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DOI: 10.1177/27325016231161841
journals.sagepub.com/home/fac
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3D Infant Orthopedic Nasal Molding System for Improved Outcomes in Cleft Nasal Deformity

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Abstract

The cleft nasal deformity is a hallmark feature in infants with cleft lip and palate and includes deviation of the caudal septum, slumping of the ipsilateral lower lateral cartilage, and obstructed nasal breathing. Presurgical orthopedic protocols have been developed and refined over several decades now, with limited evidence supporting their long-term efficacy, particularly with respect to the nasal form. Some limitations relate to variabilities in studies and data outcome measures. We have designed a novel method of presurgical orthopedic molding using separate nasal and intraoral components. The independent nasal device, called the Rhinoplastic Appliance System, is introduced in this technique article and shown to have distinct biomechanical advantages for the unilateral and bilateral cleft nasal deformity, both presurgically before lip repair, and postsurgically for nostril maintenance. As with CAD/CAM technologies for the intraoral plate component of traditional NAMs, we believe digital preparation of RAS devices, combined with separate oral appliances, can allow for more effective and reproducible presurgical infant cleft molding with less operator dependence and family/patient burden.

Keywords

cleft lip and palate, cleft nasal deformity, nasoalveolar molding, presurgical orthopedics, rhinoplastic appliance system, passive orthopedic appliance

Introduction

One of the most noticeable features in newborns with cleft lip ± palate (CLP) is a nasal deformity. Patients with cleft lips have altered anatomy, including a short philtrum and abnormal orbicularis muscle insertion into the cleft margin and alar wing. In addition, the infants have a predictable pattern of nasal deformity including a caudally dislocated nasal septum from a displaced anterior nasal spine of the maxilla, a shortened columella, attenuated and flattened lower lateral nasal cartilage on the cleft side with a flared alar base, and an inferiorly rotated upper lateral nasal cartilage.[1] The characteristic septal deviation deformity, which can be associated with hyperplasia of the inferior turbinates, may lead to paradoxical nasal obstruction.[2] CLP therefore results in disruption of the nasal foundation, with collapse of the nasal tip structures.[3]

Despite continuous refinement over the years, pre-and post-surgical orthopedics is still a work in progress.[4] The "single-appliance" presurgical nasoalveolar molding (NAM) technique proposed by Grayson et al[5] in 1999 has gained popularity over the last 2 decades due to its efficacy in reducing the severity of clefts in early infancy. NAM aims to improve nasal symmetry with a nasal stent on the cleft side attached to the maxillary plate in CLP patients. This appliance is manually manufactured and must be exchanged because of dentoalveolar growth or cleft reduction. The nasal stent is mounted onto the new plate.[6] This procedure lengthens visiting hours for patients and parents or requires more treatment sessions and increases feeding difficulties. For the past 22 years, our institutions have used a NAM modification, the "passive orthopedic appliance (POA)," with progressive changes in the maxillary obturating appliance and the nasal molding.[7] Unfortunately, due to its operator-dependent nature and complexity, this technique is difficult to implement on a massive scale. Additionally, the heterogeneity of study designs, outcome variables, follow-up periods, and inadequate data

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reporting have made it impossible to calculate effect sizes and perform reliable clinical trials and meta-analyses. According to an analysis done by van der Heijden et al,[8] all studies evaluating the efficacy of NAM on nasal asymmetry had a low grading of recommendation level.

The advent of new digital technologies including computer-aided design and manufacturing (CAD/CAM) have allowed for the production of customized appliances for presurgical orthopedic treatment in newborn babies with clefts,[9-11] with the promise of improved clinical efficacy and reproducibility of results, as well as standardization of treatment protocols. The purpose of this technique report is to showcase an innovative approach to pre- and postsurgical infant orthopedics in CLP patients focused on independently resolving the nasal and intraoral (palatal) components of the anatomical tissue discrepancies. The molding of the nasal structures is accomplished using a digitally designed and manufactured appliance called "Rhinoplastic Appliance System" (RAS). This technique improves results by creating a more manageable and effective treatment plan for all providers who may treat craniofacial patients (orthodontists, pediatric dentists, and craniofacial surgeons). Treatment of patients with nasal appliances at a young age can potentially reduce the number of future operations because of the malleability of cartilage molding in a newborn.[12] Moreover, postsurgically, experience has shown that the application of a dynamic nasal splint can contribute effectively to maintaining the surgical results by opposing forces of contracture.[13]

Technique/Device Description

Following the principles of presurgical NAM, applied and re-evaluated continuously for 20 years, we have developed a protocol that separates the management of the nasal and intraoral (palatal) aspects of the anatomic defect caused by the cleft using independent devices for the nose and maxilla. The nasal device, referred to as the RAS, was designed by the first author in 2003 to correct the patient's caudal nasal septal deviation, improve the nostril circumference and columellar length, elevate the nasal tip, and approximate the soft and hard tissue borders of the cleft defect. It specifically and individually addresses the vertical and transverse asymmetries of the nose, displacing the septum and nostrils to a more physiological and esthetic position before surgery and subsequently maintaining the correction of the nasal cartilage, avoiding its collapse after surgery.[14] The device, with its intranasal retention, has greater control to manipulate the nasal septum *laterally* in an infant with CLP. The system allows the for the nose to be maintained in a corrected position since the appliance has a lateral component with hooks, drawn by an elastic band taped to the infant's cheeks. This is in contrast to the traditional NAM appliance, which straightens the nasal septum with an extension up from the intraoral plate on the affected cleft side. The traditional NAM appliance is difficult for parents and professionals to keep in the correct position and mainly raises the tip of the nose, without correcting the deviation of the septum. Originally, the RAS devices were custom made, however now they are digitally designed and manufactured via CAD/CAM technology and are sequentially applied.

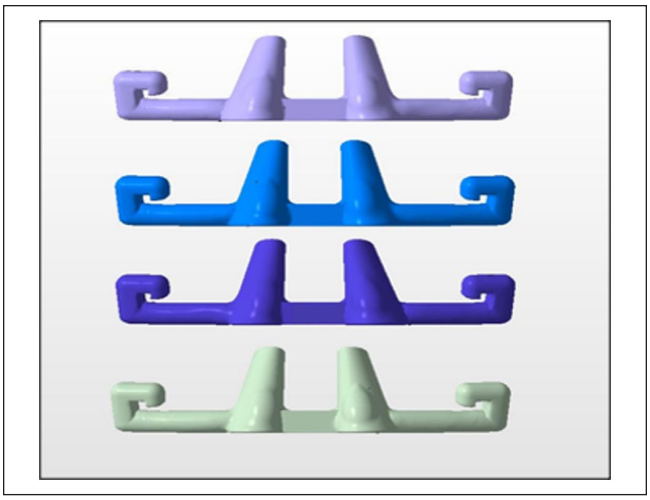

Figure 1. A kit of 4 sequential appliances with a progressive increase in the dimensions of the intranasal stent.

The implementation of our system requires a kit of 4 appliances (Figure 1) that are sequentially exchanged during the active phase of treatment. The smallest appliance is initially fitted and activated, and then should subsequently be changed every month, increasing the size to modify the nasal structures in preparation for primary lip repair surgery. The kits are available for right and left unilateral and bilateral CLP. The RAS is composed of the following elements (Figure 2):

(1) A nasal prosthesis consisting of intra-nasal extensions (stents) that are inserted into the nostrils, united by a columellar support and 2 lateral arms ending in hooks
(2) Two protective pads that avoid direct contact between the adhesive tapes that sustain the elastic elements and the patient's skin
(3) One adhesive labial tape
(4) Two adhesive tapes that sustain the elastic elements that provide the orthopedic forces

For the intraoral component of the presurgical orthopedic (PSO) intervention, different manufacturing methods have been proposed, harnessing the possibilities of CAD/CAM computerized digital technology. These have included simulating the modification of the alveolar segments, and transferring them as data sets to 3D printer devices, producing high precision, sequential palatal molding devices.[10,11,15] For the RAS, we are similarly using a nasal appliance system made with 3D technology and biomedical clear resin. The

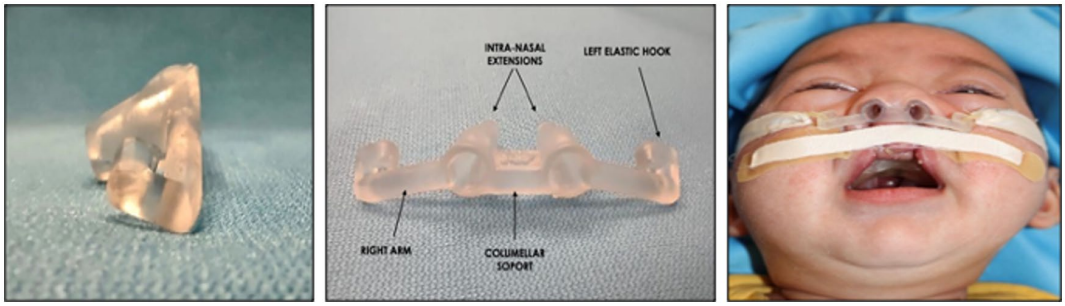

Figure 2. (Left) Lateral view; (Middle) frontal view; and (Right) clinical implementation of the rhinoplastic appliance system (RAS), with labial taping.

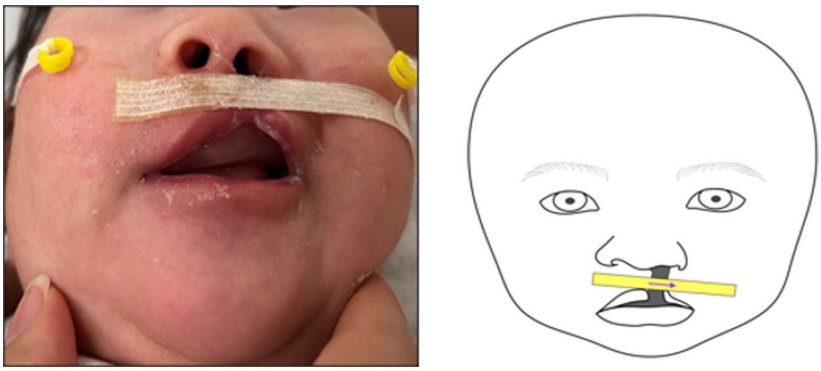

Figure 3. (Left) Example of labial tape; (Right) example of labial tape with slight downwardly tilted force vector.

nasal devices are based on models of normal anatomy and come in 4 sizes. Each size sequentially molds the nasal tissue over 3 to 4 weeks, allowing elongation of the columella, better projection of the nose, straightening of the nasal septum, and improvement of the shape and size of the nostrils.

Methods

As noted above, the RAS has been designed to pre-surgically modify the shape of the nostrils, correct the deviation of the nasal septum, elongate the columella, and improve the vertical asymmetry of the nasal alae in patients with CLP. Before the activation of the RAS, the labial tape is applied, approximating the labial borders of the cleft (Figure 3, left). In most cases, the tape must be slightly tilted toward the cleft side, producing a vertical downward force vector that will be crucial to the biomechanics of columellar elongation (Figure 3, right).

Once the labial tape is properly fitted, the appliance is inserted passively into the patient's nostrils, serving as a reference for the application of the protective pads and elastic-tapes. The level of force generated by the elastics is proportional to the distance they are stretched when coupled to the hooks. In this position, due to the vertical asymmetry of the nasal alae, the device will usually be tilted toward the cleft side (Figure 4, left). Initially, the elastic element of the non-cleft side is incorporated into the device's hook, followed by the engagement of the elastic element of the cleft side. The longer distance of engagement produces a larger *diagonal* force on the cleft side (blue arrow) (Figure 4, right) which, in turn, produces a horizontal force (dotted blue arrow) that promotes the correction of the nasal asymmetry by pushing the cartilaginous portion of the nasal septum toward the cleft side. Since the RAS was tilted before the engagement of the elastic elements, due to the diagonal disposition of the elastic force (blue arrow), it will *rotate* in a counterclockwise direction (dotted curved blue arrow), promoting the elevation of the nasal ala of the cleft side (Figure 4, right).

The therapeutic effect of the device uses light forces that produce stretching of the soft tissues (producing mild temporary ischemia of tip of the nose) and cartilage (Figure 5, right). Pressure is exerted on the tissue when size increments of the intranasal component are applied, or when tapes are adjusted, regulating the height of the nostrils, and straightening the nasal septum. The deficiency in the height of the columella is corrected by (1) the combined effect of the upward push produced by the incremental size of the

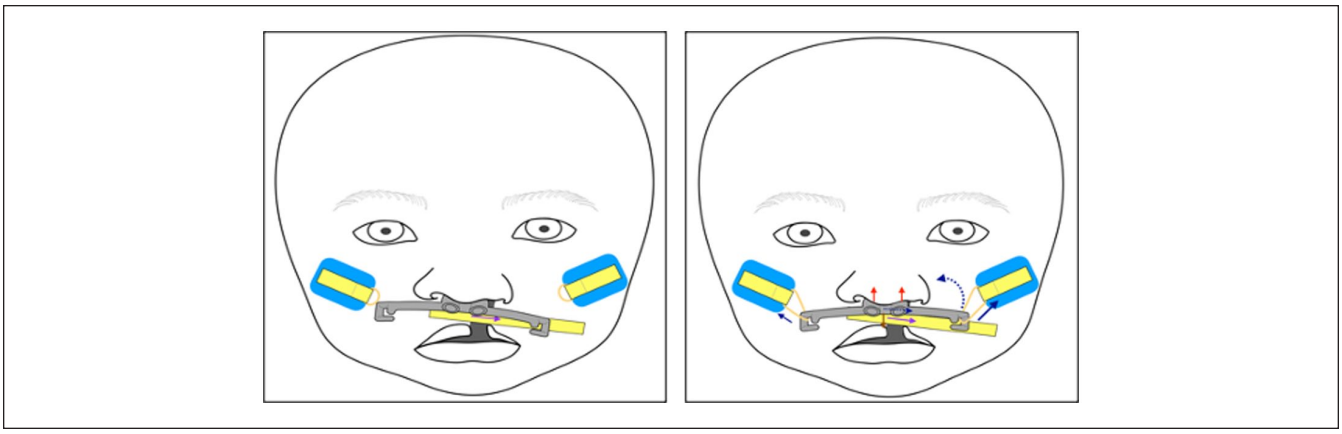

Figure 4. (Left) Appliance passively positioned; (Right) appliance actively positioned with elastic engagement.

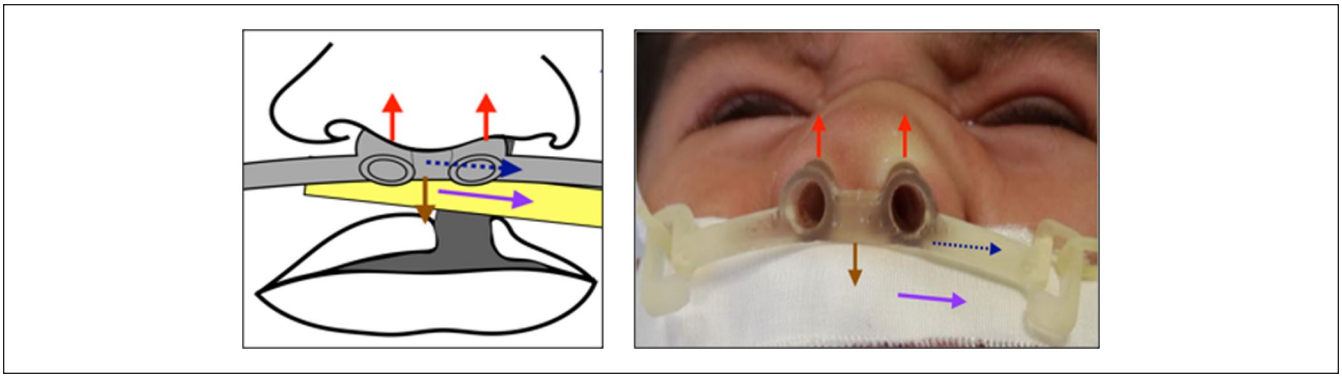

Figure 5. (Left) Individual forces produced by the RAS acting on soft tissues; (Right) mid ischemia is produced on the left nasal rim by columellar elongation forces.

intranasal extensions (red arrows), (2) the forces produced in the opposite direction, by the downward push of the columellar support (brown arrow), and (3) by the downward "pull" on the upper lip by the slightly tilted labial tape (purple arrow) (Figure 5).

Results

We typically use 1 or 2 different appliances in pre-surgical treatment, depending on the severity of the defect, with treatments starting in the first postnatal month, and patients returning approximately every 3 weeks for monitoring once compliance has been demonstrated. Clefts less than 7 mm wide can be corrected exclusively with the RAS combined with lip taping. In wider unilateral (Figure 6) and complete bilateral (Figures 7 and 8) clefts, we use separate RAS and intraoral (palatal) appliances when it is necessary to bring the premaxilla to the midline and align the occlusal plane. While RAS devices are now produced with CAD/CAM technology, intraoral devices are still largely being made with traditional manual methods of progressive adjustment, however are also amenable to production with digitized methods in the future. Using the RAS separately from the intraoral appliance offers important advantages, in that it is more comfortable for the baby and easier for physiological feeding. Preliminary observations from our institution indicate that with the current scheme, infants with even complete clefts of the lip, alveolus, and palate are ready for lip repair surgery by 4 to 5 months of age.

Post-surgical RAS treatment can be helpful to prevent the relapse of nasal asymmetry and avoid cicatricial stenosis that may occur in the caudal-most portion of the nasal cavity of CLP patients, resulting in a micronostril.[16] This is especially important given the consensus that long-term postoperative lip repair results are often far from ideal, despite the modern emphasis on primary nasal correction. This undesirable outcome is attributed to tissue and cartilage memory during the healing process.[17] We have attempted to continue post-operative use of the RAS for 3 to 6 months to optimally maintain nostril shape, however this may be limited by patient tolerance with advancing age.

Discussion

The impact of respiratory physiology in patients with CLP is of much interest, however studies are limited. In the craniofacial skeleton, the nasal septal cartilage and the sphenothmoidal and spheno-occipital cranial synchondroses are distinguished from

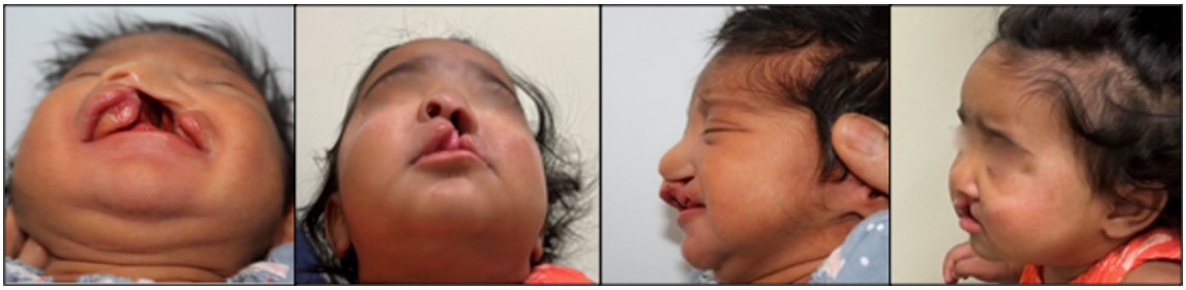

Figure 6. Unilateral case before and after presurgical molding with 2 appliances. (Left 2 panels) Basal view, before (left) and after (right); (Right 2 panels) lateral view, before (left) and after (right).

Figure 7. Bilateral case before and after presurgical molding with 2 appliances. (Left 2 panels) Basal view, before (left) and after (right); (Right 2 panels) lateral view, before (left) and after (right).

Figure 8. Basal view of a bilateral case before (left) PSO after (left middle) PSO; after surgical correction, short-term (right middle); and after surgical correction, long-term (right).

other craniofacial cartilages in possessing intrinsic growth potential. Growth of the nasal septal cartilage outstrips the growth of other skeletal and soft tissues in the midface to such an extent that it is the pacemaker for the growth of the face and the anterior portion of the skull.[18] Patients with unilateral CLP deformities commonly develop nasal airway obstruction, necessitating septoplasty at the time of definitive rhinoplasty.[19] For this reason and those mentioned previously, our pre-and post-surgical orthopedics treatment protocols allocate more emphasis on addressing the nasal defect and its functional and anatomic manifestations.

Successful implementation of a PSO treatment protocol in cleft patients requires a considerable expenditure of time and effort on behalf of both medical professionals and their families.[20] Due to the simplicity and reproducibility of the activation process, the RAS reduces these burdens. Most traditional

versions of the NAM appliance frequently overexpand the nostril on the side of the cleft due to the difficulty encountered by the caregiver in applying the appropriate level of force on the nasal stent when activated at home.[21] While the NAM appliance produces forces on the nose that originate in the maxillary plate, altering the levels of force during feeding, the RAS applies a mechanically independent force anchored on facial soft tissues, favoring a more stable force system and faster adaptation to the treatment.

Although PSO treatments were popularized more than 60 years ago, the evidence related to this topic is still weak and inconclusive, mainly due to the difficulty of the study designs and data collection[22] as well as continued uncertainty as to the effects on maxillary morphology and facial growth.[23] The difficulties involved in controlling variables such as type of appliance, hours of use, and replicability of at-home activation further complicate the prospects of undertaking reliable clinical studies. The fact that the RAS uses standardized manufacturing procedures and activation protocols in which force levels are determined by pre-calculated distances of elastic deformation, offers new possibilities in terms of methodological sound experimental study designs. There are more than 120 reports of PSO treatments, but little scientific evidence. The lack of a validated measurement instrument makes it hard to design a clinical trial to demonstrate the effectiveness of PSO and decreases the level of evidence in clinical recommendations. Future work will include standardized trials incorporating the RAS design, in addition to separately produced intraoral plate components, both produced with the aid of CAD/CAM technology. Such efforts are currently underway in our institution as well as partnering cleft treatment centers in Mexico City.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethical Approval

Institutional Review Board approval was obtained for this work under a generalized umbrella review, subproject #7, Nicklaus Children's Hospital Division of Plastic Surgery, S. Anthony Wolfe, MD (2019105RI).

Informed Consent

Informed consent was obtained by the authors (from the infants' parents) on behalf of Nicklaus Children's Hospital (Miami, FL, U.S.A.) for the use of identifying patient photographs included in this report.

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