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Preliminary results for a three-dimensional printer novel approach in nasal molding for infants with unilateral cleft lip and palate

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ABSTRACT

Background: Presurgical orthopedics (PSO) can be used to treat patients with cleft lip and palate. PSO is a powerful tool for changing nasolabial and palate structures during the 1st months of life, helping improve the restoration of form and essential functions. **Materials and Methods:** This preliminary retrospective pilot study evaluated treatment efficacy with a nasal PSO protocol using the Rhinoplasty Appliance System (RAS) in seven centers in Mexico that specialize in managing children born with cleft lip and palate. Twenty-five unilateral cleft lip and palate patients (UCLP) (9 females and 16 males) were treated by an orthodontist trained in the RAS system. The severity of the cleft, treatment time, the number of devices used during treatment, and clinical complications were documented using a survey. The results were measured using standardized submental view two-dimensional photographs. Two independent investigators evaluated one angular variable and two lineal variables before and after treatment. The columella deviation angle was measured.On the affected side, between the line in the middle of the columella from anthropometric points Sn-Prn and reference line between Sn-Sbal. The lineal variables that compare the healthy and cleft sides were measured as ratios. The lineal variables were nostril width (this measurement is taken from all are anthropometric points to the columella on the horizontal plane) and nostril height (from the nasal base plane to the upper part of the nostril). All relevant data are within the paper and its supplementary information files. **Results:** The resulting data were analyzed using a paired *t*-test ($P < 0.05$). The results of nasal symmetry showed a statistically significant decrease in the nostrils' midline deviation and horizontal symmetry and a statistically significant increase in unilateral patients' vertical symmetry and tip-base angle. Of the reported complications, 36% of orthodontists did not express any concerns. Meanwhile, 32% experienced skin irritation on the cheeks, 16% faced challenges with treatment attachment, 12% encountered difficulties in applying the correct tractions on the rubber bands, and 4% required a larger device from the system. **Conclusions:** In these preliminary results, the Rhinoplasty Appliance System (RAS) streamlines procedures, enhances reproducibility, and offers a practical solution for nasal protocols in managing cleft lip and palate in infants. RAS improves nasal symmetry, displaying promising outcomes in holistic care for infants with UCLP conditions. This research highlights the potential of three-dimensional printing and innovative digital approaches to revolutionize presurgical interventions for pediatric patients, particularly in infant nasal PSO.

Key words: Cleft lip, cleft palate, cleft lip and palate, nasoalveolar molding, presurgical orthopedics, Rhinoplasty Appliance System

INTRODUCTION

This study introduces an innovative approach to nasal presurgical orthopedics (PSO) for infants with

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unilateral cleft lip and palate (UCLP) by implementing a three-dimensional (3D) printer-based Rhinoplasty Appliance System (RAS).

Traditionally, various methods have been employed in PSO to enhance effectiveness, with nasoalveolar molding (NAM) introduced by Dr. Grayson and Cutting in 1995 being a notable shift.[1] The complexity of nasal deformities in patients with cleft lip and palate presents an ongoing challenge for surgical teams.[2] This deformity, characterized by a congenital upward deviation in nasal tips, varies in severity and is classified with a deficiency in description in the literature.[3,4] While the articles discuss nasal deformities, few delve into their physiology, function, and relation to facial growth.[5] Nasal deformity impacts breathing, esthetics, facial growth, speech, and behavior from birth.

Maintaining proper nasal ventilation is crucial for achieving optimal facial growth. PSO has proven to be a powerful tool in the early months of a cleft patient's life, aiming to restore anatomy and physiology. Studies show that most children with complete UCLP exhibit more oral than nasal breathing, and early verbal breathing patterns can be challenging to correct.[6,7]

In the past, our team at Nicklaus Children's Hospital in Miami used a modified NAM appliance passive orthopedics appliance (POA) for many years with satisfactory results.[8] However, professionals still recognize the limitations of conventional techniques, such as the operator-dependent nature, steep learning curve, and laborious manufacturing processes. This study addresses the difficulties professionals and caregivers face.[9,10] Clinical complications, such as overexpanded nostrils, highlight the challenges in applying the required force on the nasal stent, while the dependency on frequent appointments further burdens patients and caregivers.[11-13]

A 3D printer-based Rhinoplasty Appliance System (RAS) was developed to overcome these challenges and facilitate large-scale implementation. This system offers a set of sequential, incremental devices that can be used by professionals or caregivers with training in the device. Using sequential devices reduces the need for frequent in-person visits and highly specialized human resources.[14,15] The RAS, initially manually designed and patented, transitioned to a 3D printer system [Figure 1a and b]. The initial concept of the appliance was derived from custom-made plaster replica models of normal nasal anatomy and a 3D scan of the noses of an 8-month-old noncleft baby and a 2-year-old noncleft baby. It will be described in a future article. This allowed for an assessment of nostril size in newborns and how it evolves during the first 2 years of life. The resulting device, created from biocompatible resin meeting stringent medical standards, is FDA approved, ISO 13485 certified, and suitable for contact with skin and mucous membranes. The RAS is a practical option promising to facilitate the management of nasal deformities of infant patients born with cleft lip and palate.[16,17]

The nasal correction kit includes four sequential appliances, molded from the normal nasal anatomy as described before, each featuring a gradual increase in the dimensions of the intranasal stent. This specialized kit meticulously addresses vertical and transverse asymmetries of the nose. It focuses on individually correcting these asymmetries by strategically applying vector forces displacing the septum and nostrils, aligning them to a more physiological and esthetically pleasing position before surgery [Figures 2 and 3].

RAS offers two kits tailored to those with UCLP on the left or right side. The stent on the defect is larger, while the opposite, relatively normal side keeps the standard measurement. This clever design ensures that the normal side keeps an average size while the affected cleft side stent increases. The goal is clear: straightening the septum and elevating the lateral alar cartilage on the affected side, harmonizing the nasal features, and contributing to functional and esthetic improvements.

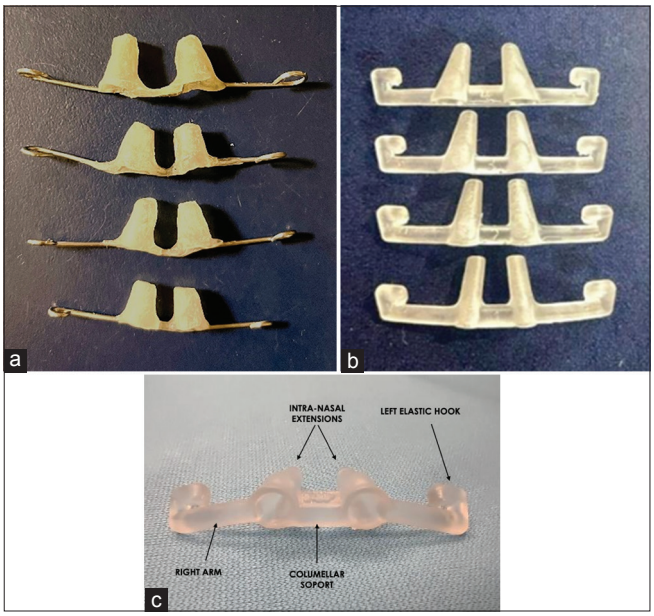


Figure 1: (a) A nasal prosthesis kit in four different sizes custom-made, b) A nasal prosthesis kit three-dimensional printer in four different sizes, (c) Appliance description: intra nasal extensions, columella support, and two lateral arm finishes in hooks

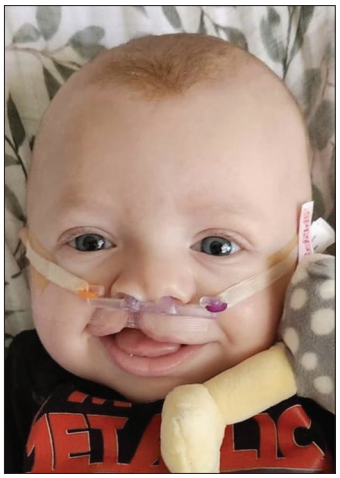


Figure 2: The vertical and transverse asymmetries of the nose

The Rhinoplasty Appliance System (RAS) comprises the following elements:

1. A nasal prosthesis kit in four different sizes [Figure 1b] consisting of intranasal extensions (stents) inserted into the nostrils united by columellar support and two lateral arms ending in hooks [Figure 1c]
2. Two protective pads that avoid direct contact between the adhesive tapes that sustain the elastic elements and skin of the patient [Figure 3]
3. One labial adhesive tape [Figure 3]
4. Two adhesive tapes that sustain the elastic elements that provide the orthopedic forces [Figure 3].

In cases where the cleft size between the alveolar segments exceeds 7 mm, the application of distinct intraoral and nasal appliances becomes necessary. Specifically, a maxillary device is recommended in complete unilateral patients with a cleft exceeding 7 mm before nasal molding. Conversely, when the gap measures 7 mm or less, the treatment can be efficiently executed solely using the Rhinoplasty Appliance System (RAS). Drawing from our experience with the custom-made system, it has been observed that nasal molding alone is feasible when the cleft side is 7 mm or less.[8]

The Rhinoplasty Appliance System was used to correct the patient's nasal septum deviation and deformation, improve nostril size and columella elongation, elevate the nasal tip, and stretch prolabium before CL surgery. In our experience, using a nasal appliance separately from the oral device is better for the baby and more effortless for physiological feeding, which is vital to consider.

In postsurgical treatment, many articles show that it is essential to maintain the nasal results and

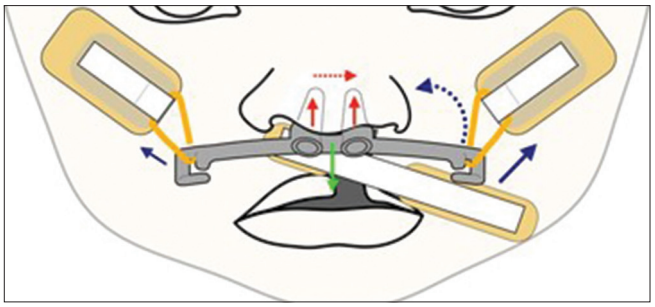


Figure 3: Two protective pads that avoid direct contact between the adhesive tapes

prevent relapse on the cleft side and stenosis on the nostrils.[18-20] In our protocol, the surgeon sutures the commercially available silicone stents and keeps them for 3 weeks. After that, the stents are removed, and the clinician uses RAS number 2 or 3, depending on the nostril size. We keep it for 6 months after surgery [Figures 4 and 5].

MATERIALS AND METHODS

This retrospective pilot study was conducted across seven specialized centers for managing patients with cleft lip and palate in Mexico. Ethical approval for the pilot study was obtained from the corresponding ethics committees of each center.

The study focused on centers implementing the Rhinoplasty Appliance System (RAS), wherein a kit of four appliances is sequentially exchanged during the active phase of treatment. The process begins with fitting and activating the smallest appliance. Subsequently, based on the severity of the cleft, professionals exchange it for the next sequential appliance whenever an increase in size is considered necessary to modify nasal structures in preparation for primary lip repair. All patients were treated by an orthodontist who had undergone prior training in the RAS, which included comprehensive protocols, a manual, parent brochures, and a workshop.

The treated sample consisted of 25 unilateral patients, distributed in five unilateral mild cleft sizes that do not exceed a 5 mm gap between the alveolar segments. Three unilateral moderate sizes that do not exceed an 8 mm gap between alveolar segments and 17 unilateral severe clefts with a gap of more than 8 mm. Mild: The separation between the alveolar segments is non-existent or less than 3 mm. Moderate: The separation between the alveolar segments is about 5 to 8 mm. Severe: The separation between the alveolar segments is 8 or more mm [Table 1]. Use the cleft severity table on methods. Nasal symmetry in unilateral cleft patients was evaluated using standardized two-dimensional photographs.

Three variables, two lineal and one angular, were considered in a submental view. Two independent professionals performed all measurements. Nostril Width: Measure from the AL alare anthropometric landmark point to the columella on the horizontal plane. The anthropometric point is likely a specific point on the nose used as a reference. The columella is the strip of skin between the nostrils. This measurement helps determine the width of the nostril from a specific point on the nose to the center of the columella. Nostril Height: Measure from the nasal base plane to the upper part of the nostril. The nasal base plane is a horizontal reference plane, likely defined in the study. This measurement helps assess the height of the nostril from the base of the nose to the upper part of the nostril. Columella Deviation Angle: Measure the angle on the affected side between the line in the middle of the columella from anthropometric points Sn-Prn and the reference line between Sn-Sbal. Anthropometric points Sn-Prn likely represent the length of the nose, and Sn-Sbal could be another reference line.

Table 1: Cleft severity

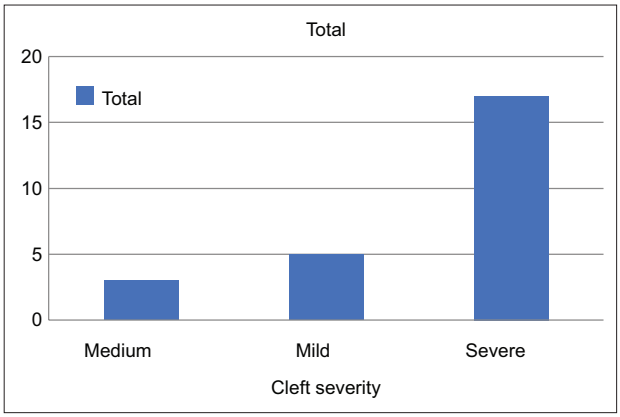


Figure 4: A unilateral case after surgery

Figure 5: Post surgery

Patients were treated with RAS devices made in 3D printers exclusively for the pilot study.

After the PSO treatment, orthodontists were surveyed to collect information regarding treatment time, how frequently they used the following size from the system, how many sizes of appliance sequences they needed in each case, and patient complications [Figures 6 and 7].

RESULTS

This preliminary study reports 25 patients with UCLP, 9 females and 16 males. The RAS results were analyzed with a paired *t*-test ($P < 0.05$); the results of nasal symmetry showed a statistically significant decrease in the nostrils' midline deviation and horizontal symmetry and a statistically significant increase in unilateral patients' vertical symmetry and tip-base angle.

The RAS devices address concerns such as nasal septum deviation, nostril size enhancement, columella length improvement, nasal tip elevation, and the harmonization of tissue borders within the cleft defect.

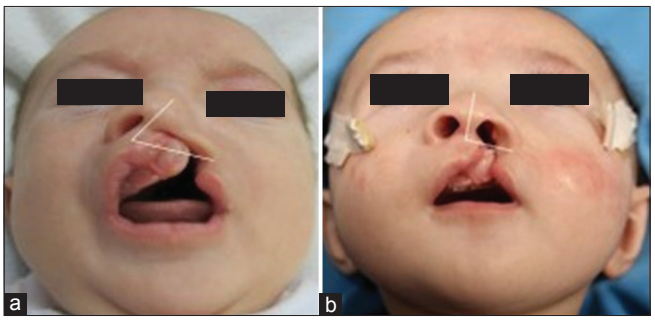


Figure 6: Before and after presurgical orthopedics (a) and nasal molding angle measures between the columella deviation angles (b)

Our results from photography analysis showed that all the unilateral patients improved on the nasal base to columella angle and increased the septum by an average of 37% [Table 2 and Figures 8 and 9] before and after PSIO. After surgery, results are in Figures 4 and 5. Only one patient out of 25 had minimal improvement, and the professional reported that parents did not use the device consistently on the patient. The other patients showed a significant improvement in nasal septum straightening, nasal symmetry, and elevation on the lateral alar on the cleft side.[21]

According to the survey, Treatment Duration: Most participants, constituting 72% of the total, used the system for 4 months. A significant portion, 16% of the participants, utilized the system for 5 months. 8% of the participants reported using the system for 3 months. The smallest percentage, 4% of the participants, had a treatment duration of 2 months. These findings provide a comprehensive view of how long participants engaged with the system, with a predominant number using it for 4 months.

For the interval time, professionals exchanged devices for a larger size: 19 (76%) patients every 4 weeks, 4 (16%) patients every 3 weeks, 1 (4%) patient every 5 weeks, and 1 (4%) patient every 8 weeks [Table 3]. Concerning the number of devices clinicians needed to finish the nasal PSO, the results were that 68% of clinicians used the four sizes (complete RAS system), 24% used three sizes, and 8% used two sizes. Regarding the professional difficulties or patient complications, 9 (36%) patients did not have complications or difficulties, 8 (32%) patients had skin irritation, 4 (16%) patients had poor treatment attachment, 3 (12%) patients had problems using the correct force on the rubber bands, and in 1 (4%) patient, the clinician considered that the patient needed a bigger device not available in the kit [Table 4].

DISCUSSION

We found a benefit in the presurgical treatment of UCLP of doing nasal molding separate from maxillary molding. This method allows professionals and caregivers to do the treatment on fewer appointments than other methods. This distinctive method not only offers practical benefits in terms of efficiency but also holds the potential for global standardization, providing a feasible treatment option that can be disseminated through professional education to benefit a larger population of patients worldwide. The simplicity and the use of different premade sizes of the RAS system also offer the advantage of experienced clinicians assisting new clinicians remotely.

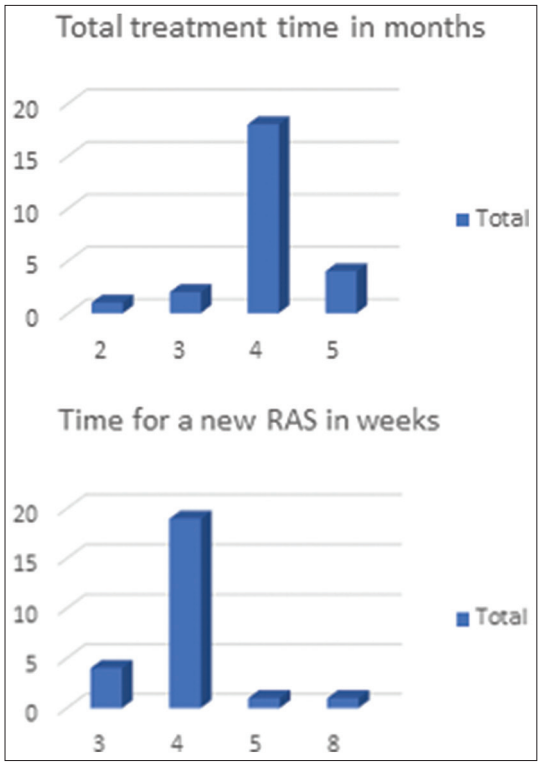
As a pilot study, our current findings emphasize the necessity of ongoing research and continual system refinement and methodology refinement. This iterative process is crucial to optimize its applicability and maximize the positive impact on patients undergoing PSO. This approach recognizes the evolving nature of medical interventions and underscores the commitment to advancing health-care practices to improve patient outcomes.

The broader discussion surrounding PSO, particularly in the context of cleft lip and palate surgery, prompts considerations about the optimal timing of interventions, especially in newborns. As part of the presurgical

Table 2: The results of nasal symmetry were the media analyzed by comparing the healthy side to the cleft side showed a statistically significant paired *t*-test (*P*<0.05)

Measurement	Mean with a standard deviation	Increase/decrease	P-value
Deviation	7.68 ±6.45	Decreases	0.00025*
Vertical	2.125±2.01	increases	0.00074*
Horizontal	5.8±4.88	Decreases	0.00025*
Tip	26.18±16.59	increases	0.00001*

Table 3: Treatment time is in months, and the time between rhinoplasty appliance system sizes is in weeks



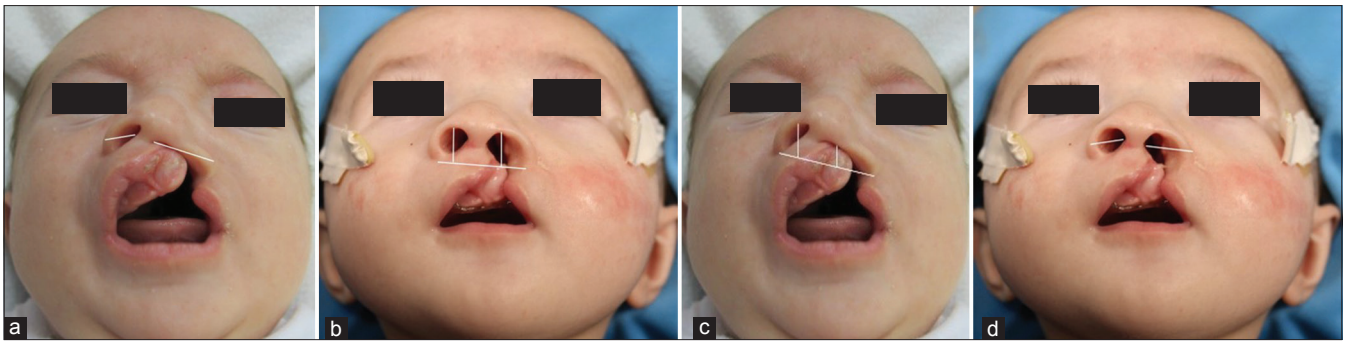


Figure 7: (a-d) Lineal measures before and after presurgical nostril wide and height were evaluated as radios, and symmetry was assessed by comparison of cleft versus noncleft side

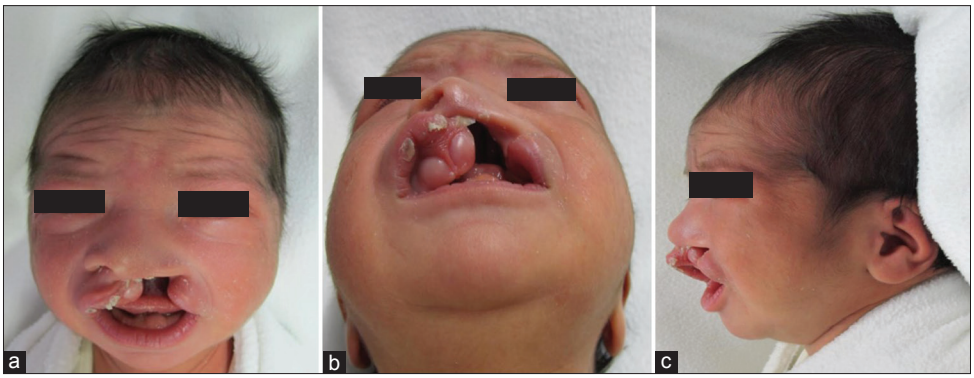


Figure 8: 2 weeks old newborn before PSIO with complete unilateral cleft lip and palate case. (a) Frontal view CUCLP, (b) basal view, (c) Lateral view

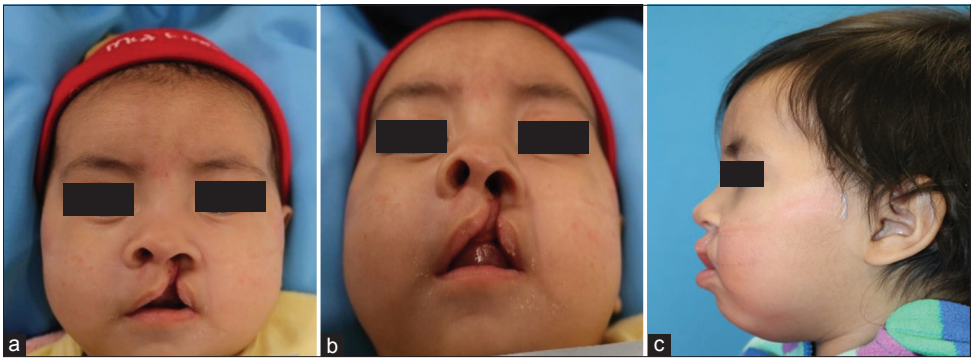
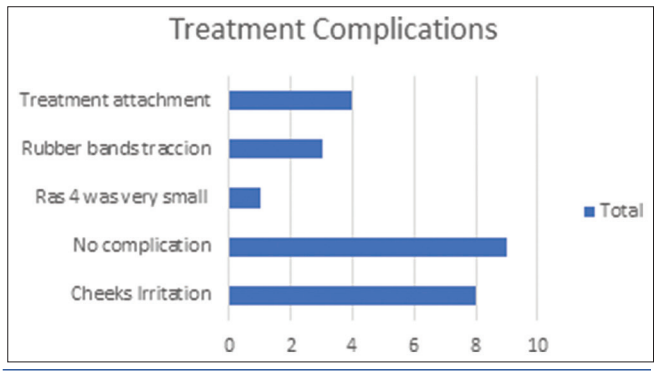


Figure 9: 5 months newborn after PSIO with complete unilateral cleft lip and palate. (a) Frontal view, (b) basal view, (c) Lateral view

Table 4: Professional complications during treatment



treatment, nasal molding emerges as a promising avenue, offering the potential to address nasal septum alignment and enhance newborn breathing. Given the vital role of proper breathing in craniofacial growth and functional development, this innovative approach becomes particularly pertinent.

Moreover, recognizing the long-term implications of cleft palate conditions, including sleep apnea, attention deficit disorders, allergies, and underdeveloped maxillary sinuses, highlights the need for a comprehensive approach to care. The prospect of conducting further

studies to assess the impact of nasal molding on respiration during the early months of life and its potential influence on the lifelong health and well-being of patients with cleft lip and palate underscores the significance of ongoing research in this critical area of medical science.

CONCLUSIONS

Implementing the RAS is a notable advancement, streamlining procedures and augmenting reproducibility. In addressing the demand for alternative nasal protocols, RAS provides a practical solution, equipping health-care professionals with more accessible tools. Notably, the observed enhancements in nasal symmetry stand as a testament to the promising outcomes achievable through RAS, particularly in the holistic care of infants grappling with UCLP conditions. This research sheds light on the potential of innovative digital approaches, particularly 3D printing, to reshape and improve the landscape of presurgical interventions for pediatric patients, revolutionizing infant nasal PSO.

Acknowledgment

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Nil.

Conflicts of interest

The RAS, initially manually designed and patented (Mejia 2021), transitioned to a 3D printer system [Figure 1a and b]. Invention name: Mejia, M (2021) RHINOPLASTY APPLIANCE AND METHOD (56) OF FORMING THE SAME, Patent No.: US 11,166,835 B2, U.S. Patent and Trademark Office.

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