CASE REPORT

Presurgical Nasoalveolar Molding and Columella Lengthening for Early Unilateral Cleft Lip and Palate Rehabilitation: A Comprehensive Clinical Case Report

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Abstract

Aim and background: In presurgical infant orthopedics, nasoalveolar molding (NAM) therapy has gained more popularity worldwide in treating patients with cleft lip and palate because of its soft tissue nasal corrections in addition to the alignment and close approximation of the cleft alveolar segments, which gives better postsurgical stability and esthetics with minimal tissue tension and scar formation to the patients.

The clinical case report aims to show the better post-NAM results, which had helped to gain better postsurgical results in terms of stability and aesthetics in hard and soft tissues.

Case description: This clinical report presents the case of a 9-day-old female patient reported in the Department of Orthodontics and Dentofacial Orthopedics at Sharad Pawar Dental College and Hospital with nonsyndromic complete left-sided unilateral cleft lip and palate with a defect size of 13 mm, which was treated with a presurgical nasoalveolar molding (PNAM) appliance. NAM therapy takes advantage of circulating maternal estrogen, bringing pliability to the cartilage cells for presurgical cartilage molding. This has resulted in the closure of the alveolar defect from 13 mm to <2 mm in the present case, with nasal symmetry restoration due to nasal stent within 12 weeks of active treatment; hence, the postsurgical esthetics are maintained.

Conclusion: NAM therapy remained beneficial and had significant aesthetic results in reducing the cleft defect size. Because of this, it gained a big surgical advantage in reducing tissue tension, primary stability, and scar formation.

Clinical significance: By reducing the cleft defect size of the dysplastic maxillae, which brings the lip segments and alveolar segments closer to each other, it thus reduces the defect size for primary lip and palate repair. This serves as the biggest advantage for maxillofacial and plastic surgeons to maintain the posttreatment results with redefined aesthetics and symmetry.

Keywords: Case report, Cleft lip and palate, Nasoalveolar molding, Nasoalveolar molding therapy.

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Introduction

The most common incongruity causing abnormal craniofacial development during the gestational period is known to be cleft lip and palate (CLP). According to the global survey of epidemiology, the incidence of CLP was found to be 1:600 newborns,[1] running in decreasing order from highest in Asian populations followed by Caucasians and Africans.[1] The frequency of the same in the Indian population was 3,500 patients per year.[1] The predisposing factors concerning such conditions are mainly genetic and environmental in etiology, which fail to meld maxillary and median nasal processes and palatal processes during the 5th to 12th week of intrauterine life (IUL).[2] This malformation in the craniomaxillary region can affect a patient's whole life and deteriorate normal functions, such as feeding difficulties, hearing, speech difficulties, poor oral hygiene, and recurrent upper airway infections.[2] Hence, approaching the condition and treating the malformation in a multidisciplinary way stand paramount.

Since the 1950s, "presurgical infant orthopedics (PSIO)" has been pursued as an auxiliary neonatal therapy for the rectification of CLP.[3] However, these traditional approaches have remained insufficient to achieve soft-tissue nasal cartilage corrections.[3] Presurgical nasoalveolar molding (PNAM) brings a transformative shift from the conventional PSIO techniques. It has gained popularity in achieving soft tissue nasal cartilage corrections along with alveolus molding.[3] The concept of PNAM was introduced by Grayson in 1993.[4] The fundamental intent of nasoalveolar molding (NAM) therapy is to decrease the severity of the initial cleft defect by the method of serial negative sculpturing and passive molding of the alveolar fragments with an intraoral molding prosthesis and correction of soft tissue nasal deformity (nonsurgical elongation of columella) with the help of wire and acrylic nasal stent.[4] Reducing the defect size presurgically by PNAM has the advantage of allowing surgeons to have reduced postsurgical scar and contracture of the tissues, with better retention of the corrected deformity.[4] After the depletion in the

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alveolar cleft width is established, the nostril base and lip segments have improved alignment.[4] NAM appliance consists of the passive intraoral acrylic plate for alveolus molding, a nasal stent for nasal molding and columella lengthening, which is made of a soft liner and acrylic resin that is attached to the acrylic plate by stainless-steel, swan-shaped round wire and a downward-tilted retention arm for retention of a passive appliance with the help of elastics. The treatment of CLP starts from the early 1st week postparturition to 6 to 8 weeks of age to gain the maximal advantage of maternal circulating estrogen in fetal blood, which corresponds to the elevated hyaluronic acid. This interferes with intercellular linkages and offers plasticity to the cartilage cells and alveoli and delivers more stable results postintervention.[5–7]

Case Description

Patient's Information

A 9-day-old female patient reported to the Department of Orthodontics and Dentofacial Orthopedics in Sharad Pawar Dental College and Hospital, Sawangi (Meghe), Wardha, Maharashtra, India. The case report is presented according to the CARE guidelines checklist.[8]

Clinical Findings

On clinical and physical examination, unilateral left-sided complete CLP was identified and diagnosed with a cleft defect of size 13 mm (Figs 1 and 2).

On examination by a physician, it was diagnosed as a nonsyndromic cleft defect. The physician's approval and the patient's informed consent for NAM therapy were taken before the intervention.

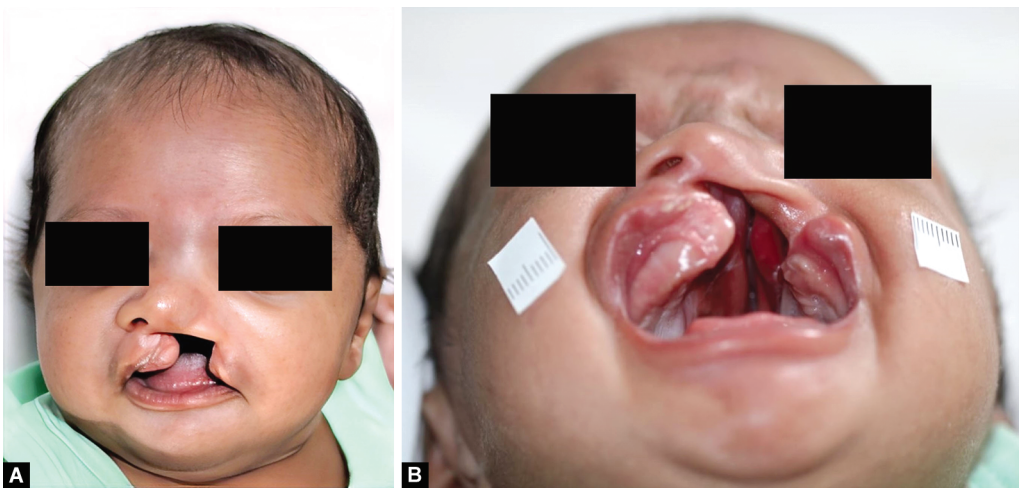
Diagnostic Assessment

Intra-alveolar cleft impression was procured with a "heavy-bodied polyvinyl siloxane" fast-setting impression material (Hydrorise™; Zhermack) for preparation of the diagnostic cast and assessment of the defect (Fig. 3).[9]

At the time of impression taking, the baby was turned upside down in position, with the tongue positioned in a forward direction. This facilitated the drainage of oral cavity fluids, which reduced the chances of their aspiration in the airway tract.[9] Proper care was taken to obtain the fine details of the soft tissues, undercuts, buccal and labial sulcus, buccal and labial frenum, and the extent and depth of intra-alveolar cleft defect for alveolar prosthesis fabrication. After the set of

Fig. 2: Pretreatment intraoral occlusal view with the intra-alveolar defect size of 13 mm

Fig. 3: Pretreatment maxillary impression for the diagnostic cast



Figs 1A and B: Pretreatment photographs: (A) Frontal view shows deviated alar cartilage; (B) Worm's eye view shows depressed medial and lateral crus of the nose on the affected side

impression material, the impression tray was removed, and a thorough examination of the oral cavity was done to check for the residual particles of impression material left over. Impression was then poured with a type-III gypsum product (Kalstone™; Kalabhai Karson™, Mumbai, India) to procure a definitive cast. After cast retrieval and duplication for working, the defect was measured from the alveolar base with the help of a vernier caliper, which was found to be 13 mm in size.

Appliance Fabrication

All the undercuts on the working cast were blocked out with the help of the modeling wax. The spacer adapted on the working cast, on which the prosthesis of 3 mm thickness was fabricated with the help of clear heat-cure acrylic resin (DPI Heat Cure™), for structural stability and future trimming of intaglio alveolar surface during serial negative sculpturing.[9] The alveolar plate was then trimmed and polished thoroughly to have nonirritant, rounded borders for the intraoral tissues. The extra-oral retentive arm was constructed with cold-cure resin (DPI Cold Cure™) and attached to the molding prosthesis with the help of a wire component (0.9 mm SS) at 40° downward angulation for prosthesis retention (Fig. 4).[9]

This wire component helped to angulate the retentive arm whenever it was required to change the force system for alveolar molding. It is to be attached to the molding plate vertically between the upper and lower lips at rest and transversely between the attachments of an upper cleft lip at the point of approximation when pulled together.[3] Grooves were made on the bottom of the retentive arm for elastic attachment. On the next clinical appointment, a try-in of the molding prosthesis was carried out to check for blanching of the intraoral tissues, and the acrylic was trimmed off the molding prosthesis wherever the tissues were blanched.[9]

Therapeutic Intervention

A broad basal micropore adhesive tape (1 × 2 inch) was placed on the patient's cheek superolateral to the oral commissure line bilaterally. These broad tapes served as an anchorage for smaller micropore tapes (0.25 × 4 inch), which are attached to the round elastics (red) at one end.[3] These elastics were stretched to double their diameter to place them over the retentive arm of the molding prosthesis for its retention against the palate (Fig. 5).

This gentle continuous force of 100 gm was generated and delivered to the alveolar segments.[4] Lip taping (3M™) was started 15 days after the appliance delivery to primarily adjust the patient with a preexisting molding prosthesis.[3] The adhesive tape was clung to the noncleft side and stretched onto the cleft side by bringing the philtrum and columella to the center. All required instructions were given to the patient to change these round elastics and tapes periodically to maintain proper force delivery for alveolar molding through the prosthesis. On every 15th day of the appliance delivery, the patient was kept on recall for intraoral examination to check for any inflammatory signs and indignation. Sequential modifications were made on the intaglio surface of the molding prosthesis to decrease the width of the intra-alveolar defect and align the segments in an arch form.[9,10] This was done by the selective addition of PermaSoft™ soft lining material and selective reduction on the opposite side at the alveolar intaglio surface of the molding prosthesis. Thus, the closure of the intra-alveolar segments and reduction of the defect size bring lip segments close to each other and reduce the width of the nasal base, which in turn brings laxness in the alar cartilaginous rim, beneficial for nasal molding.[9,10] As evidenced, after achieving the intra-alveolar defect size of 5 mm, lower nostril molding was started by integrating a nasal stent in the alveolus molding appliance at the labial flange area near the wire component of the retentive arm (Fig. 6).[3]

Fig. 5: Alveolar molding plate positioning

Fig. 4: Molding plate with retentive arm at 40° downward angulation

Fig. 6: Integration of the nasal stent for nasal molding

It was fabricated by bending 0.9 mm round SS wire (Konark™) into a "swan-neck" shape for functional lip movements and to get access for lip taping. The part (molding bulb) that got inserted into the nostril was "kidney-bean" shaped (bilobed) and made of cold-cure acrylic resin (DPI Cold Cure™).[3] This provided anterior thrust to the nasal cartilage. The superior lobe of the bulb was inserted into the left nostril to lift the depressed nasal dome until blanching was seen, and the inferior lobe of the bulb was used to lift the alar rim (Fig. 7).[3]

Follow-up and Outcomes

The active treatment was carried out for 12 weeks after the appliance delivery. The appliance was kept passive during the first week of appliance delivery for adaptation during the initial phase with the infant and parents. After the second week, the lip taping (Fig. 7) and trimming were started sequentially on the intaglio or tissue surface of the molding plate to align the shorter segment and reduce the width of the alveolar defect. The defect size was reduced to 5 mm after 5.5 weeks of the appliance delivery. At this stage, the nasal stent was incorporated with the molding appliance for nasal molding (Fig. 7). At the end of the NAM therapy, the intra-alveolar defect size, which was 15 mm, was reduced to <2 mm (Fig. 8).

Columella lengthening was achieved; a flattened nasal dome on the affected side was lifted up, and hence, nasal symmetry was achieved presurgically (Fig. 9).

Depiction of the comparison of the defect size at various time intervals (Figs 10 and 11).

After completion of the NAM therapy, the patient was given a referral to the Department of Oral and Maxillofacial Surgery for primary lip closure (cheiloplasty) after 4 months. Figure 12 shows the postsurgical image immediately after surgery.

The patient was kept on regular follow-up after surgery to avoid postsurgical complications. A minimal amount of scar tissue was formed with very little tissue tension after surgery. Figure 13 shows photographs 15 days after suture removal, with a very minimal amount of scar tissue formation, reduced postsurgical tissue tension, lengthening of the columella, and lifted nasal dome with morphofunctional correction in the external airway.

Further, the palatal closure surgery will be done at 9 months of age.

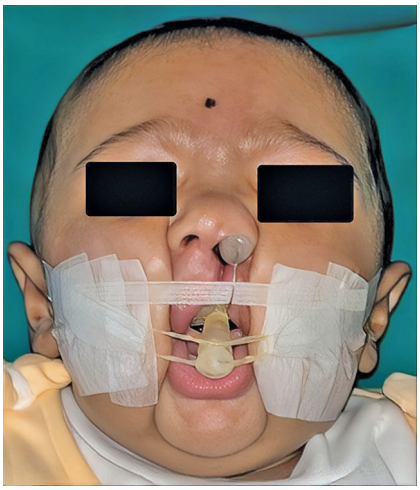


Fig. 7: Intranasal placement of nasal stent and lip taping

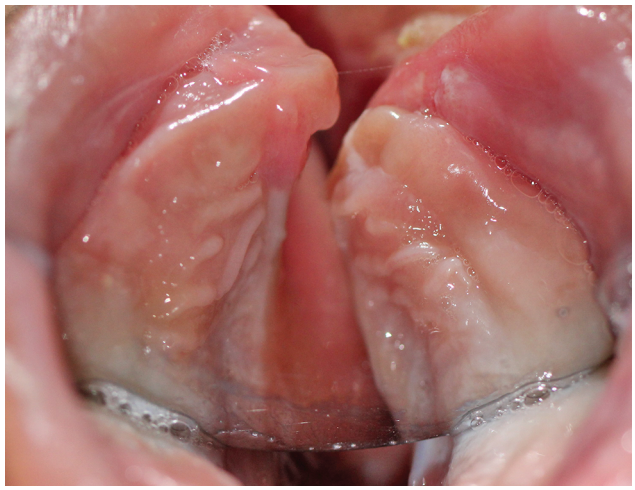
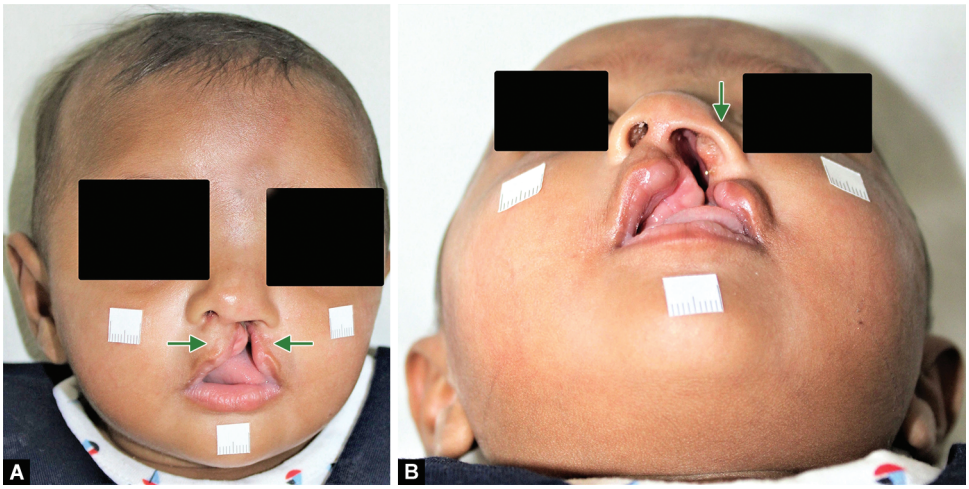
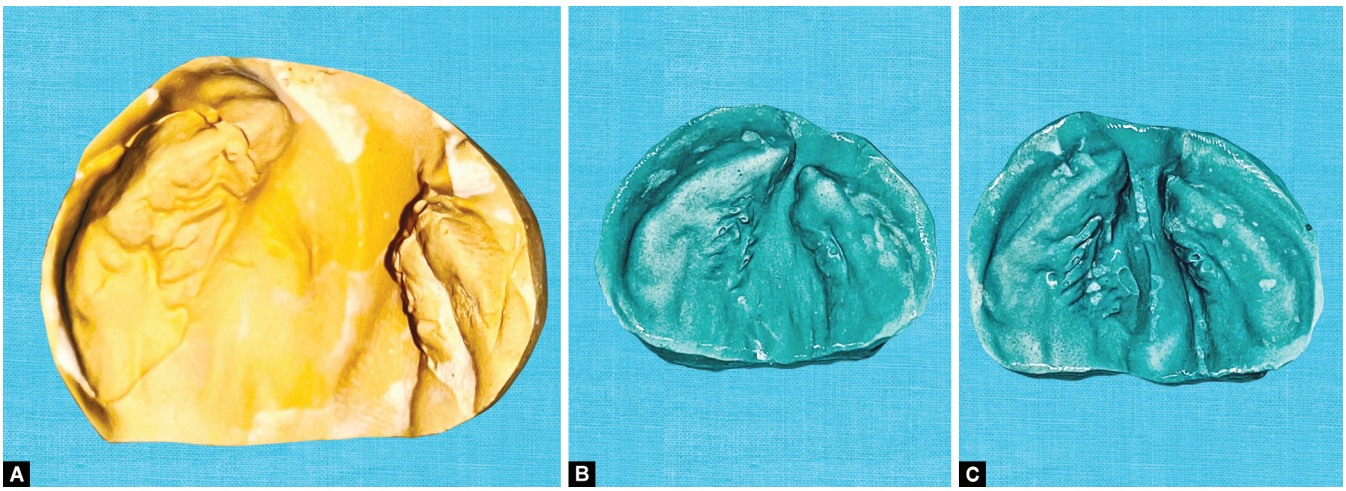


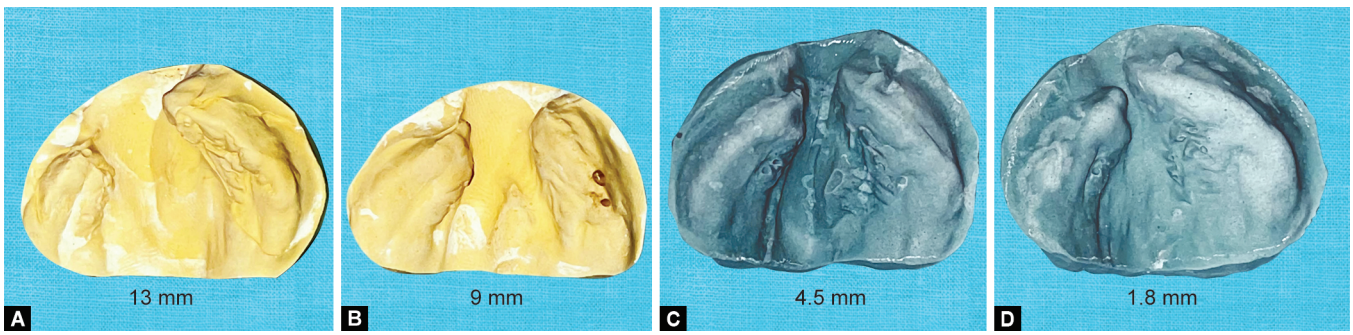
Fig. 8: Post-NAM intraoral occlusal view



Figs 9A and B: Post-NAM extraoral photographs: (A) Frontal view shows the reduced size of the labial cleft defect; (B) Worm's eye view shows a lifted nasal dome



Figs 10A to C: Comparison of casts before and after NAM therapy: (A) Represents the pretreatment cast with a defect size of 13 mm; (B) Represents the post-treatment cast with a defect size of <2 mm; (C) Represents the cast with a defect size of 5 mm, the stage at which the nasal stent was incorporated

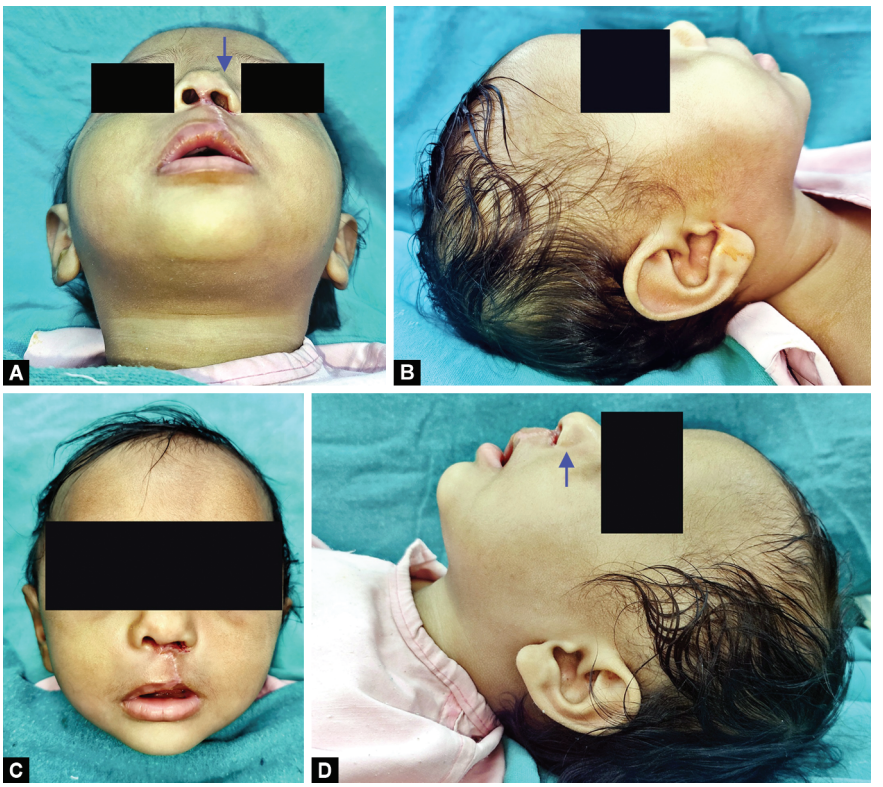


Figs 11A to D: Sequential alignment of the alveolar segments: (A) Intraalveolar gap of 13 mm before alveolar molding; (B) Intraalveolar gap reduced to 9 mm; (C) Intraalveolar gap further reduced to 4.5 mm at which nasal stent was incorporated with molding plate; (D) Stage at completion of presurgical nasoalveolar molding. Intraalveolar gap finally reduced to 1.8 mm and alignment of greater and lesser cleft segments was achieved

Fig. 12: Primary cheiloplasty (immediately after surgery) shows closely approximated labial segments with no tissue tension

Discussion

Presurgical infant orthopedics was first instigated by "McNeil" in the 1950s and was additionally improvised by others.[11] This method has been utilized for aligning and converging the alveolar segments preoperatively and hence encourages primary surgical repairs in cleft patients.[12,13] Presurgical nasal molding also has been instigated as an adjuvant neonatal therapy for preoperative rectification of nasal irregularity by using the malleability of alar cartilage soon after birth.[13] Grayson et al. put forward the amalgamation of presurgical orthopedics and nasal molding as a newly discovered approach called PNAM for correcting the alveolar cleft and the nasal deformities.[10] The major objectives of PNAM are rearranging the alveolar lesser and greater halves, approximating the clefted segments, molding the dearranged nasal cartilaginous matrix, and lengthening the affected columella.[14,15] After NAM therapy, the primary cheiloplasty should be done after 3 months of age.[9,13] Liou et al., in their study, found that there was a significant relapse in nasal symmetry in the 1st year postsurgery and then attained stagnancy in another 2 years after surgery.[13] There was a marked dissimilar growth pattern/recurrence seen between the noncleft and cleft sides in the first year postsurgery. The overall growth of the nasal cartilage on the affected side was comparatively less.[13] To compensate for the differential growth that led to relapse in the first year postsurgery, Liou et al. recommended the reduction of the cleft defect primarily with the help of NAM, which should be followed by overcorrection at the time of primary cheiloplasty and maintenance of surgical results by using nasal conformer.[13]



Figs 13A to D: Postsurgery photographs after 15 days of suture removal show decrease in tissue tension after approximation, elevation, and retention of alar cartilage, and thus nasal symmetry is achieved: (A) Worm's eye view shows postsurgical soft tissue nasal correction achieved equivalent to noncleft side; (B) Right profile view of noncleft side; (C) Frontal view shows postsurgical tension-free cleft lip approximation; (D) Left profile view of cleft side shows postsurgical correction of lateral crus of nose and columella lengthening achieved by presurgical nasal molding resembling the soft tissue nasal anatomy of noncleft side

Conclusion

Presurgical nasoalveolar molding is an amalgamation of alveolar molding and nasal tissue molding. This technique utilizes the principle of tissue expansion by harmonizing the hard and soft tissues before primary cheiloplasty to give better and well-acceptable postsurgical results with minimal scar tissue formation, reduces tissue tension, and hence provides better stability to the tissues with less postsurgical tissue contractures. Future revision esthetic surgeries can be avoided by PNAM.

Clinical Significance

In early neonatal age, NAM therapy has its advantage in hard and soft-tissue manipulation before surgery by taking advantage of the increased level of hyaluronic acid in the intercellular matrix of the cartilaginous tissue, which corresponds to the increased maternal estrogen in the infant's blood. Due to this, the tissue becomes plastic, less elastic, and compatible with molding nonsurgically. Thus, reducing the cleft defect size of the dysplastic maxillae, which brings the lip segments and alveolar segments closer to each other, and molding the depressed and dysplastic nasal cartilage on the affected side serves as the biggest advantage for maxillofacial and plastic surgeons to maintain the post-treatment results with redefined aesthetics and symmetry.

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