AJO-DO

Presurgical nasoalveolar molding with 3D printing for a patient with unilateral cleft lip, alveolus, and palate

Jie Zheng, Hong He, Wenying Kuang, and Wenjun Yuan
Wuhan, Hubei, China

An 8-day-old male infant with unilateral cleft lip, alveolus, and palate had a wide alveolar defect, soft tissue deformity, and a markedly sunken nasal wing at the cleft side. The patient was treated with a series of 3D-printed molding plates and synchronously with a nasal hook. The cleft edges moved closer by 9 mm at the alveolar ridge and the nasal wing was lifted considerably. Split-type 3D printing of presurgical nasoalveolar molding helped to reduce the cleft gap, improve the arch form, approximate lip segments, and distinctly improve the morphology of the nose by correcting the flattened nasal wings. (Am J Orthod Dentofacial Orthop 2019;156:412-9)

Clefts of the lip and palate (CLP) are common birth defects of complex etiology,[1] affecting 1 in 700 live births.[2] The nose, lips, and maxillary arch of the newborn are often severely distorted and asymmetric.[3] Treatment of CLP ideally involves a multidisciplinary team. Presurgical nasoalveolar molding (PNAM) was first described by Grayson et al[4] and is a widely accepted approach applied before cheiloplasty for infants with CLP. The treatment goals are to reduce the width of the cleft gap with reduction in soft tissue and cartilaginous deformity, to align and approximate the intraoral alveolar segments, and to allow a surgical repair with minimal tension.

Modification of PNAM is required to gradually approximate the alveolar segments and minimize the nasal deformity. The procedure requires weekly visits over a period of 2-3 months, which is time consuming, especially for patients who live in remote regions. In addition, manual grinding is an experience-based manipulation; the efficiency of treating patients with the use of the PNAM appliance has a positive correlation with the clinician's skills.[5] It is also worth mentioning that the integrated structure of the traditional PNAM appliance has some drawbacks. For example, force acting on the nose tip produced by the nasal stent will produce a reaction force, which goes against the retention of molding plate and makes the molding plate move downward. It is unfavorable for projecting the nasal tip; moreover, there exists a risk that the molding plate will be dislodged and obstruct the airway.[5]

Three-dimensional (3D) printing has been hailed as a disruptive technology that will change manufacturing.[6] New applications are being developed in orthodontics. For example, computed models can suggest stages between current and desired teeth positions, and aligners are designed and created precisely for each stage. Inspired by this application, we attempted to apply digital technology to presurgical orthopedics for infants. First, we modified the integrated structure of the traditional PNAM into a split type. Then we designed and printed the intraoral molding plates. To reduce the number of follow-up visits, we used 3D software and equipment to precisely compensate for the alveolar growth in each appliance.

The present case report describes the treatment of a neonate with unilateral CLP by molding the nasolabial morphology and alveolar arch with the use of 3D printing of the PNAM device.

DIAGNOSIS AND ETIOLOGY

An 8-day-old healthy male neonate unable to breast-feed and swallow because of lip and palatal

Department of Orthodontics, School and Hospital of Stomatology, State Key Laboratory Breeding Base of Basic Science of Stomatology, and Key Laboratory for Oral Biomedicine of Ministry of Education, Wuhan University, Wuhan, Hubei, People's Republic of China.
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Address correspondence to: Wenjun Yuan, Associate Chief Physician, Department of Orthodontics, School and Hospital of Stomatology, State Key Laboratory Breeding Base of Basic Science of Stomatology (Hubei-MOST), and Key Laboratory for Oral Biomedicine of Ministry of Education (KLOBM), Wuhan University, #237 Luoyu Road, Hongshan District, Wuhan 430079, People's Republic of China; e-mail, yuanwenjun@whu.edu.cn.
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defects was brought to the Department of Orthodontics, School and Hospital of Stomatology, Wuhan University, for consultation. Before treatment, a recent medical examination report including hemanalysis, cardiac color ultrasound, and chest radiography was provided by the patient's parents, which was not significant. History of infection, vitamin deficiencies, drug ingestion, chemical stimulus, and traumatic stress during pregnancy was absent.

Two- and 3-dimensional extra- and intraoral photographs were taken and information regarding the objectives and proposed treatment was provided to the parents, who gave consent for their son's treatment.

He had a markedly sunken nasal wing at the cleft side and alveolar segment displacement (Fig 1). The intraoral cleft gap was 9.4 mm. The defect was diagnosed as a nonsyndromic unilateral complete cleft of lip, alveolus and palate. A stepwise approach was planned for PNAM, followed by surgery.

TREATMENT OBJECTIVES

The treatment objectives were to reduce the width of the cleft gap, to close the intraoral alveolar segments and normalize the alveolar arch form, to approximate bilateral lip tissues and distinctly improve the morphology of the nose by correcting flattened nasal wings, and finally to allow a surgical repair with minimal tension.

TREATMENT ALTERNATIVES

The following options were established for the patient based on the principal objective of PNAM treatment: to reduce the severity of the initial cleft deformity.[5]

1. Application of the traditional alveolar molding plate in conjunction with a nasal stent[4] or a nasal hook. The typical method for traditional PNAM is modifying a single plate over time, which requires weekly follow-up visits.
2. Application of the split-type PNAM, which contains a series of 3D-printed molding plates and a nasal hook. The typical method for novel PNAM is using the alveolar molding plates sequentially. A nasal hook independent from the molding plate is used synchronously. A series of molding plates are printed and delivered to the parents at every visit. The follow-up visits are monthly. The neonate's parents chose this option.

TREATMENT PROGRESS

Impression making

After explaining the procedure and treatment goals to the parents, an impression was made. The infant was fully awake, without anesthesia. His head was gently held in a slightly supine position during the impression procedure. He was allowed to cry freely to ensure the adequate breathing during the impression-making procedure. The hospital setting allowed for rapid response by an airway team in the event of an emergency.

To quantitatively and objectively evaluate the alveolar molding effect, establishment of a stable 3D coordinate system was necessary. Frankfort horizontal plane (FH plane) was suggested as the reference plane to develop the dimensional numeric model, considering the fact that it is independent from the maxilla. Consequently, a simple face bow was designed and used for transferring the FH Plane to the tray (Fig 2, *A*). Impressions for the maxillary casts were taken in a special pediatric impression tray by an orthodontist using silicon rubber (GC Corporation, Tokyo, Japan) at the initial visit and after treatment. Care was taken to ensure that the material registered the border regions of the maxilla as well as the cleft region. When the material had fully polymerized, the impression was removed and inspected to ensure that all desired landmarks had been captured (Fig 2, *B*).

Impression scanning and molding plate making

The 3D models were acquired with the use of a laser scanning machine (3Shape, Copenhagen, Denmark; Fig 2, *C*). A computer-aided design (CAD) and reverse-engineering software system was applied to judge the digital geometric 3D model and formulate the treatment design (Rhinoceros 5; Robert McNeel & Associates, Seattle, Wash). The CAD molding plate procedure was divided into 8 steps by selective removal and sequential addition of resin, as shown in Figure 3. To direct the premaxilla of the greater segment inward, a 0.5 mm thickness of acrylic resin was added to the inner labial aspect of the premaxilla of the alveolus portion of the appliance, and an equal amount of resin was removed from the palatal aspect. To direct the lesser segment and the posterior alveolus of the greater segment outward from the cleft, acrylic resin was selectively removed from the inner labial aspect of the alveolus (~0.5 mm), and an equal amount of resin was added on the palatal aspect.

The standard tessellation language (STL) file was then exported from the planning software and imported into the 3D printing software to set up and complete the print. The molding plate was oriented to minimize

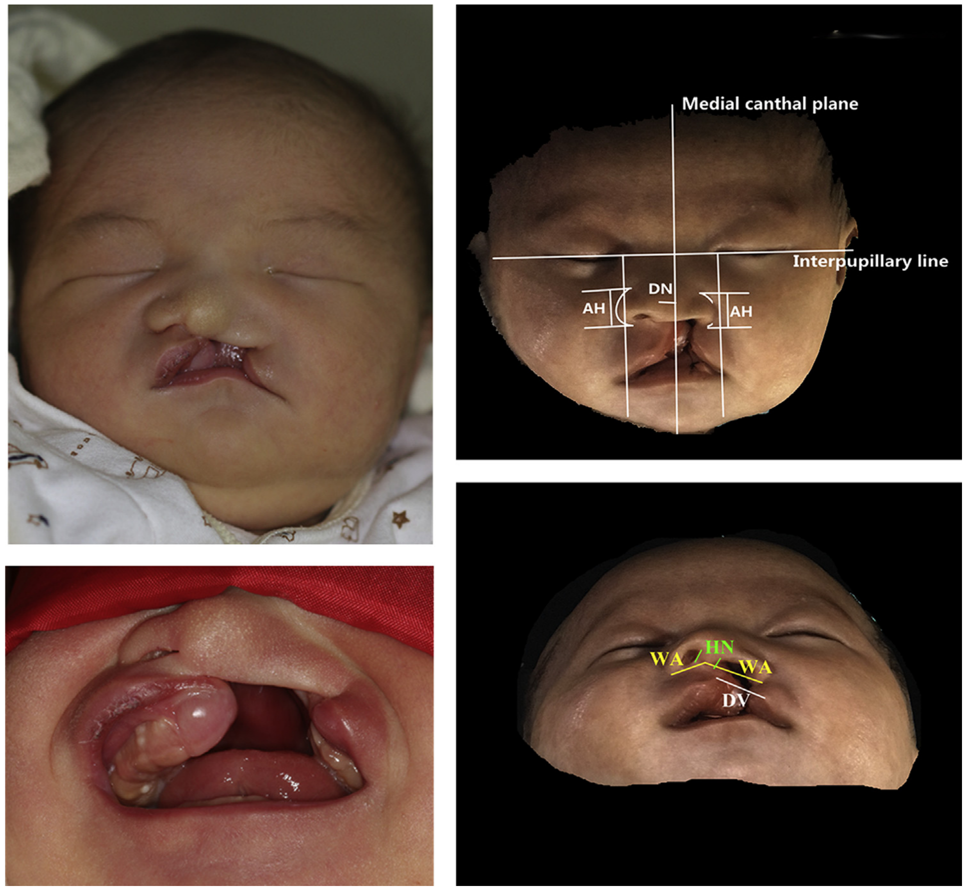

Fig 1. Extraoral 2D and 3D records before PNAM treatment. The images show an 8-day-old male infant with cleft lip, alveolus, and palate. Nasolabial measurement variables were marked as shown in the right figures.

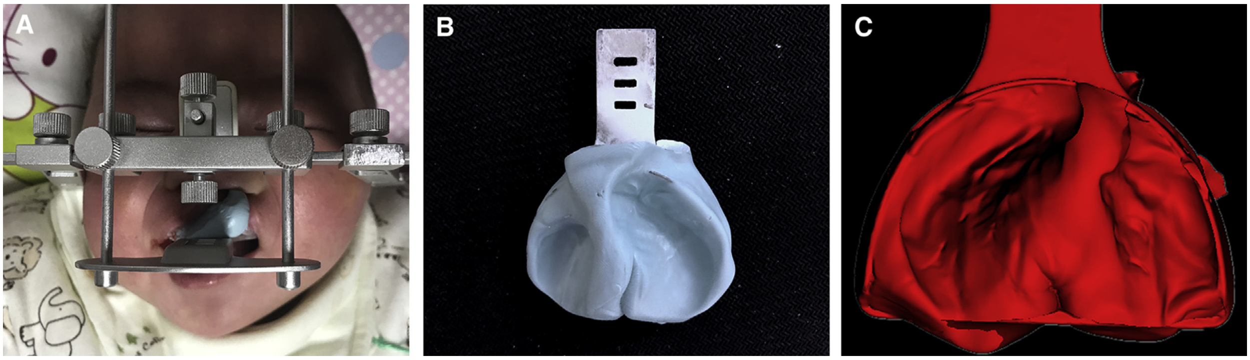

Fig 2. Impression making and scanning. **A,** A simple articulator was used to transfer the Frankfort plane. **B,** An impression was made with a silicon rubber. **C,** Impression scanning.

cross-sectional peeling forces during printing and to allow for the drainage of excess resin, and support points were added in areas that did not interfere with an accurate fit of the molding plate (Micro Plus 3D printer; EnvisionTEC, Gladbeck, Germany; Fig 4, *A*). The acrylic resin (Vertex-Dental, Soesterberg, The Netherlands) volume used was 5.65 mL, and the settings for the print were 20-mm layers in the *z*-axis with a print time of

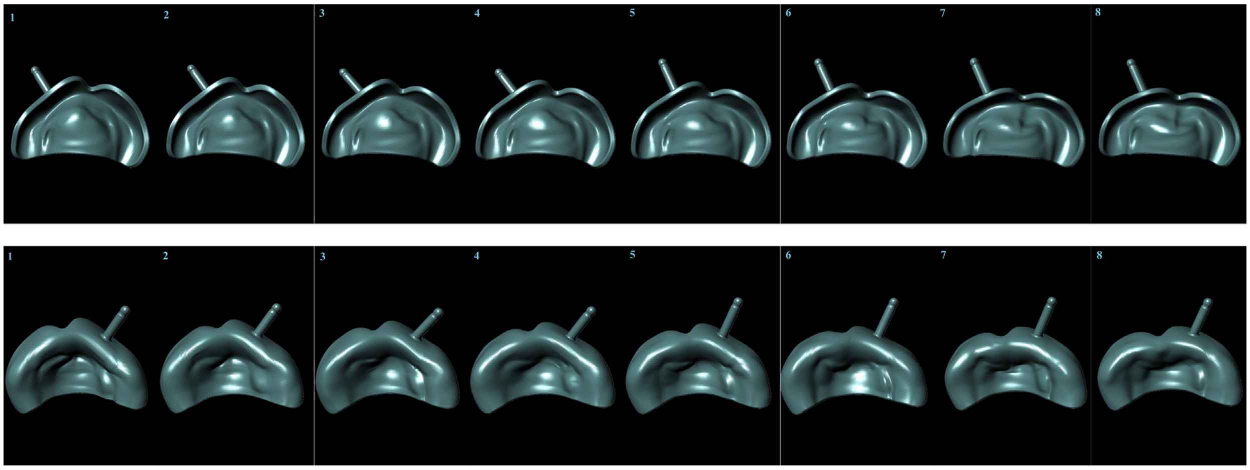

Fig 3. The CAD molding plate procedure was divided into 8 steps by selective removal and sequential addition of resin.

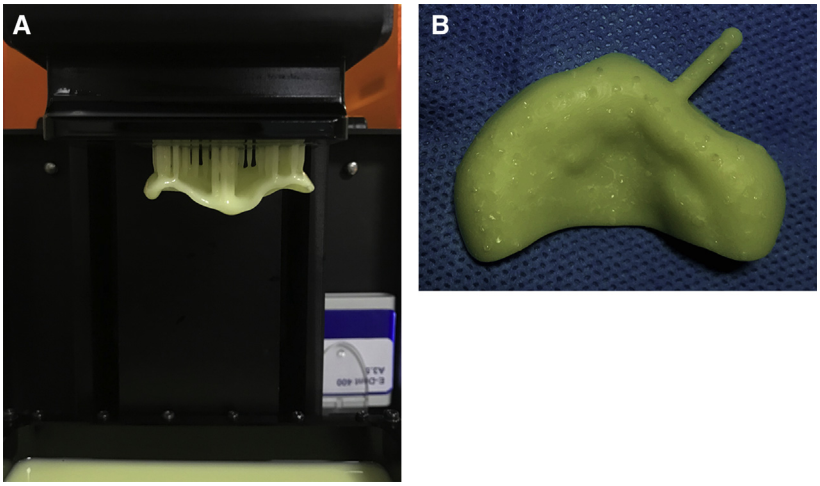

Fig 4. Stereolithographic printing of molding plate. **A,** Printed molding plate on the platform printer. **B,** After trimming, removal of supports, and autoclave sterilization.

1.5 hours. Next, the molding plate was removed from the build platform, rinsed twice with 95% ethanol for 20 minutes and allowed to air dry. Complete polymerization was accomplished with the use of a polymerization chamber by exposure for 10 minutes. Supports were removed and the molding plate was then autoclave sterilized (Fig 4, *B*). A series of appliances were then created (Fig 5). Each pair of PNAM appliances consisted of a molding plate and a retentive button.

Molding plate insertion, taping, and nasal hook hanging

The traditional PNAM was joined with 2 parts: molding plate and nasal stent. Force from the nasal stent lifts the nostril apex and defines the top of the columella, while reacting force from the nostril apex retracts the noncleft alveolar process.

Interaction force between nasal stent and intraoral molding plate would reduce the efficiency and accuracy of the whole device. To eliminate the above interaction force, we designed the split-type PNAM. The molding plate and the nasal appliance were separate. Independent action of the nasal hook can improve nasal tip projection and better decrease the nasal alar base width.

Artificial skin (Hydrocolloid Adhesive Medical Tape; 3M Co) was clung to the infant's cheek as the base to protect the skin. Steri-Strip was used to bring lips together. The parents were instructed to start taping

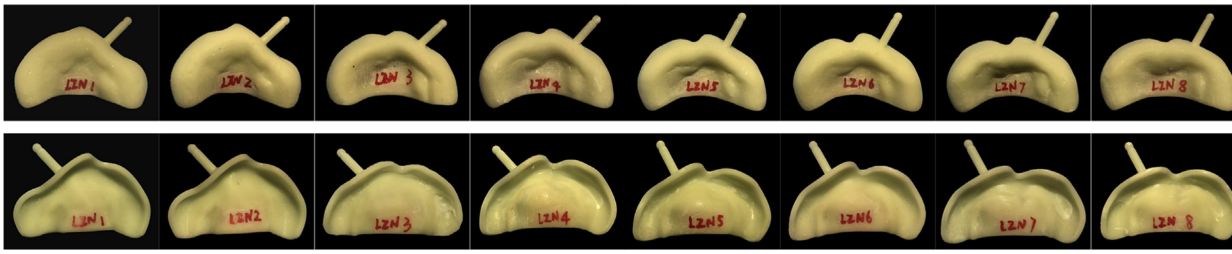

Fig 5. A series of 3D-printed molding plates.

Fig 6. Molding plate insertion, taping, and nasal hook hanging. **A,** Artificial skin and Steri-Strip were applied to the infant's cheek. **B,** Two extra layers of 3M tape were used for molding plate insertion and nasal hook hanging.

the lip across the cleft gap, from one side of the cheeks to the other side (Fig 6, *A*). The Steri-Strip was pulled to achieve an activation force of approximately 120 g at first, and decreased by 20 g per plate until the force was reduced to 60 g, which was maintained until cheiloplasty. Two extra layers of 3M tape (Transpore White Surgical Tape) were used to enhance the depressing force for the premaxilla. Although a lip adhesion alone produces uncontrolled orthopedic effects, a lip-taping force, in conjunction with a molding plate, yields a controlled movement of alveolar segments in a predetermined direction. For molding plate retention, denture adhesive cream (Polident; GSK, Brentford, U.K.) was first smeared on the tissue surface of the molding plate. After inserting the molding plate, the infant was observed for a few minutes to ensure that he was able to suckle while the appliance was stabilized with an index finger. Thinner tape strip was subsequently wound around the retentive button and affixed to the base tape to hold the appliance against the palate (Fig 6, *B*).

The molding plate was then inserted and changed to the next one sequentially each week. The parents were trained to insert, tape, remove, and clean the appliance. Instructions were given to keep the appliance in the oral cavity for 24 hours a day except during cleaning. Tapes were changed 2-4 times a day to make sure that the orthopedic force was strong enough, because suckling would wet the tapes.

We creatively used a nasal hook to replace the traditional nasal stent to support the deformed nasal alar cartilage adequately. A nasal hook was used synchronously with the intraoral molding plate. Active molding of the nasal cartilage was begun by taping the end of the tape on the noncleft-side forehead (Fig 6, *B*). The determination of the magnitude of the force was standardizing on whether the nose tip turned white, if not, the force should be increased. Tape connecting the nasal hook and the forehead was changed daily to make sure that the orthopedic force was strong enough to elevate the nasal tip on the cleft side.

Fig 7. Extra- and intraoral records after PNAM treatment (left) and after cheiloplasty (right).

The patient was recalled on a monthly basis, with basic records and minor adjustments made.

TREATMENT RESULTS

Through this procedure, the shape of the alveolus and nose were gradually molded to resemble the normal shape of these structures.

Pre- and posttreatment extraoral photographs revealed an improvement in the vertical level of the nasal alae (Fig 7). Height difference of the nasal columns on both sides was reduced from 1.85 mm to 0.36 mm and close to 0 after surgery. Discrepancy in the vertical level of both sides of the alae, which was 2.43 mm, was reduced to 0.55 mm before surgery and reached 0.14 mm after cheiloplasty. Nasal tip deviation from the midsagittal reference plane reduced from 6.14 to 2.16 mm before surgery and became 0 after surgery. Discrepancy of the alar base width decreased from 9.67 mm to 3.83 mm and reached 1.49 mm after cheiloplasty. Distance between the ridges of vermilion on the cleft side decreased from 16.68 mm to 8.67 mm during

PNAM treatment. See Supplemental Table I for changes of the nasolabial soft tissues.

By the end of PNAM treatment, the cleft gap was reduced with the 2 alveolar segments nearly contacted with a proper maxillary alveolar arch form (Fig 7). The posterior transverse alveolar width remained constant throughout the treatment, with remarkable diminution in the anterior cleft alveolar width. The sagittal arch length of the noncleft alveolar bone decreased obviously. The alveolus of both sides showed rotation toward the midsagittal plane, with the midline of the upper denture being corrected. See Supplemental Table II for changes of the maxillary alveolar arch.

DISCUSSION

In the 1990s, Grayson et al introduced the PNAM concept, which continues to play a significant role in neonatal CLP treatment.[4] This approach is preferred by certain orthodontists because it produces improved results, allows repositioning of the maxillary alveolus and surrounding soft tissues, and is beneficial for restoring normal oral function.[7-9] The reduced intersegment distance following PNAM improves arch symmetry and stability and thus may prevent arch collapse in the long term.[10]

Despite its advantages and usability, there exist some inadequacies in its clinical application. First, the PNAM devices currently reported in the literature are mostly unibody architectures.[5,7,9,11-15] Interaction force between nasal stent and intraoral molding plate would reduce the efficacy and accuracy of the whole device. Second, the efficacy of treating patients with the PNAM appliance has a positive correlation with clinical skills.[5] Besides, the curative effect is affected by the operation of patients' parents to some extent. They usually have no professional backgrounds in medicine and are unable to control the magnitude of force exerted on cheeks by the Steri-Strip. Excessive force will engender quick alveolus rotation, which can lead to overlap of bilateral maxillary segments, resulting in asymmetric alveolar arch form and T-type deformity. Conversely, insufficient force would hinder the closure of the bilateral segments before cheiloplasty, increase the surgical tension and adversely affect the maxilla after natal development, contributing to an unesthetic concave profile.

It is imperative to modify procedures to make the treatment cost-effective. The case we presented was a neonate with unilateral CLP treated with the use of an innovative split-type 3D-printed PNAM. A series of molding plates were designed by reverse-engineering software, printed by 3D printer, and delivered to infant's parents at the first visit. The 3D-printed alveolar molding device was used synchronously with a nasal hook. The complex system of forces was thereupon unified to achieve the goals of guiding the maxilla to shift more effectively and shaping the nose more precisely.

The basis of 3D printing a PNAM is to set up a stable 3D coordinate system for evaluating the alveolar molding regularity. That is the foundation for PNAM treatment digitization. In the available literature, the landmarks making up the reference plane were mostly on maxilla.[7,8,16,17] It is worth mentioning that the premaxilla region is obviously influenced by orthopedic interventions. Therefore, whenever descriptions of relative movements in 3D analyses are related to a basis reference plane defined by the anterior points, they seem to be of limited value.

The Frankfort horizontal plane is an important spatial reference plane in orthodontics, representing the habitual postural position of the head.[18] More importantly, it is a reference plane independent from the maxilla. This makes it possible to describe the dislocation of the cleft palate segments objectively. The tuberosity points are defined as constructed points at the junction of the crest of the alveolar ridge with the outline of the tuberosity in which the alveolar ridge and the outer surface of the tuber maxillae meet.[19] They locate next to the pterygoid process of sphenoid bone. The distance between 2 tuberosity points is correlated with the distance between the pterygoid processes and appears to be most stable in relation to the large variety in form of infants' maxillas.[17] Therefore, the present study chose the Frankfort horizontal plane as the reference plane to construct the 3D evaluation system, and the line between 2 tuberosity points served as the y-axis. A novel 3D coordinate system independent from the premaxilla was then constructed to evaluate the molding effect of the alveolus.

As shown above, the width of the alveolar cleft and the sagittal arch length of the anterior arch decreased with the use of the split-type 3D-printed PNAM. The upper denture midline deviation was also corrected. The contour of the alveolus was normalized without collapsing the alveolar segments. The relative width of the nose was generally perceived to be narrowed by the nasal hook treatment, with a simultaneous lengthening of the columella. The alignment of the nasal base region was achieved by bringing the columella toward the midsagittal plane, and the symmetry of the nostril apertures was improved.

The advantage of split-type 3D-printed PNAM treatment can be summed up as precision, personalization, and simplicity.

By applying the digital technology to the manufacture of the molding plate, expected movement distance and rotation angle of the alveolar segments can be precisely designed before treatment. The elastic tape is used to regulate the magnitude of the force. A nasal hook independent from the intraoral device is used synchronously with the molding plate, which props up the nasal tip stably in the right direction.

The patient's personalization is represented by days of birth, cleft type, width of cleft gap, etc. Personalized treatment plan should be designed considering neonate's initial state. Patients with a narrow cleft gap, for example, need alveolus outward growth first and rotation behind to avoid the overlapping of 2 alveolar segments; although it is diametrically opposite for neonates with a wider cleft gap. Personalization of the surgeon also needs to be considered. Different surgeons have different surgical characteristics. A number of surgeons require a total closure of the gap before cheiloplasty whereas some others want a certain gap to be reserved to facilitate a vomer flap operation. Application of digital technology in 3D printing of PNAMs can set the final state of alveolar molding in the initial stage to meet the individual needs.

As mentioned above, weekly visits of PNAM treatment are inconvenient for CLP families in remote regions. Digital technology assists physicians in designing and printing all of the intraoral molding plates before treatment. The data compensating for the alveolar growth is preset precisely in a series of PNAM devices. Under this circumstance, the digital approach can greatly decrease the subsequent visit frequency. Moreover, even inexperienced physicians in remote areas can treat CLP patients via assistance provided by cleft lip and palate centers in advanced cities.

Although this workflow is practical and can produce an accurate outcome, this report is only a preliminary trial in 1 patient. Further longitudinal studies with long-term follow-up will provide more evidence. The potential for digital technology in PNAM treatment is great.

CONCLUSIONS

This clinical report describes an innovative approach to the management of an infant with unilateral CLP. Split-type 3D-printed PNAM treatment helped to reduce the cleft gap, improve the arch form, approximate lip segments, and distinctly improve the morphology of the nose by correcting flattened nasal wings.

SUPPLEMENTARY DATA

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ajodo.2018.04.031.

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