)	-0.33±0.43	-1.5 – 0.0	-0.61 – -0.06	Decreased	0.01*
Nostril height on the cleft side (NHC)	1.06±1.14	0.0 – 4.0	0.34 – 1.79	Increased	0.011*
Columella length (CL)	0.69±0.51	0.0 – 1.25	0.36 – 1.01	Increased	0.007*
Columella deviation (CD)	-0.48±0.43	-1.25 – 0.0	-0.75 – -0.20	Decreased	0.003*
Nose protrusion from subnasale (NPS)	0.00±0.11	-0.25 –0.25	-0.07 – 0.07	No change	1.000

*indicates a statistically significant difference ($p < 0.05$)

Table 4. Measurements of differences of the upper lip morphology between pre- (T1) and 2 weeks after-(T2) KKU PNAM therapy for 12 participating subjects

Upper lip	Mean±SD (mm)	Range (mm)	95% CI (mm)	Change:Increased/ Decreased	*p*-value
Soft tissue gap (SG)	-1.79±0.94	-3.0 to 0.0	-2.39 to -1.19	Decreased	0.000*
Vermillion gap (VG)	-2.00±0.86	-3.5 to -0.75	-2.55 to -1.45	Decreased	0.000*
Upper lip protrusion from subnasale (UPS)	-0.71±0.69	-2.0 to 0.0	-1.15 to -0.27	Decreased	0.004*

*indicates a statistically significant difference ($p<0.01$)

significance, was small and not clinically important. Singh et al[13] reported no significant change in alar base width when utilizing digital 3D stereophotogrammetry to evaluate changes in nasal morphology in 10 UCLP patients undergoing Grayson PNAM therapy. The change of nostril width on the cleft side in the present study was significantly decreased when treated with the KKU PNAM. This result concurs with many publications[6,9,12-16] using different types of PNAM. On the contrary, Mishra et al[17], studying a mixed group of 23 unilateral and bilateral cleft lip and palate patients, reported no significant differences in nostril width after Grayson PNAM therapy but no strapping was used in that study and no intraoral contraction screw plate. The finding of nostril height increase on the cleft side in the present study agrees with other studies using Dynacleft with nasal elevator[6] and Grayson PNAM[1,12,15,16,18]. The change of columella length and columella deviation were significantly improved. The columella length was increased (T1-T2), while columella deviation was decreased. This concurs with Liou et al., 2004[15] who studied 25 Taiwanese infants with UCLP treated with Grayson PNAM. The finding showed a significant increase in columella length compared to before the PNAM treatment. Berggen et al[9] and Monasterio et al[6] reported similar findings on the improvement of the columella deviation, but their measurements revealed changes in the angle not the length of the deviation. The nose protrusion (from subnasale to vertical tangent to nose tip) after using the KKU PNAM was maintained between T1 and T2. This agrees with Singh et al[13] and Gomez et al[18] who observed no significant changes at the tip of the nose (Prn-Sn).

The treatment with the KKU PNAM also produced a significant decrease in the soft tissue lip gap from T1 to T2. This result agrees with previous findings using PNAM with molding plate from Ezzat et al[12] and Kecik and Enacar[14] who found a significant reduction in cleft width. Interestingly, Monasterio et al[6] also found a significant reduction in cleft width, even though no molding plate was used in their PNAM system.

The PNAM technique is claimed to correct deviated alveolar segments by improving the arch form and nasal symmetry before cheiloplasty. Treatment objectives of PNAM technique are to correct or reduce the nasal deformity (nasal cartilage, nasal tip projection, alar base, position of columella and philtrum), elongate the columella, align and approximate alveolar segments[4]. Furthermore, the other objectives of PNAM technique are to facilitate surgical closure with lessening of surgical scar[3], reduce the likelihood of further nasal surgery, reduce the need for secondary alveolar bone grafts if gingivoperiosteoplasty (closure of the alveolar cleft) is included in the treatment[19,20], close oronasal fistulae and reduce labial deformities.

Conclusion

Measurement changes between at starting and 2 weeks after the treatment with KKU PNAM in the patient with cleft lips and palate indicated some improvement in the nasal and lip morphology. These nasal changes in conjunction with the reduction of the alveolar segments obtained by the lip strapping and nasal molding device of the KKU PNAM, lead to improved and favorable nose and upper lip morphology which possibly could help towards improvement of the surgical outcomes of cheiloplasty and nasal surgery. However, the present study only demonstrated short-term nose and lip results. More studies are still needed to evaluate long-term controlled clinical trials outcomes after the PNAM treatment and to determine the number of nose and lip revisions.

What is already known on this topic?

Several studies[1,5,6,9,10,12,13,21] have been reported on assessments of the effects of various types

of nasal-alveolar molding appliance. To date, no evaluation has been made regarding the efficacy of the Khon Kaen University PNAM appliance (KKU PNAM).

What does this study add?

The present study was to evaluate the effectiveness of the KKU PNAM in treating UCLP patients by measuring the changes of the nose and upper lip morphology, with the aim to facilitate cheiloplasty and nasal surgery and enhanced the clinical outcomes.

Remark

These patient's parents gave permission by signing the consent form for the use of all clinical photographs in this report for publication.

Acknowledgements

The author would like to express greatest appreciation and sincere gratitude to Associate Professor Keith Godfrey for his valuable advices. This research was supported by Faculty of dentistry, Khon Kaen University and the Center of Cleft Lip-Cleft Palate and Craniofacial Deformities, Khon Kaen University under Tawanchai Royal Grant Project.

Potential conflicts of interest

None.

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การประเมินรูปร่างจมูกและริมฝีปากภายหลังการปรับแต่งจมูกและสันเหงือกก่อนการทำศัลยกรรมในผู้ป่วยปากแหว่งเพดานโหว่ด้านเดียวสมบูรณ์ ด้วยการวัดโดยตรง

มนเทียร มโนสุดประสิทธิ์, เอกสิทธิ์ มโนสุดประสิทธิ์, สิริญา ภัทรศิริพร

วัตถุประสงค์: *เพื่อประเมินการเปลี่ยนแปลงของรูปร่างจมูกและริมฝีปากบน 2 สัปดาห์ภายหลังการปรับแต่งจมูก และสันเหงือกก่อนการทำศัลยกรรมในผู้ป่วยปากแหว่งเพดานโหว่ด้านเดียวสมบูรณ์*

วัสดุและวิธีการ: *กลุ่มตัวอย่างประกอบด้วยผู้ป่วยปากแหว่งเพดานโหว่ข้างเดียวสมบูรณ์จำนวน 12 คน ที่ได้รับการรักษาด้วยการดัดแปลงเครื่องมือปรับแต่งจมูกและสันเหงือกก่อนการทำศัลยกรรมชนิดยึดบริเวณหน้าผากของ มหาวิทยาลัยขอนแก่น (KKU PNAM) ทำการวัดโดยตรงก่อนเริ่มการรักษา (T1) และหลังการรักษา 2 สัปดาห์ (T2) การวิเคราะห์ด้วยสถิติแพร์ แซมเปิล ที เทสต์ เพื่อเปรียบเทียบระหว่าง T1และ T2*

ผลการศึกษา: *2 สัปดาห์หลังจากการรักษา มีการเปลี่ยนแปลงอย่างมีนัยสำคัญทางสถิติในการวัดที่ T2 เมื่อเทียบกับ T1 ($p<0.05$) พบว่ามีการเพิ่มขึ้นอย่างมีนัยสำคัญทางสถิติของความยาวโคลัมเมลลาและความสูงรูจมูกด้านรอยแยก ขณะที่ ส่วนเบี่ยงเบนโคลัมเมลลาและความกว้างรูจมูกด้านรอยแยก มีค่าลดลงอย่างมีนัยสำคัญทางสถิติ นอกจากนี้ขนาดรอยแยกสันเหงือกบน ขนาดรอยแยกริมฝีปากบน และความยื่นริมฝีปากมีค่าลดลงอย่างมีนัยสำคัญทางสถิติ*

สรุป: *การเปลี่ยนแปลงรูปร่างจมูกและริมฝีปากได้รับการปรับปรุงโดยแถบคาดริมฝีปาก และเครื่องมือปรับแต่งปีกจมูกชนิดยึดบริเวณหน้าผากของมหาวิทยาลัยขอนแก่น ซึ่งอาจช่วยให้ผลของการทำศัลยกรรมตกแต่งริมฝีปากและจมูกดีขึ้น*