Case Report

Correction of extreme premaxilla malposition in an infant with BCLP using PNAM: a case report

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ABSTRACT

Introduction: Cleft lip and palate are the most common congenital abnormalities occurring in the orofacial region. Infants with this condition need to be treated as early as possible to achieve satisfactory treatment outcomes. Presurgical Nasoalveolar Molding (PNAM) is a non-surgical treatment used to reshape and realign the gingiva, lips, and nasal openings to reduce the severity of the cleft and reposition the premaxilla before labioplasty is performed. This case report aims to explain the use of PNAM to correct an extreme premaxilla malposisition that accompany complete bilateral cleft lip and palate in infants. **Case Report:** A month old infant came to Hasan Sadikin General Hospital with his parents with clinical conditions of complete bilateral cleft lip palate (CBCLP) and a protrusive and rotated premaxilla. PNAM was fabricated for the infant and periodic follow-ups were conducted to correct the lip and palate cleft as well as the premaxilla abnormalities to normal anatomical conditions. At the start of the treatment, the infant weighed 4.3kg, and the cleft gap measured 9mm. At the end of the treatment, the infant weight was 6.1kg, and the cleft gap was reduced to 6mm. Then the infant was referred to the Department of Oral Surgery for a labioplasty. **Conclusion:** BCLP requires a multidisciplinary approach. This case shows PNAM's success as a standard for improved surgical outcomes, function, and aesthetics.

KEYWORDS

CBCLP, extreme premaxilla position, PNAM installation

INTRODUCTION

Cleft lip and palate are the most common congenital abnormalities occurring in the orofacial region.[1,2]The genesis of clefts is complex, involving both environmental and genetic factors. Maternal smoking, maternal age, and folic acid insufficiency are risk factors that have been associated with the formation of clefts.[3,4] A child with cleft lip and/or palate not only has an aesthetic deformity but also significant functional morbidity, including stunted maxillofacial growth, speech abnormalities, difficulty eating and feeding, hearing loss, and/or recurrent ear infections. Living with a cleft poses a major health burden, despite the fact that it is typically not life-threatening[5]. Children with cleft lip/ palate (CLP) suffer a range of medical problems that include feeding difficulties at birth due to problems with oral seal, swallowing and nasal regurgitation, hearing difficulties due to abnormalities in the palatal musculature, and speech difficulties due to nasal escape and articulation problems. Beyond the physical effects on the patient,

CLP also has significant psychological and socioeconomic effects on both the patient and their family, including disruption of psychosocial functioning and decreased quality of life (QOL)[6,7]

Bilateral complete cleft lip and palate (BCLP) is widely recognized as the most severe subtype of common orofacial clefts.[8,9] This condition is characterized by significant variations in cleft anatomy, particularly in the premaxilla and prolabial segment. The premaxilla may exhibit different positions—it can project forward, rotate upward under the nose, or, in some views, appear absent. These variations are attributed to the mobility of the premaxilla in BCLP, as it is fixed apically to the vomer bone, which can cause abnormalities in its alignment. Additional factors influencing the premaxillary position include pressure exerted by the tongue and lip, as well as the forward growth of midline structures and lateral processes.[10] The interplay of these forces during early development contributes to the distinct anatomical differences observed in BCLP cases.

Moreover, one unique feature of bilateral complete cleft lip and palate is the absence of muscle tissue beneath the prolabial skin.[11,12] This absence not only affects the functionality of the upper lip but also has implications for surgical planning and long-term outcomes in rehabilitation. The complexity of BCLP anatomy underscores the need for a comprehensive, multidisciplinary approach to treatment. This typically involves coordinated efforts from surgeons, orthodontists, prosthodontists and speech therapists to address both functional and aesthetic challenges.

Pre-surgical Nasoalveolar Molding (PNAM) is a prosthetic device that can passively reposition the alveolar and nasal cartilage segments before the labioplasty procedure.[13,14] NAM treatment preceding the surgical intervention is used to reposition the nasal cartilages, to approach the alveolar processes, centralize the pre-maxilla, and to elongate the deficient columella.[15] Additionally, PNAM will cover the cleft, thus serving as a feeding aid to prevent choking and allowing the baby to obtain proper nutritional intake.[16,17]. Achieving satisfactory results in the surgical correction of bilateral cleft lip and palate deformities poses additional challenges particularly if it is accompanied with an asymmetry of the palate and alveolar segments and a protruding and displaced pre-maxilla.[18]

After the anatomical position was corrected, and the infant reached the rule of 10, the infant was referred to an oral surgeon.[19] This case reports aims to explain the use of PNAM to correct an extreme premaxilary malposition accompanying complete bilateral cleft lip and palate in infant. In this case report, during the course of treatment, there was a change in the position of the nasal stent due to the movement of the premaxilla, which caused a narrowing of the lip cleft. As a result, the position of the nasal stent was also adjusted.

Case Report

A one-month-old infant weighing 4.3 kg came to Hasan Sadikin General Hospital with his parents. The baby was referred from a local hospital and had not received any treatment after birth. Upon clinical examination it was found that the baby had complete bilateral labiognatopalatoschizis. An extraoral examination showed that the columella appeared protrusive and rotated towards the right, the left nostril was widened so that the nostrils looked asymmetrical (Figure 1).

After obtaining approval from the paediatrician and family members, impression of the maxilla was carried out with elastomeric impression material. The baby cried throughout the impression to ensure a maintenance of airway. Seven days after the impression the infant weighed 4.8kg and the appliance (PNAM) was inserted into the baby. The appliance must have good adaptation and retention in the baby's oral cavity. The baby was observed for several minutes after insertion of the device. The baby should be able to breastfeed without feeling nauseous or short of breath. After observing the appliance, it can be used for one week for the first stage of adaptation. Then the baby came to the hospital once a

week for 2 and a half month until the baby was ready to be referred to oral surgery for labioplasty. Several device adjustments were performed to correct the cleft lip and palate and premaxilla abnormalities to normal anatomic conditions during the visit. (Figure 2).

Seven days after insertion the infant's weight increased to 4.9kg, the premaxilla cup was grinded on one side and relined on the other side to move the premaxilla. Strapping (non-woven adhesive) was used on the baby's lips. 14 days after insertion, the infant weighed 4.9 kg.

Figure 1. Infant initial condition

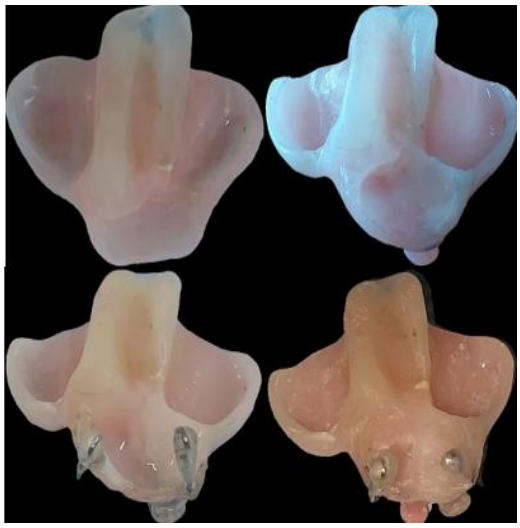

Figure 2. Appliance modification during treatment

An additional button was added to the left side of the premaxilla to aid in repositioning. There is a change in the strapping position due to the addition of a button. The strapping was given a tighter pulling force towards the right, where the premaxilla would be moved; on the left side, the strapping acted as an anchorage.

After 28 days of insertion, the baby's weight reached 5.5 kg, and the gap was measured, showing a reduction to approximately 6 mm, leading to the placement of a nasal stent in the baby's nose. After 49 days of insertion, the baby's weight was 5.9 kg. There were changes in the nasal stent position due to the premaxilla already moving, and the nasal stent was pressing against the premaxilla. Therefore, the nasal stent on the right side needed to be replaced with a new one in a different position to alleviate pressure on the premaxilla.

After 77 days of insertion, the baby's weight reached 6.1 kg. The premaxilla had shifted upward, prompting the button to be repositioned and additional buttons to be placed. These buttons were placed on the right and left sides of the premaxilla to serve as anchorage. 84 days after insertion, the baby met the

surgical criteria, and a referral was made to the oral surgery department for labioplasty.

Figure 3 Figures 1a The initial PNAM with one button to pull the premaxilla to the center position, Figures 1b the premaxilla at the center so another butter was added

Figure 4 Post Labioplasty

DISCUSSION

The treatment of complete bilateral cleft lip and palate (CBCL) presents unique challenges due to the complexity of the associated anatomical deformities. In this case, the extreme malposition of the premaxilla necessitated an effective and targeted approach using Presurgical Nasoalveolar Molding (PNAM). This prosthetic device serves as a non-surgical intervention to align and stabilize the alveolar ridges, reduce cleft severity, and improve outcomes for subsequent surgical procedures. Many scientific studies have reported the benefits of PNAM in correction of cleft lip and nose deformities in patients with complete unilateral and bilateral clefts. [20,21]

The effectiveness of this therapy can be affected by the timing of its initiation. PNAM treatment should be started as early as possible to maintain the achieved symmetry. Neonatal hyaluronic acid, which increased transiently with elevated estrogen levels, acts as a temporary barrier between intercellular material, giving cartilage a temporary lack of elasticity. If started later, the results may be less than satisfactory.[22]

In this case report, PNAM was started when the infant was 1 month old. This is almost the ideal time to start the therapy for a desirable outcome. Kinouchi et al., stated in their case report that the average age to start PNAM therapy is the 26th day after birth, while in their case, the therapy was started at 49.7 days after birth. The outcome of this therapy can also be enhanced by adequately supporting the appliance against the palatal tissues and by taping the lip segments together across the cleft.[23] Lip taping helps restore normal oral function, make surgical lip closure easier, and improve the overall outcome of the presurgical therapy.[24]

Strapping was used on the baby's lips seven days after insertion. Lip adhesion taping is helpful in stabilizing the plate. Fourteen days after the insertion, an additional button was added to the left side of the premaxilla to pull the premaxilla. A case report by Devya et al., reported three cases of infants with cleft lip and palate that used lip taping with an orthodontic elastic band looped around the retentive arm as a method to bring the lip segment closer together.[25] Active movement of the major segment was achieved through forces exerted by stretched elastics and over build-up of soft liner. This technique utilizes both active force application and passive growth to close the cleft space.[25] These will cause the orthopedic force of the premaxilla. Selective grinding created an empty space that became a place where that particular segment was sliding.[26]

The progression of this case demonstrated the effectiveness of PNAM, as evidenced by the reduction in the cleft gap from 9 mm to 6 mm and the improved anatomical positioning of the premaxilla. Breh et al. have the same result from his case that the gap was getting closer until 6 mm, allowing progression to the next stage of treatment.[26] PNAM is used to normalize the upper lip, alveolus, and nostrils by using a palate device attached to a nasal stent.[23] Nasal stenting has been used for the maintenance of the alar cartilage form in clefts with stenosis.[27]

After 28 days of insertion, when the alveolar cleft width had been reduced to 6 mm, a nasal stent was placed in the baby's nose, so that nasal cartilage moulding may begin. Adding the nasal stent to correct the nasal cartilage deformity before the reduction of the alveolar cleft width to less than 6 mm may cause unfavourable results such as the lengthening of the circumference of the alar of the nose.[23] Bhatia et al., also reported a case of a 1-month-old baby with bilateral cleft lip and palate treated with nasoalveolar molding with help of nasal stent that was added when the cleft alveolar gap was reduced to 5 mm, the nasal stent was added on the right side for moulding of the right columella.[28]

There were several changes in the nasal stent position along the way due to the ongoing movement of the premaxilla. After the premaxilla had shifted to the upper position, the button needed to be repositioned and additional buttons also needed to be placed. The consistent monitoring and adjustment of the PNAM device played a critical role in addressing the specific needs of this patient. Periodic follow-ups enabled modifications to the nasal stent and strapping positions, ensuring continuous progress. Such an approach aligns with the multidisciplinary treatment philosophy advocated in cleft care, emphasizing collaboration between paediatricians, surgeons, orthodontists, and prosthodontists. After the baby met the surgical criteria, a referral was made to the oral surgery department for a labioplasty.

The success of PNAM in this case aligns with findings in existing literature, which underscore its role in improving feeding, reducing nasal regurgitation, and ensuring proper weight gain in infants with clefts[2]. Additionally, PNAM's ability to centralize the premaxilla and elongate the deficient columella further supports its utilization in managing severe BCLP cases.[12] These outcomes are particularly significant as they facilitate better surgical results during labioplasty and contribute to improved functional and aesthetic outcomes.

While this case report on the use of Presurgical Nasoalveolar Molding (PNAM) to address extreme premaxilla malposition in infants with bilateral cleft lip and palate (BCLP) provides a successful outcome, the following limitations should be acknowledged: the treatment outcomes were assessed over a relatively short

period, up to the point of referral for labioplasty. Longer-term follow-up is necessary to evaluate the stability of the anatomical corrections and surgical outcomes. While weight gain was noted, detailed assessments of nutritional intake and growth patterns were not explored, which may influence overall treatment outcomes.

Bilateral complete cleft lip and palate (BCLP) is widely recognized as the most severe subtype of common orofacial clefts. The complexity of BCLP anatomy underscores the need for a comprehensive, multidisciplinary approach to treatment. Coordination among oral surgeons, orthodontists, prosthodontists and speech therapists are needed to address both functional and aesthetic challenges to achieve outstanding outcomes, alongside parents and caregivers.

Future studies could further investigate these aspects to optimize the treatment protocol and explore its applicability in resource-limited settings. The limitations of this report include the fact that the premaxilla of the baby was not sufficiently rotated to the intended place because, at the end of the treatment, the baby became uncooperative when using the appliance.

CONCLUSION

The successful application of PNAM in an infant demonstrated its effectiveness in reducing the alveolar cleft gap, enhancing nasal symmetry, and supporting weight gain, all of which are pivotal for preparing infants for subsequent surgical interventions. Therefore, PNAM should be recommended for all patients with BCLP to improve surgical outcomes and improve aesthetics and function. The implication of this case report is to show the treatment planning for a patient with extreme premaxilla malposition to give the best result of soft tissue management before referral to an oral surgeon, ensuring the surgery can be done with the most desirable outcome.

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