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Original Article

Modified presurgical nasoalveolar molding for patients with neonatal complete bilateral cleft lip and palate having a severely malpositioned premaxilla

Jiansuo Hao*, Quan Wan*, Jiayu Liu, Wenli Wu, Jiameng Liu, Chongdai Luo, Zijun Gao, Fan Li, Yiyang Chen, Zhiyuan Lu

*Department of Oral and Maxillofacial Surgery, Stomatology Medical Center, Guangzhou Women and Children's Medical Center, Guangzhou Medical University, Guangzhou, P. R. China. *Equal contributors.*

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Abstract: Objective: To develop a modified presurgical nasoalveolar molding (MPNAM) with a premaxillary appliance, connected with two stainless steel wires and evaluate its therapeutic efficacy in newborns with complete bilateral cleft lip and palate (BCLP). Method: A total of 41 patients with neonatal complete BCLP having a severely protruded and deviated premaxilla were retrospectively selected from January 2017 to November 2019. All patients received the MPNAM device with a premaxillary appliance which was worn until cheilorrhaphy. Plaster casts from pre- and post-MPNAM treatments were scanned using a three-dimensional laser scanner, and the changes were recorded. Facial photographs of patients were taken during the treatment. Result: The average MPNAM treatment duration was 59.8 days. In all cases, the protrusive and deviated premaxilla was rapidly retracted and set into a suitable position after MPNAM treatment. The relative deviation distance and alveolar cleft width were significantly reduced. Both mid-palatal arch width and posterior arch width were increased. In addition, none of the patients developed any complications during the MPNAM treatment. Conclusion: Our MPNAM device was able to rapidly centralize the malpositioned premaxilla and reduce the alveolar cleft defect. This device can be applied in presurgical orthodontic treatments for patients with complete BCLP having a severely protruded and deviated premaxilla.

Keywords: Bilateral cleft lip and palate, malpositioned premaxilla, morphometric analysis, nasoalveolar molding, presurgical orthopedic treatment

Introduction

Cleft lip and palate is a congenital malformation of maxillofacial development in humans, which seriously affects a child's facial appearance and phonic function, even causing psychological problems [1, 2]. Complete bilateral cleft lip and palate (BCLP) is the most complex cleft lip deformity, which requires interdisciplinary cleft team management. A protruded premaxilla and columellar deficiency are common features in patients with BCLP [3, 4]. This significantly alters the facial form and structure, which increase the difficulty of surgery with respect to facial esthetic outcomes compared to unilateral clefts. In these circumstances, the retraction of the premaxilla and the restoration of a satisfactory facial form are especially crucial.

Although great advances in reconstructive surgery have significantly improved the quality of repair for clefts of the lip, alveolus, and palate, a single surgical technique cannot correct a protruding premaxilla [5]. Therefore, many presurgical maxillary orthodontic treatments have been used in the past to align the premaxillary segment and obtain the desired clinical result. Presurgical nasoalveolar molding (PNAM) is a preferred presurgical orthopedic treatment to correct the deformed nasal cartilage and reduce the cleft gap [6, 7]. The therapy significantly reduces the surgical difficulty of reconstruction, and it has been widely used in many hospitals in China [8]. However, using a standardized PNAM approach for patients with BCLP having a severely malpositioned premaxilla makes it particularly difficult to accomplish the midline correction of the premaxilla [9].

Therefore, these cases usually require a second surgery to achieve acceptable nasoalveolar esthetics, leading to an increase in scar formation [10]. The centralization and retraction of the displaced premaxilla into a more anatomic position is essential to achieving a better facial aesthetic outcome for these cases.

In this study, we developed modified PNAM (MPNAM) with a small premaxillary appliance connected with two elastic chains for the restoration and centralization of a severely protruding/deviated premaxilla in patients with complete BCLP. This study retrospectively evaluated the clinical effects of MPNAM on alveolar segments.

Materials and methods

Patients

The study was carried out at the Department of Oral and Maxillofacial Surgery, Guangzhou Women and Children's Medical Center, Guangzhou, China from January 2017 and November 2019. The patients had been treated consecutively by a single surgeon. The inclusion criteria for the patients were as follows: (1) infants with complete BCLP and obviously deviated protuberant premaxilla, (2) infants who started MPNAM treatment within 1 month of birth and with a minimum of 3 kg body weight, (3) infants with no other craniofacial deformities and systemic disease, (4) infants with primary surgical interventions by the same operator, and (5) infants with available medical records and photographs obtained pre- and post-treatment. Patients with incomplete cleft lip and palate or syndrome were excluded from the study. A total of 13 girls and 28 boys who met the criteria were included in this study. The mean patient age was 19.6 days (range, 14-26 days) at the initiation of MPNAM. This retrospective study was approved by the Institutional Ethical Committee of Guangzhou Women and Children's Medical Center (2019-32700). Written informed consent was obtained from the parents of all patients participating in the study.

Modified PNAM treatment

After the initial clinical evaluation, impressions of the intraoral cleft were taken at the second visit in the outpatient clinic without anesthesia. The impression was taken under the condition wherein airway emergency was available. The infant was held in an inverted position to prevent aspiration of the impression material. The impression was made using silicone impression material (Silagum, DMG Medical Devices Co. Ltd., Hamburg, Germany) and placed into the oral cavity. The mouth was examined for residual impression material after the tray was removed. The impression was then poured in dental stone (Heraeus, Heraeus Kulzer Dental Ltd, Hanau, Germany) to obtain a working model cast and a study model cast.

The boundaries of the palate and premaxilla were marked following a careful evaluation of the plaster model. The nostril position was marked, and excess dental stone was removed. The cleft region and the undercut areas were filled with base plate wax (Shanghai Medical Instruments Ltd., Shanghai, China) to approximate the contour. The working cast was then fabricated with auto-polymerizing acrylic resin (GC Corporation, Tokyo, Japan) to cover the alveolar ridge and the palate. It was extended outward to the vestibular folds and backward to the maxillary tuberosity and the trailing edge of the hard palate. After the resin was cured, the molding plate was sawed along both the alveolar ridges that separated the premaxillary part from the remaining molding plate. The anterior part was called the premaxillary appliance and the remaining part was called the palatal plate. Two stainless steel wires (Shanghai Medical Instruments Ltd.) with a 0.5 mm round tip were used to connect the premaxillary appliance and the anterior and middle of the palatal plate. Then two nasal stent wires made of 0.8-mm round stainless steel were embedded into both cleft sides of the palatal plate and emerged using acrylic resin. The extended aspect of the wire was bent to form a loop (1-1.5 mm in diameter) for activation and the tip of the wire was covered with soft silicone (Ufi-Gel P, Voco, Germany) to form a nasal bulb. The MPNAM device comprised two nasal stents: a premaxillary appliance and a palatal plate (**Figure 1**).

After fabrication was completed, the MPNAM device was thoroughly polished to ensure a smooth border to avoid trauma to the surrounding tissue. The MPNAM device was placed in the oral cavity and denture adhesive was used to retain the appliance in the mouth. The alveolar segments and nasal molding were molded

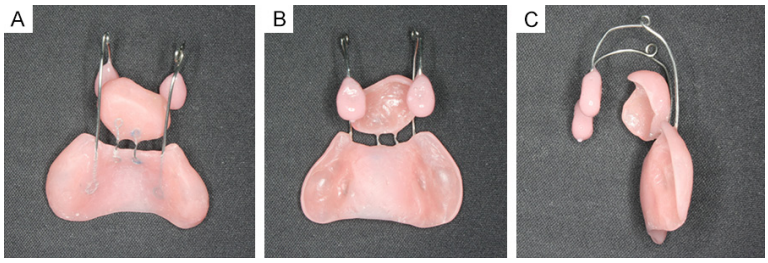

Figure 1. Photograph of modified PNAM device with the nasal segment attached to the molding plate. A. Frontal view. B. Dorsal view. C. Lateral view.

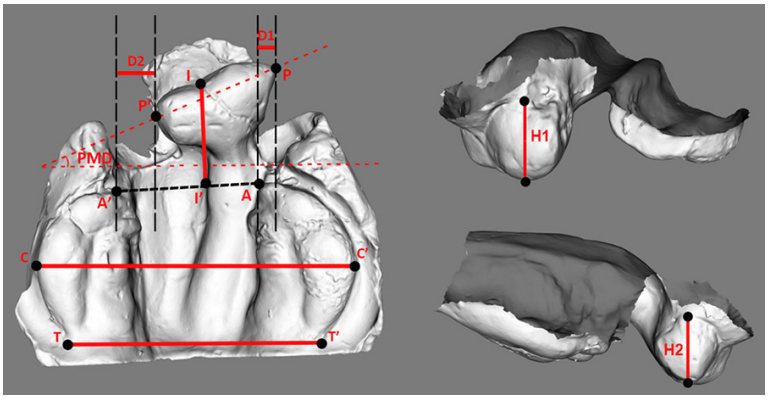

Figure 2. Anatomic landmarks of the maxillary cast. I, Incisal point; P, P', the right and left cleft edges of the premaxilla; A, A', the right and left cleft edges of the lateral segments; C, C', the cuspid points; T, T', the tuberosity points; I-I', the length of perpendicular line from I point to the AA' line; D1, the horizontal distance of deviated side in premaxilla and D2, the horizontal distance of offside in premaxilla, H1, the height of deviated side in premaxilla and H2, the height of offside in premaxilla; C-C', mid-the width of palatal arch; T-T', the width of posterior arch; premaxillary deviation (PMD), deviation of premaxillary segment.

simultaneously, and the nasal bulb was placed inside the cleft nostril. The curvature of the nasal stent wire was adjusted to ensure that the loop did not touch the lower lip. A horizontal adhesive tape (Micropore Surgical Tape; 3M, MN, USA) was placed from one side of the cheek, and then around below the loop of the wire to the other side. One more adhesive tape was placed over the premaxillary appliance to enhance the depressing force on the premaxilla. MPNAM was retained for 1-3 months, and patients needed to visit our clinic every 2 weeks for MPNAM device adjustment until lip repair.

Primary cheiloplasty

The MPNAM treatment was terminated when the alveolar cleft was reduced to less than 3 mm and the impression was taken at post-treatment. The mean age of patients post-MPANM treatment was 79 (range: 51-114) days. Cheiloplasty was performed by the same experienced surgeon in infants at an age of more than 3 months. Reconstructive surgery was performed to preserve the original length of the anterior lip.

Measurements

The casts of all patients were obtained before and after MPNAM treatment and scanned using a Carestream Intraoral Scanner (CS3600, Carestream Health, TX, US) to produce a 3D image. The landmark analysis was performed based on 3D images of casts using CS imaging software (Carestream Health, TX, USA). The landmarks on the maxillary casts are shown in **Figure 2**.

Statistical analysis

All statistical analyses were performed using SPSS version 22.0 (SPSS Inc., IL, USA). The changes in casts (pre-treatment and post-treatment) were analyzed using the two-tailed paired-sample *t* test, and P value < 0.05 was considered a significant difference.

Results

Patients treated with an MPNAM device

The appearance of the MPNAM device is shown in **Figure 1**, including its frontal (**Figure 1A**), dorsal (**Figure 1B**) and lateral view (**Figure 1C**). A total of 41 patients with complete BCLP having a severely deformed premaxilla treated with a MPNAM were included in this study. The average age of patients at the initiation of the MPNAM treatment was 19.6 days (range: 14-26 days). The average duration of MPNAM treatment was 59.8 days (range: 29-88 days). No complications were found in all patients during MPNAM treatment.

Table 1. Maxillary cast analysis

Measurement	Pre-treatment (Mean ± SD)	Post-treatment (Mean ± SD)	*P* value
D1 (Deviated side)	2.65 ± 0.49 (mm)	1.02 ± 0.69 (mm)	< 0.001
D2 (Offside)	4.17 ± 1.36 (mm)	1.48 ± 1.00 (mm)	< 0.001
I-I'	8.21 ± 2.69 (mm)	3.83 ± 1.61 (mm)	< 0.001
C-C'	40.56 ± 2.42 (mm)	41.96 ± 2.41 (mm)	< 0.001
T-T'	38.23 ± 2.03 (mm)	40.10 ± 1.88 (mm)	< 0.001
P-P' (Premaxilla width)	15.88 ± 0.65 (mm)	15.82 ± 0.67 (mm)	0.440
A-A' (Alveolar cleft width)	19.71 ± 0.75 (mm)	17.48 ± 1.09 (mm)	< 0.001
H1 (Deviated side)	9.83 ± 0.52 (mm)	9.81 ± 0.54 (mm)	0.568
H2 (Offside)	8.01 ± 0.74 (mm)	8.00 ± 0.68 (mm)	0.672
PMD	16.05 ± 5.48 (°)	3.86 ± 3.49 (°)	< 0.001

Note: PMD, premaxillary deviation.

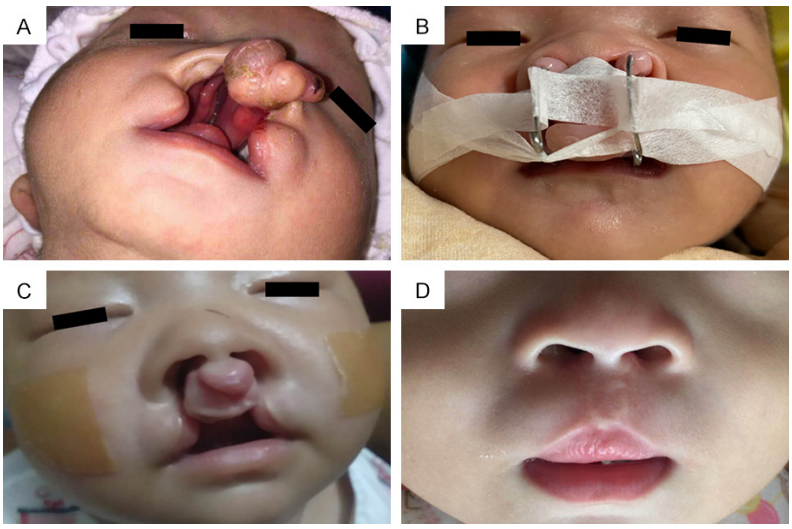

Figure 3. A female infant with a complete bilateral cleft lip and palate with severely displaced maxilla. The age at the start of MPNAM treatment was 21 days, and the treatment duration was 49 days. (A) Frontal view prior to MPNAM treatment. (B) Intermediate photograph with MPNAM and taping. (C) Frontal photograph of post MPNAM treatment and (D) 6 months after cheiloplasty. MPNAM, modified presurgical nasoalveolar molding.

Treatment outcomes

A change in the premaxilla and palatal cast measurements was clinically evident after MPNAM treatment. The landmarks on the maxillary casts are shown in **Figure 2**. A significant reduction was found in the distance of premaxillary protrusion (I-I') after treatment (8.21 ± 2.69 mm vs 3.83 ± 1.61 mm, P < 0.001). Both sides of the relative premaxillary horizontal travel distance (deviated side: D1 and offside: D2) were significantly decreased (pre-D1 vs post-D1, 2.65 ± 0.49 mm vs 1.02 ± 0.69 mm; pre-D2 vs post-D2, 4.17 ± 1.36 mm vs 1.48 ± 1.00 mm, P < 0.001). The width between the middle parts of alveolar segments (C-C') was increased (40.56 ± 2.42 mm *vs* 41.96 ± 2.41 mm, P < 0.001). Also, a significant increase in the posterior arch width (T-T') was found (38.23 ± 2.03 mm vs 40.10 ± 1.88 mm, P < 0.001). There was a significant decrease in alveolar cleft width (A-A') after MPNAM treatment (19.71 ± 0.75 mm *vs* 15.82 ± 0.67 mm, P < 0.001). In addition, the deviated premaxilla was derotated from 16.05 ± 5.48° to 3.86 ± 3.49° (P < 0.001). While there were no obvious changes in premaxilla width (P-P', 15.88 ± 0.65 mm vs 15.82 ± 0.67 mm) and both sides of premaxilla height (9.83 ± 0.52 mm vs 9.81 ± 0.54 mm; 8.01 ± 0.74 mm vs 8.00 ± 0.68 mm). The detailed results are shown in **Table 1**.

Case report

A typical case report is illustrated in **Figure 3**. A female infant had a complete BCLP with severely displaced maxilla (**Figure 3A**). The age at the start of MPNAM treatment was 21 days, and the treatment duration was 49 days in this case. The deviated premaxilla was retracted and set to a proper position by continuous MPNAM treatment from the frontal photographs (**Figure 3B** and **3C**) and no obvious deviation was observed on the philtrum midline after cheiloplasty (**Figure 3D**). **Figure 4** shows the 3D models of casts pre- and post- MPNAM treatment in this case (**Figure 4A** and **4B**). Significant changes were observed in the position of the maxilla and the width of the posterior dental arch by comparing their superimposition (**Figure 4C**).

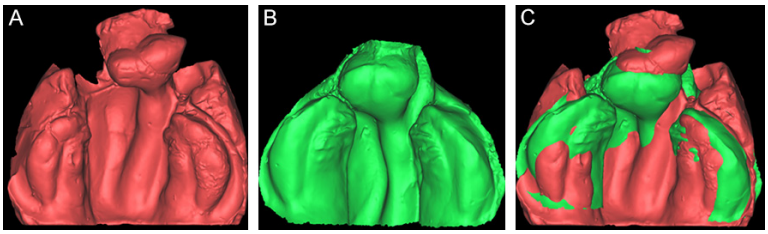

Figure 4. 3D scanned maxillary cast photographs. A. Pre-MPNAM treatment 3D model of scanned maxillary cast photograph. B. Post-MPNAM treatment 3D model of scanned maxillary cast photograph. C. Superimposition. MPNAM, modified presurgical nasoalveolar molding.

Discussion

Treatment of a complete BCLP deformity is challenging. Patients with complete BCLP often have obvious protrusion and severe dislocation of the premaxilla, resulting in severe disruption of the nasolabial structures. Thus, they require multidisciplinary treatments. PNAM has become one of the most common preoperative orthodontic treatment modalities. The advantages of PNAM, including a decrease in the width of the cleft, an improvement in the nasolabial symmetry, elongation of the columella, and facilitation of primary surgical repair, have been intensely studied. In many reports published to date, cleft lip and palate repair surgery is an effective way to improve the nasal cartilage deformity; however, it often leads to a flat nasal tip and short columella in patients with complete BCLP after surgical repair [11, 12]. The application of PNAM provides a new opportunity to reduce the nasal and alveolar deformity and the number of surgeries. Many scientific studies have reported the benefits of PNAM in correction of the cleft lip and nose deformity in patients with complete unilateral and bilateral clefts [13-15].

However, in certain cases, PNAM cannot provide the ideal result, particularly in patients with a severely protruding/deviated premaxilla. Ideally, a protruding/deviated premaxilla should be shifted to a suitable position prior to surgical correction so as to enhance the esthetic appearance of the face. To date, very few studies have been performed on difficult cases with a severely malpositioned premaxilla. In a study, Li et al. [8] developed a novel PNAM with a retraction screw that could rapidly correct nasal deformities and centralize the malpositioned premaxilla. Kiya et al. [16] described a novel appliance comprising premaxillary and palatine process plates connected with two elastic chains. They achieved reduced cleft widths and premaxillary protrusion within 1 month after the treatment. However, this appliance was invasive. Monga et al. [17] described the fabrication of a silicone cup-bonnet appliance and its successful use in two cases of BCLP.

In our study, we developed a modified device with the addition of a premaxillary appliance connected with the palate using two steel wires to align the cleft segments. Our MPNAM technique takes advantage of the transitory plasticity of the cartilage following birth, thus simultaneously improving the nasolabial symmetry and centralizing the deviation of the premaxilla in difficult cases. Therefore, we could perform one-stage cheiloplasty with low tension. Previous studies indicated that more beneficial outcomes could be achieved with PNAM if the treatment was started as early as possible after birth [18, 19]. A newborn has a high level of hyaluronic acid, which makes the cartilage highly flexible, vulnerable, and adaptable into a new position. The plasticity of the nasal cartilage lasts for 3-4 months and it decreases as the infant grows older. In our series, the average age for applying MPNAM device was 60 days (range: 29-88 days), which was close to the ideal cartilage-molding period in previous studies [20, 21]. During treatment, patients treated with MPNAM exhibited a significant improvement in nostril symmetry. The nasal columella was gradually prolonged, and the rotated premaxilla was gradually retracted and centralized to its normal anatomical position. It was also found that both sides of the alveolar gap were significantly reduced in all cases. As a result, the rotated premaxilla was retracted and centralized to the middle line, and the width of the alveolar gap was effectively reduced 2 months after the PNAM treatment so that primary cheiloplasty was carried out on time. This decreased the number of outpatient clinic visits before the surgery. This repositioning allowed for a less invasive surgery and improved the esthetics of the nasolabial complex after primary lip repair surgery.

Using the modified procedure, the soft acrylic resin premaxillary appliance was used to apply

force to rotate the premaxilla to its optimal position, and the denture adhesive was used to hold the appliance in position so that it was non-invasive to the soft tissue. Soft silicone in the nasal stents facilitated movement of the nasal alar cartilage toward the normal position.

Our modification had several advantages. This improvement reduced anesthetic exposure within our protocol. The direct contact appliance made of resin or silicone material was flexible and soft, which offered patient comfort. The modified PNAM technique significantly reduced the duration of active treatment in comparison to conventional PNAM treatment, which allowed for better compliance and cooperation by the parents/caretaker because the appliance should remain in the mouth for 24 h. Furthermore, this appliance was non-invasive for infants and simple to set up. Finally, the inner side of the MPNAM device had buffer space that was beneficial for palatal development, which was the reason why the width between the middle and posterior parts of the alveolar segments (C-C' and T-T') was increased after MPNAM treatment.

This study had several limitations, including its retrospective nature and small sample size limited to a single institution. This modified PNAM treatment required high levels of expertise and cooperation from families as well as weekly adjustments by the orthodontists. We started utilizing this modified appliance from November 2016; thus, we could not report its long-term results in this manuscript. Further investigation is needed to analyze the long-term effects, including the effect on maxillary growth, tooth formation, and nasal shape.

Conclusion

Our MPNAM is not only easy and atraumatic to apply but also time-effective by approximately 2 months. Further, it produces highly desirable outcomes. Moreover, it can simultaneously correct the nasolabial and palatal deformities, and also retract and centralize the deviated premaxilla in patients with BCLP. Therefore, our appliance could be used in patients with complete BCLP patients having a severely malpositioned premaxilla.

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We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us.

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Disclosure of conflict of interest

None.

Address correspondence to: Zhiyuan Lu, Department of Oral and Maxillofacial Surgery, Stomatology Medical Center, Guangzhou Women and Children's Medical Center, Guangzhou Medical University, Guangzhou 510632, Guangdong, P. R. China. E-mail: lzy890120@163.com

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