

Presurgical Infant Orthopedics for Cleft Lip and Palate: A Review

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Abstract

Cleft lip and palate deformities are some of the most common facial and oral anomalies. Severe cleft forms are associated with severe nasolabial deformities, and present a significant surgical challenge in order to achieve a functional and aesthetic outcome. Presurgical infant orthopedics has been used in the treatment of cleft lip and palate for some centuries. Starting with the McNeil method, several methods and modifications had been developed by different clinicians over time. However, there is no consensus in the literature on infant orthopedic methods and their benefits. Therefore, the aim of this review is to discuss presurgical infant orthopedic methods and their advantages and disadvantages. Presurgical orthopedics allows not only the alignment of cleft segments, but also molding alar cartilages and nose tip. In addition, this procedure allows performing primary alveolar grafting or gingivoperiosteoplasty to establish a union bone at the cleft side as well. However, there have been some studies reporting that there was no positive effect of presurgical orthopedics on the maxilla and maxillary arch. There is still no consensus in the literature on the best protocol for orthopedic and surgery methods for the treatment of cleft lip and palate in infants.

Keywords: Cleft lip and palate; Presurgical infant orthopedics; Nasoalveolar molding

Introduction

Clefts of the lip and palate are some of the most common facial and oral anomalies. These anomalies can appear with considerable variation in form and severity [1]. The maxillary structure of the cleft lip and palate is divided in two or three segments by the cleft of the palate and alveolus. A unilateral cleft defect is characterized by a wide nostril base and separated lip segments on the cleft side. Severe cleft forms are associated with severe nasolabial deformities, and present a significant surgical challenge in order to achieve functional and aesthetic outcomes [2]. The affected lower lateral nasal cartilage is displaced laterally and inferiorly, resulting in a depressed dome, increased alar rim, oblique columella, and overhanging nostril apex [3]. When associated with cleft palate, the nasal septum deviates to the non-cleft side, with an associated shift of the nasal base [4]. The bilateral cleft lip and palate may be symmetrical or asymmetrical, depending on the equality of involvement on both sides. In these patients, both nasal chambers are in direct communication with the oral cavity, and the turbinates are clearly visible within both nasal cavities. The premaxilla may be small or large, and projects considerably forward from the facial aspect of the maxilla [5].

Cleft lip and/or palate patients have feeding, functioning, aesthetic, speech, and psychological problems; and therefore, are best managed through a team of experts [6]. Management of cleft lip and/or cleft palate is a process that starts in infancy and continues in adulthood. These patients undergo many surgical procedures throughout life. Numerous methods and treatment strategies have been developed over the years to reduce the number of surgeries. Despite the fact that there have been many advances in surgery, certain orthopedic corrections prior to the primary surgery are still required in patients with cleft lip and palate. For this purpose, presurgical infant orthopedics is suggested for achieving better surgical outcomes. Since it is widely accepted the intervention of multidisciplinary teams for treating cleft patients, various methods have been developed for presurgical infant orthopedics. The aim of this review is to summarize the current state of knowledge of the effects of presurgical infant orthopedics (PSIO) on long-term outcomes of different treatment protocols. In this review, the advantages, disadvantages, effects on maxillary growth, and dentoalveolar arches, as well as in speech and complications of some

presurgical infant orthopedics methods will be discussed. A PubMed search was performed using the terms PSIO, presurgical nasoalveolar moulding and its long-term results and related articles were selected for the review. In addition, limitations of these studies will be discussed.

Presurgical Infant Orthopedic Methods

Some presurgical implementations in infants were developed a few centuries ago. Facial binding or adhesive tape strapping was used centuries ago to narrow clefts before surgery [7]. The use of a bonnet and strapping to stabilize the premaxilla after surgical retraction has also been reported [8]. Head is still used today to retract the premaxilla [9], and T traction using an external device has been reported as useful for surgical procedures in short-term [10]. All procedures, which at the time were mainly performed by orthodontists or by the surgeons, were based on the ever proven assumption that a narrow and well aligned cleft would be easier to repair, with less undermining and less mobilization of soft tissues. A narrower cleft would also require less tension in the repaired lip, and thus, the aesthetic outcome, facilitation of feeding, and speech was improved [11-15]. Presurgical infant orthopedics aims at securing a good maxillary arch form in an acceptable relation with the mandible, and at restoring normal oral function [7]. It also corrects angulation of the palatal shelves to a more horizontal position [16]. It is generally accepted that modern presurgical infant orthopedics started with the McNeil technique [17]. Since McNeil, many researchers have published their own methods for obtaining proper growth and development of the face, and for improving surgery results. The Hotz appliance, and then the Latham device were introduced for aligning the cleft segments [18-23]. Some authors used a combination of these appliances like in the Brogan technique, which combines the

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McNeil technique and the Hotz plate [7]. Later, Grayson and Cutting described the “presurgical nasoalveolar molding (PNAM)” concept for molding not only cleft segments but also nasal appearance [24]. Certain investigations reporting short- and long-term results of these methods mentioned above are summarized below.

Active appliances

McNeil Method: McNeil was the first in aligning presurgically the alveolar parts in cleft lip and palate patients [17]. He suggested the use of serial appliances to approximate cleft alveolar segments. By molding the palatal segments into the correct position using a series of acrylic plates, McNeil believed that this would produce a normal maxilla, while reducing the alveolar and palatal cleft at the same time. McNeil and Burston claimed that soft tissues overlying the hard palate were stimulated to grow, and they also added that neonatal maxillary orthopedics could control and modify the postnatal development of the maxilla [7]. In addition, the use of a series of acrylic plates may be favorable for patients who have to travel long distances and are unable to visit the orthodontics clinic weekly. Another advantage may be foreseeing the final position of the alignment arches.

Latham device: The other appliance used for aligning cleft segments is the Latham device, which was introduced by Dr. Latham [25-27]. In this approach, forces are applied using a pinned palatal appliance in order to manipulate mechanically the maxillary segments into close approximation, which is followed by alveoloperiosteoplasty and lip adhesion. According to Drs. Latham and Millard, these alignments allow the performance of gingivoperiosteoplasty (GPP), providing stabilization of the maxillary segments and reconstruction of the nasal floor [27]. Bercowitz et al. reported a longitudinal study in unilateral and bilateral cleft lip and palate treated with the Latham device. They also performed periosteoplasty in all cases, and compared the results to patients treated with a non-orthopedics procedure without GPP, and treated just with a lip adhesion method. They found a higher frequency of anterior and posterior crossbite in the presurgical orthopedics group [28]. Some authors have commented that those findings might be the result of the periosteoplasty procedure [7]. Dr. Latham applied the Latham device with less extensive surgery in cases of bilateral cleft lip and palate, and he assessed their dental occlusion and lateral head radiographs at 5 years of age. He found greater values for cephalometric measures in maxillary length, maxillary prominence, and ANB angle compared to previous cases [29].

In another long-term study, more anterior open bites and posterior crossbites were found in unilateral and bilateral cases compared to non orthopedics and periosteoplasty group [30]. In the study of Chan et al., in which they evaluated active appliances longitudinally, dental models of patients with unilateral cleft lip and palate (UCLP) were assessed using the Goslon Yardstick. GPP and lip adhesion were performed both in non-orthopedic and in orthopedic groups. No significant differences were found in Goslon scores between the two groups. The authors concluded that Latham procedures did not affect dental arch relations in preadolescent children with UCLP [31]. Similarly, Allareddy et al. stated that, outcomes are predictable without any major adverse events or complications by using Latham device [32]. Besides, it was stated that Latham device could be useful in unusual cleft cases [33].

Passive appliances

Hotz appliance: The Hotz appliance, also known as the Zurich approach, in which arch alignment is achieved by grinding away the acrylic in specific areas, was introduced after the McNeil technique. Although there is no strict research about the outcomes of the Hotz plate, it was stated that this method had a tremendous impact on cleft patients [20]. According to Hotz and Gnoinski, the primary aim of presurgical orthopedics is not to facilitate surgery or to stimulate growth, as postulated by McNeil, but to take advantage of intrinsic

developmental potentials. Therefore, the Zurich approach, after a lip operation is performed at the age of 6 months, palate repair is postponed until 5 years of age [22]. These authors concluded that orthopedic guidance combined with optimal timing of surgery has beneficial effects. In a study investigating the short-term effects of the Hotz plate, a harmonization in the vertical and transverse positions of the segments was found in the plate group compared to the control group [34]. In another study with 4 years follow-up of the Hotz plate group, the width of the palate was larger in the Hotz plate group than in the control group, but no difference was observed in the anteroposterior distance of the palate between the groups [13]. In addition, Sasaguri et al., investigated the long-term effects of the use of the Hotz plate and lip adhesion. They found that arch width and length of the anterior part of the maxillary were larger in the Hotz (+) group than in the Hotz and lip adhesion group, and in the group without Hotz plate and palatoplasty, at 5 years of age. The anterior part of the maxillary arch was wider in the Hotz group than in the other two groups [35]. In another long term study, Silvera et al. concluded that The two-stage palatoplasty in combination with application of the Hotz' plate had good effects on the maxillary growth than one stage palatoplasty without Hotz plate up to the age of 12 years [36].

Nasoalveolar Molding Appliance: In 1993 Grayson et al. introduced the PNAM concept, which continues to play significant role in neonatal cleft lip and/or palate treatment [24]. This approach is preferred by certain orthodontists because it produces improved results, and allows repositioning of the maxillary alveolus and surrounding soft tissues. Grayson and colleagues have reported many studies about PNAM treatment and they suggested the use of this appliance for improving nasal appearance, which results in less secondary nasal surgeries. This procedure also minimizes the need for later alveolar bone grafting, allows GPP, as well as effective retraction of the protruded premaxilla, and lengthening of the deficient columella [37,38]. In addition, produces limited maxillary growth disturbance [39]. PNAM has become very popular among orthodontists because of its nasal molding effect [40]. It is also suggested to correct septal deviation in early ages without surgery, since nasal cartilages are able to mold easily in the first postnatal 2 months because maternal estrogen provides the molding for the nasal cartilages.

In the PNAM approach the orthodontist adjusts the appliance every 1–2 weeks in 1 mm increments by removing hard acrylic resin, and adding soft acrylic resin. Once the maxillary alveolar segment gap is less than 6 mm, a nasal stent can be added to the appliance using acrylic resin placed on 0.036 inch-thick wire. The stent is positioned 3–4 mm into the nostril just below the soft tissue triangle of the nose. The size and shape of the stent is adjusted by adding soft acrylic to help create a “tissue expander” effect on the length of the cleft-side columella, as well as to reposition the malpositioned lower lateral cartilage. This process can take several months and results in a delay of the definitive cleft lip repair until approximately 4–5 months of age. PNAM should ideally begin before 6 weeks of age to take advantage of the early plasticity of nasal cartilages [18].

The first goal of PNAM in bilateral cases is to move the premaxillary segment posteriorly and medially, while preparing the lateral alveolar clefts to come in contact with the premaxilla [40]. The posterior lateral palatal shelves are molded to the appropriate width to accept the premaxilla. The premaxilla is retracted and derotated as necessary using the molding plate in conjunction with external tape and elastics. In addition, another important point is the elongation of the columella [38]. Cutting et al. stated that a saddle should be placed at the lip and columella junction. This saddle produces a separation between lip and columella that is expanded along an anterior vector, while the prolabium is stretched downwards using tape. Several months of appliance adjustments are often required. They also reported that much of the nasal tip shape produced through presurgical molding was

lost within a few weeks because of the fibroadipose tissue deposited between the widely separated nasal domes. Therefore, they suggested removing the fibro-fat tissue from between the nasal domes of the lower lateral cartilages, and suture them together in the midline without an external incision.

Grayson pointed out that multiple nasal surgical revisions are often indicated to approximate the nasal symmetry, because surgical techniques for managing nasal deformity are lacking. He also pointed out that in bilateral cleft lip and palate (BCLP), the deficient columella and ectopic premaxilla are the primary reconstructive challenges. Multiple nasal surgeries are required, which often result in excessive scarring at the columella-prolabial junction, and lack of nasal projection [41]. Therefore, he emphasized the importance of nasal molding before surgery in the early neonatal period. In addition, some other appliances for nasal molding have been reported [42]. However, the permanence of the improvement in nasal symmetry and appearance using PNAM remains controversial; however, there is a trend towards a positive effect. Liou et al. reported that nasal asymmetry was significantly improved after nasoalveolar molding in infants with cleft lip and palate; but after the primary closure of the cleft lip and nose, there was a significant relapse of the nasal asymmetry in the first year postsurgery, which remained stable afterwards. This relapse was the result of a significant differential growth between cleft and noncleft sides in the first year postsurgery [43]. Similarly, Pai et al., who used the nasoalveolar molding (NAM) appliance in their study, concluded that there was some relapse of nostril shape in width (10%), height (20%), and angle of columella (4.7%) at 1 year of age, compared to their presurgical status [44]. Therefore, the use of a nasal stent has been suggested after primary lip closure, at least for 6 months. Nonetheless, in a long-term study, it was found that the change in nasal shape is stable until early childhood, and it was emphasized that the symmetry in nose shape was maintained [45]. In another longitudinal study with 8 year follow-up, the rate of residual fistula was assessed, and it was found that NAM in conjunction with nasal floor closure contributes to a low incidence of oronasal fistulae [46]. Another longitudinal study investigating the effects of NAM approach on further surgery requirements reported that NAM-prepared patients were more likely to have less severe clefts, present the best surgical outcomes, and need less revision surgeries compared to patients not prepared with NAM [47].

The average age at the time of appliance is another concern. Grayson and Maull focused on starting neonatal period (within first one month) for better nasal esthetics results, while in the Latham technique, started within 8-11 weeks [32]. However, Shetty et al., evaluate the effects of nasoalveolar moulding (NAM) in complete unilateral cleft lip and palate infants presenting for treatment at different ages. Study groups comprised: group I treated with NAM within 1 month of age; group II treated with NAM between 1 and 5 months of age. This study concluded that the effects of NAM were most significant in group I. Group II patients also benefited from NAM, although to a lesser extent. This study validates the use of NAM in infants presenting late for treatment [48].

Limitations of Presurgical Infant Orthopedics Studies

Since cleft lip and palate treatment requires multidisciplinary approaches, outcomes may be affected at any stage of treatment. Therefore, the pure effect of PSIO appliances is very difficult to assess because of the variety in timing and sequence of treatment protocols for both surgery and orthodontics. The major difficulty when comparing different presurgical orthopedic methods is the type of surgical technique, and whether performing sequential palatal closure (one or two stage) or GPP, at what age, the experience of the surgeon [49-52]. The debate on the ideal time for hard palate closure is not over so far. Furthermore, because of the different timing for gathering representative sample sizes, and the inability to obtain untreated

control groups, the comparison of the effects of different orthopedic appliances is an almost impossible task. Another limitation is that previous studies, except one, have not examined the severity of clefts in infants. Peltomaki et al. found that patients with large clefts and small arch circumference, arch length, or both, showed less favorable maxillary growth than those with small clefts. Therefore, further randomized control studies comparing the effects of PSIO within different orthopedic and surgical techniques are needed for assessing long-term results [53].

Another surgical issue is completing the osseous union at the cleft region at the time of primary lip closure, which may affect maxillary growth, and therefore, influence the outcome of presurgical infant orthopedics. Different methods have been used for this purpose, and some of them, such as primary alveolar grafting, have been abandoned because of the detrimental effects on maxillary and facial growth [54-56]. As minimal invasive methods such as GPP provide bone union, there is a debate regarding the effect of the combination of this technique with presurgical infant orthopedics. Two cleft groups were compared in a study; one group underwent presurgical alveolar molding followed by GPP at the time of lip repair, while the other group did not undergo molding and GPP. The authors investigated whether narrowing of the cleft parts and GPP diminished the need for bone-grafting later. The results of this study showed that all patients in the control group required bone grafts, while 60% of patients treated with presurgical orthopedics and GPP did not need a secondary alveolar bone graft in the mixed dentition [37]. However, although these benefits have been stated by many authors, there is no consensus regarding the utility of GPP or secondary alveolar bone grafting [37]. Another issue is that most of the anterior growth of the maxilla takes place by the age of six years [57]. Similarly, Wood et al. were unable to demonstrate any clear impairment of maxillary growth in patients treated with GPP compared to patients not treated with this technique [58].

Complications

Some studies have reported complications in soft and hard tissues using PNAM therapy [59-61]. Grayson and Maull reported some problems including soft tissue breakdown, intraoral ulcerations, and failure to apply tapes and elastics, cooperation issues, and the eruption of neonatal teeth during treatment. They reported that common areas of breakdown were the frenulum attachments, the anterior premaxilla, or the posterior fauces, as the molding plate is retracted. They also reported that the intranasal lining of the nasal tip can become inflamed if too much force was applied by the upper lobe of the nasal stent [61]. The other most frequent problem was the development of cheek skin rashes [40]. In the study of Lewy-Bercowsky et al., soft and hard tissue complications were mentioned. Contact dermatitis due to repeated removal of tapes, meganostril produced by improper positioning of the nasal stent, overactivation of the nasal stent resulting in bruises or petechiae in the dome area were mentioned as soft tissue complications. Neonatal teeth eruption during treatment, or premature eruption of the incisors due to the pressure exerted by the acrylic plate, which creates a T-shape maxillary arch after the use of the molding plate, were reported as hard tissue complications [61].

In a unique study, the effects and complications of two PSIO treatment methods were compared. The authors stated that both Grayson and Figueroa nasoalveolar molding improved nasal deformities, and reduced alveolar gaps in a similar manner; however, the Figueroa technique was associated with fewer oral mucosal complications and better efficiency [62].

Opinions Against Presurgical Orthopedics

Despite the fact that the usefulness of these methods has been pointed out, presurgical orthopedic procedures have been stated as unnecessary in some studies of the Eurocleft project. One of these

studies was performed by Kuijpers-Jagtman and Prahls Andersen analyzing neonatal orthopedics of the Zurich approach, for over 20 years. According to their longitudinal observations, neonatal orthopedics is not the best approach. They conducted a randomized clinical trial named "Dutchcleft" in three centers, and compared infant orthopedics and non-orthopedics groups in relation to general, orthodontic, and cost effectiveness, as well as speech effects of these approaches. Regarding general effects, there was no difference between the groups in weight for age, length for age, or weight for length. When they assessed the maxillary arch form and dimensions, they found that cleft gap was reduced significantly in the orthopedic group; however, no significant differences were found between the groups after lip closure [7]. Furthermore, Prahls et al. found that infant orthopedics did not prevent collapse of the maxillary arch [14]. Therefore, in the Dutchcleft study there were no observable effects on occlusion and jaw relationships at the ages of 4 and 6 years [63]. Evaluation of speech and language development showed that at one year of age, children who wore plates presented an enhanced production of alveolar sounds; however, at the age of 1.5 year, when the plate was no longer used, a limited effect on speech was observed [64]. At 30 months of age, the phonologic development of the orthopedic group was normal or delayed, while most children in the non-orthopedic group presented an abnormal development [65]. Taking into account the results of the Dutchcleft trial, there is no need to perform infant orthopedics for unilateral cleft lip and palate.

Papadopoulos et al. also investigated the effectiveness of presurgical infant orthopedics using a systematic review [66]. They showed that there were no significant differences in craniofacial and dentoalveolar changes, indicating that PSIO treatment had no effect on cleft lip and palate patients. The limited evidence derived from this study does not seem to support the short- or long-term effectiveness of PSIO in these patients. Furthermore, Van der Heijden et al. performed a meta-analysis, and inferred that the results of studies on nasolabial molding were inconsistent in relation to changes in nasal symmetry, although there was a trend towards a positive effect [67]. In a similar manner, Uzel and Alparslan concluded in their systematic review that presurgical infant orthopedic appliances have no long-term positive effects in patients with cleft lip and palate and that more randomized controlled trials are necessary. They also added that the encouraging results on the effect of nasolabial molding appliances on nasal symmetry need to be supported by future randomized controlled trials [50].

Presurgical infant orthopedics has been investigated in terms of cost-effectiveness. The main principle of cost-effectiveness analysis is to estimate the cost and treatment outcome compared to an alternative treatment. The total cost of presurgical orthopedics was higher in the treatment group of the Dutchcleft study [68]. In this study, the mean medical cost for infant orthopedics treatment was US\$852. The non-orthopedics treatment group had a significantly lower mean medical cost (US\$304). Mean travel costs and indirect nonmedical costs were US\$128 and US\$231 for the orthopedics, and US\$79 and US\$130 for the non-orthopedics groups, respectively. However, the additional cost of neonatal maxillary orthopedics might be partly outweighed by the costs in speech therapy in later years, as the group treated with neonatal orthopedics had a significantly better rating for speech [69]. Based on the results of the Dutchcleft study, the authors concluded that neonatal maxillary orthopedics for unilateral cleft lip and palate is not necessary for feeding, patient's satisfaction or orthodontic reasons. Regarding speech, a positive but very limited effect was found until the age of 2.5 years.

Apparently, studies concerning presurgical orthopedics in cleft lip and palate have been heterogeneous and lacked adequate reporting. In particular, surgical time and sequence of surgery were stated as decisive factors for the final success, rather than the presurgical orthopedic

treatment type [7]. It should be kept in mind that the best surgery approach for these patients was not described, and the outcomes were affected not only by presurgical orthopedics, but also by surgery methods [70-74]. There are many surgical alternatives as there are many types of orthopedics. Future prospective longitudinal studies are needed to achieve a consensus on the effect of presurgical orthopedics, as well as the best treatment approach.

Conclusion

It can be inferred from this review that presurgical orthopedic appliances are useful for aligning cleft segments, reducing soft tissue tension and improving nasal aesthetics. Although in some investigations it was found that there were no differences between the groups that underwent presurgical infant orthopedics and those who did not; there is a trend towards a positive effect on nasal symmetry with the use of the PNAM appliance. Assessments on the effects of different combinations of cleft surgery and orthopedics methods are still needed. Therefore multidisciplinary treatment modalities are of great importance for the rehabilitation of cleft patients.

Conflict of interests

Authors have no conflict of interests to disclose.

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