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Review Article

Current status of presurgical infant orthopaedic treatment for cleft lip and palate patients: A critical review

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ABSTRACT

Rehabilitation of cleft lip and palate (CLP) patients is a challenge for all the concerned members of the cleft team, and various treatment modalities have been attempted to obtain aesthetic results. Presurgical infant orthopaedics (PSIO) was introduced to reshape alveolar and nasal segments prior to surgical repair of cleft lip. However, literature reports lot of controversy regarding the use of PSIO in patients with CLP. Evaluation of long-term results of PSIO can provide scientific evidence on the efficacy and usefulness of PSIO in CLP patients. The aim was to assess the scientific evidence on the efficiency of PSIO appliances in patients with CLP and to critically analyse the current status of PSIO. 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. The documented studies report no beneficial effect of PSIO on maxillary arch dimensions, facial aesthetics and in the subsequent development of dentition and occlusion in CLP patients. Nasal moulding seems to be more beneficial and effective in unilateral cleft lip and palate patients with better long-term results.

KEY WORDS

Bilateral cleft lip and palate; long-term results; nasoalveolar moulding; presurgical infant orthopaedics; unilateral cleft lip and palate

INTRODUCTION

Facial clefting is the second most common congenital deformity. Over the years, various treatment modalities have been attempted in these patients so as to achieve a satisfactory outcome. Infant orthopaedic (IO) treatment was first introduced by McNeil[1] and was further improved by others.[2-6] From McNeil's concept of alveolar moulding to concept of nasoalveolar moulding (NAM) many changes have also taken place in appliance design. In 1999, Grayson *et al*.[7] described a new technique to presurgically mould the lip, alveolus and nose in infants born with cleft lip and palate (CLP). The concept of NAM works on Matsuo's principle;[8] that the nasal cartilage could be moulded due to increased plasticity concurrent to increased levels of maternal oestrogen, if treatment is initiated within 6 weeks of life. Presurgical nasoalveolar moulding (PNAM) appliances have been in use as a new

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approach to traditional presurgical infant orthopaedics (TPSIO).[7,9-12] The NAM appliance consists of an intraoral moulding plate with nasal stents to mould the alveolar ridge and nasal cartilage concurrently.

Some of the major advantages of TPSIO are claimed to be the improvement in arch form, facilitation of surgical closure, and thus improvement of aesthetic outcome, facilitation of feeding, and improvement of speech.[13-18] Advocates of PNAM have stated that, beside other advantages of traditional plates, the main objectives of PNAM appliances are improving nasal symmetry and lip aesthetics while elongating the columella and correcting nasal cartilage deformity.[7,10,19-22] On the other hand, opponents have stated that all types of presurgical infant orthopaedics (PSIO) approaches are complex and expensive and might have an adverse effect on maxillary growth. None of these claims are evidence based.[23,24,16,17] As there is not yet a definitive conclusion on the subject, a review of PSIO seems to be warranted.

Evaluation of long-term outcomes of different treatment protocols has become more valuable because it is well known that the definitive outcome of treatment in patients with CLP cannot be established until facial development is complete.

The aim of the present review is to assess the scientific evidence on the efficacy of PSIO appliances in patients with CLP and to critically analyse the current status of PSIO.

METHODS

A PubMed search was performed using the terms PSIO, PNAM and long-term effects and related articles were selected for the review. As the purpose of this review was to assess the long-term effects of PSIO in CLP patients, no efforts were made to study the effects of PSIO on unilateral cleft lip and palate (UCLP) and bilateral cleft lip and palate (BCLP) patients separately. A minimum 3-year follow-up was decided, and no restrictions were made regarding the type of PSIO appliance. Similarly as surgical times and techniques affect treatment outcomes, studies on PSIO using control groups treated with different surgical methods also were not included in the study.

Long-term effects of PSIO can be studied as under[Table 1]:

- Effects of PSIO on maxillary growth
- Effects of PSIO on dentition and occlusion
- Effects of PSIO on facial appearance
- Effects of PSIO on nasal symmetry
- Effects of PSIO on speech.

Long-term effects of presurgical infant orthopaedics on facial growth, maxillary arch, and dentition and occlusion

Studies, published in the 80s and 90s, dealt mainly with the effect of PSIO on maxillary arch dimensions with little attention on other aspects of the maxillofacial growth. Most studies, however, were retrospective, had a small sample size, lacked a control group with UCLP without PSIO, had no clear outcome measures, or did not take confounding variables into account.[25-33] Only a few studies could be found that had an adequate research design to investigate the effects of PSIO.

Mishima *et al*.,[34-36] using a two-group quasirandomised design with a control group ($n = 8$) children without PSIO, found that prior to lip surgery the maxillary segments in the PSIO group ($n = 12$) moved towards the midline, and in the non-PSIO group, the maxillary segments remained in the same position or displaced laterally. At 18 months of age, the curvature of palatal shelves was less steep, and there was less arch collapse in PSIO group when compared with control group. At 4-year of age, the distance between deciduous canines and second deciduous molars was larger in the PSIO group; other variables did not differ between the two groups.

A three-centre, randomised, prospective clinical trial in Netherlands.[37,38] (Dutchcleft) evaluating the effects of PSIO showed comparable differences in maxillary arch dimensions between PSIO ($n = 27$) and the non-PSIO group ($n = 27$) until lip surgery at 15 weeks of age. However, at 12 months of age (prior to soft palate closure), no differences with respect to maxillary dimensions existed anymore between the two groups.

In the Dutchcleft studies, facial growth, maxillary arch dimension, and occlusion were further assessed at the age of 4 and 6-year.[39-41] No significant differences were found in any of the variables between the (IO^+) and (IO^-) groups when the occlusion was assessed by 5-year-old index and Huddart score. Arch width, arch depth, arch length, arch form, and the vertical position of the lesser segment were measured. Authors found no clinically significant differences between (IO^+) and (IO^-) for any of the variables.

When comparing facial growth and occlusion, the centres that practiced PSIO did not show demonstrable advantages in patients with UCLP treated with different types of PSIO appliances.[26,42-44]

Lee *et al*.[45] assessed the effects of NAM and gingivoperiosteoplasty (GPP) in 20 UCLP patients at 6-year and at 11.5-year that is, at pre-pubertal age using lateral cephalogram and found that midface growth in sagittal or vertical planes (up to the age of 9-13 years) was not affected by pre-surgical alveolar moulding and GPP.

Adali *et al*.[46] studied the effect of PNAM on arch circumference and arch form in transverse, anteroposterior, and vertical dimensions on study model sets of 75 UCLP infants using Reflex Microscope and concluded that pre-surgical orthopaedics did not produce any significant effect on the arch form.

Ringdahl (2011)[47] studied the long-term effects of NAM on mid face growth in 28 UCLP patients using photographs, study models and lateral cephalograms and concluded that there was no significant difference between moulding and nonmoulding groups in Goslon score.

Ross and MacNamera[13] analysed the long-term effect of PSIO on facial aesthetics in 20 complete BCLP patients using facial photographs and concluded that PSIO has no lasting effect on lip, nose and facial aesthetics and does not alter the need for subsequent revisionary surgery.

The Dutchcleft studies[39-41] also concluded that there was no clinically relevant effect of IO with passive plates on facial growth until the age of 6-year.

Long-term effects of presurgical infant orthopedics on nasal symmetry and nasolabial appearance

The correction of nasal deformity continues to be the greatest challenge in CLP cases.[4] It is complex and affects the shape of the nose in all three planes of space. Matsuo *et al*.[8] and Matsuo and Hirose[48] were the first to describe pre-surgical moulding of nasal cartilage in the neonate. Grayson *et al*. developed an appliance with a nasal extension attached to the anterior portion of an acrylic alveolar moulding plate, which marked the advent of PNAM appliances. Grayson *et al*.[49,50] designed their nasal stent to extend from the anterior flange of an orthopaedic appliance used to mould the cleft alveolar segments. The resultant effect was NAM appliance and could be inserted as early as possible after birth. It is suggested that the main difference of NAM from traditional presurgical infant orthopaedics is the reshaping of nasal cartilage and providing aesthetic benefits in terms of nasal tip and alar symmetry.[7,9-12] Thus, nasal symmetry was investigated in most of the studies on NAM.[59]

In a 6-year follow-up by Bennun *et al*.[51] to compare growth and cosmetic results of 97 UCLP patients using plaster models by using surface impressions of the babies revealed a better and permanent nostril symmetry, increase in the columellar length and no alar cartilage luxation in patients who had the nasal component.

Maull *et al*.[9] evaluated long-term effects of NAM on three-dimensional nasal shape in unilateral clefts and found a significant change in nasal symmetry that was also maintained long term in early childhood. However, a major limitation of this study was that the children were not fully grown, and the control group was not age matched.

Liou *et al*.[19] assessed the progressive changes of nasal symmetry with growth after NAM in 25 UCLP patients at the initial visit (T1), after NAM (T2), 1 week (T3), 1-year (T4), 2-year (T5), and 3-year (T6) after cheiloplasty using standard basilar view photographs. Nasal symmetry was assessed by the "quantity of asymmetry" (in millimetres), that was the linear difference of each measurement between the cleft and non-cleft (cleft–non-cleft). The quantity of asymmetry revealed that nasal symmetry improved after NAM (T1–T2) and further corrected after cheiloplasty (T2–T3), but relapsed in 1st year (T3–T4) and then remained stable in 2nd and 3rd years after surgery (T4–T6). Nasal symmetry was acceptable after 3-year post-operatively. The relapse in nasal asymmetry was the result of a significant differential growth/relapsed between the cleft and non-cleft sides in the 1st year post-operatively. On the cleft side, the growth of nostril height and nasal dome height was significantly less, the columella length shortened (relapsed) significantly, and the growth of nostril width and nasal basal width was significantly less than on the non-cleft side. The relapse stopped, and the nasal growth was the same between the cleft and non-cleft sides in the second and 3rd year. To compensate for relapse resulting from the differential growth in the 1st year post-operatively, the authors have recommended narrowing down the alveolar cleft by NAM, overcorrecting the nasal vertical dimension

surgically, and maintaining the surgical results by using a nasal conformer.

Lee *et al*.[20] presented a series of 26 patients with bilateral CLP treated with a NAM protocol and stated that non-surgical columellar elongation with NAM followed by primary retrograde nasal reconstruction restored columellar length to normal by 3-year and significantly reduced the need for nasal reconstruction beyond their initial repair

A 9-year follow-up by Barillas *et al*.[52] using stone cast measurements in 25 UCLP patients revealed that the nasal symmetry was improved by PNAM and was maintained at 9-year.

Sulaiman *et al*.[53] did a 15-year follow-up of pre-surgical orthopaedics, followed by primary correction for unilateral cleft lip nose in program SEHATI in Indonesia and found that the nostril height and width ratio and the height of the alar groove were significantly improved post-operatively and maintained for 15-year, but also stated that repositioning of nasal cartilage at infancy might not eliminate need for secondary correction after puberty.

A meta-analysis performed to study the effect of PNAM in unilateral cleft on nasal symmetry by van der Heijden *et al*.[54] revealed that the results of NAM were inconsistent regarding changes in nasal symmetry; however, there was a trend towards a positive effect.

Long term effects of presurgical infant orthopedics on speech

Karling *et al*.[55] evaluated the effect of T traction on speech in unilateral, bilateral cleft patients comparing them with the control group of non-cleft subjects and found no significant differences between the groups. However, the difference between mean ages of the groups (10.6-year for[IO$^+$] and 17.6-year for[IO$^-$]) should be taken into consideration when evaluating this study.

Suzuki *et al*.[56] studied the effect of pre-operative orthopaedic plate on articulatory function in 17 CLP children. Speech was assessed by speech therapists 3-year 11 months after palatoplasty. The results indicated that the continuous use of orthopaedic plate was effective in preventing palatalized articulation.

Konst *et al*.[57] evaluated language skills of children between the ages of 2 and 6-year. The results showed that the early positive effects of PSIO changed over time, and no differences were found in language development between the two groups at the age of 6-year. However, the low follow-up rate reduced the level of evidence of this randomised controlled trial (RCT).

DISCUSSION

The effects of PSIO remains a subject of controversy in the medical literature. In the current review, the long term effects of PSIO treatment were examined, mainly with respect to the passive type of appliances. From the literature reports on the effects of PSIO on maxillary arch, dentition and occlusion, it seems that PSIO has no positive effect on maxillary arch dimensions and also does not improves the dentition and occlusion of UCLP children. The effects of PSIO on speech and language development also seems to be inconclusive although maxillofacial growth and speech are greatly influenced by the type, time and number of surgical procedures and also by the skill of performing surgeon. The only reported systematic review on effects of PSIO and its long-term advantages was released by Uzel and Alparslan.[58] The authors concluded that until the age of 6, there were no positive effects on factors like facial growth, maxillary arch dimension, or occlusion when treatment included passive IO appliances. However, the authors made the distinction between PSIO and NAM appliances, stating that their review yielded the conclusion that nasal symmetry was improved with NAM. A systematic review by van der Heijden *et al*.[54] to quantify the long term effect of PNAM on nasal symmetry in unilateral cleft also reported a trend toward a positive effect of PNAM on nasal symmetry. However Chang *et al*.[59] suggested that NAM alone could not provide nostril symmetry in the long-term and that overcorrection of 20% maintained nostril height after 5-year. Use of post-surgical nasal splint appliances for at least 6 months post-operatively have been advocated by Yeow *et al*.[60] and Chang *et al*.[59] to prevent relapse following NAM. These nasal splints help maintain the alar cartilage height and prevent collapse during scar healing and beyond. Nasal and facial anatomy and texture corresponding to ethnic descent, could be an important factor for remodeling a nose in the desired shape. Similarly skills of the dentist, orthodontist, and surgeon could also be a decisive factor in achieving improved nasal symmetry.

The Eurocleft study[61] showed that centres with better outcomes were seen when there were few surgeons with

Table 1: Clinical summaries of the retrieved articles

Article	Outcome	Subjects	Age at evaluation	Follow-up	Appliance	Method/ measurement	Conclusion
Facial growth, maxillary arch, and dentition and occlusion							
Ross and MacNamera (1994)[13]	Facial appearance	40 BCLP, 20 (IO^+) */20 (IO^-)*	15.6/14.8 years		Passive plate + rubber band	Photographs, JS score assignment (rating scale and comparison ranking 1-10)	PSIO has no lasting effect on esthetics of lip and nose and facial esthetics and does not alter the need for subsequent revisionary surgery
Mishima *et al*. (2000)[15]	Maxillary arch dimension	20 UCLP, 10 (IO^+) *and 8 (IO^-)*		4 years follow-up	Hotz plate (passive appliance without extraoral strapping)	Serial study models of palate measured three dimensionally	No difference was observed in alveolar arch form, degree of palatal surface curvature, and anteroposterior distance of palate between the two groups at 4 years age
Lee *et al*. (2004)[45]	Midface growth	20 UCLP, 10 (NAM + GPP) 10 (NAM− GPP)	Prepubertal age (11.5 years)		NAM	Lateral cephalograms	Midface growth in sagittal or vertical planes (up to the age of 9–13 years) was not affected by presurgical nasoalveolar molding and GPP
Bongaarts *et al*. (2006)[40] (Dutchcleft)	Maxillary arch dimensions	45, UCLP, (IO^+)=4 years (*n*=23), 6 years (*n*=22), (IO^-)=4 years (*n*=22), 6 years (*n*=23)		4 and 6 years follow-up	Passive (passive appliance without extraoral strapping)	three-dimensional cast analysis	All significant differences between the two groups faded away at the age of 6 years. PSIO has no clinically significant effect on the maxillary arch dimension or on the contact and collapse scores
Bongaarts *et al*. (2009)[41] (Dutchcleft)	Facial growth	43 UCLP (IO^+)=4 years (*n*=21), 6 years (*n*=21), (IO^-)=4 years (*n*=20), 6 years (*n*=22)		4 and 6 years follow-up	Passive (passive appliance without extraoral strapping)	Lateral cephalogram, 23 variables	No clinically relevant effect
Muhammad Syafrudin Hak002C (2012)[63]	Maxillary growth	53 complete BCLP patients divided into three groups: 11 patients treated without Hotz's plate), 24 treated with Hotz's plate; and 18 treated with lip adhesion and Hotz's plate compared to 10 noncleft patients		5 years follow-up	Hotz plate with extraoral strapping	Dental casts	Treatment using Hotz's plate prevented collapse of the premaxilla, and the growth of the arch length was comparable to that observed in the noncleft group

(*continued*)

Table 1: (Continued)

Long-term effects of PNAM on nasal symmetry and nasolabial appearance						
Bennun (1999)[51]	Nasal symmetry	91 UCLP patients, (*n*=44) treated with occlusal plate, (*n*=47) treated with nasal component added to occlusal plate compared to 6 years old healthy control group		6 years follow-up	NAM	Anthropometric measurements on plaster models	Better and perma nent nostril symmetry, increase of columellar length and no alar cartilage luxation in patients who had nasal component
Maull *et al*. (1999)[9]	Nasal symmetry	10 UCLP patients with PNAM, 10 with only alveolar moulding without nasal stent	4.6 years for NAM, 9.1 years for control		PNAM	Nasal casts digitized 3-dimensionally by virtuose shape camera	Significant ↑ in nasal symmetry maintained long-term in early childhood
Liou *et al*. (2004)[19]	Nasal symmetry	25 UCLP patients		3 years follow-up	NAM	Standard basilar view photo graphs	Nasal symmetry improved after NAM and further corrected after cheiloplasty but relapsed in 1st year and then remained stable in 2nd and 3rd years after surgery acceptable after 3 years postoperative
Lee *et al*. (2008)[20]	Nasolabial appearance	26 BCLP, Group 1 (*n*=13) cleft repair + nasal correction, group 2 (*n*=13), NAM followed by lip repair and primary retrograde nasal correction, group 3 (*n*=130) age matched controls		3 years follow-up	NAM	Photometric technique to evaluate columellar length and postsurgical nasal results by clinical assessment by a board-certified plastic surgeon, a craniofacial orthodontist, and a prosthodontist	Nonsurgical columellar elongation with NAM followed by 1° retrograde nasal reconstr-uction restored columellar length to normal by 3 yearsandsignificantly reduced the need for secondary nasal surgery
Barillas *et al*. (2009)[52]	Nasal symmetry	25 UCLP, 15 patients treated with NAM/10 treated with surgery alone without NAM		9 years follow-up	NAM	Stone cast measurements. Four nasal anthropometric distances and two angular relationships were measured to assess nasal symmetry	The lower lateral and septal cartilages were more symmetric in the nasoalveolar molding patients compared with the surgery-alone patients. The improvement in nasal symmetry by PNAM was main-tained at 9 years

Table 1: (Continued)

							Suggests a trend toward a long-term clinical improvement in nasal and lip anatomy of UCLP patients treated with PNAM
Sulaiman *et al.* (2013)[53]	Nasolabial appearance and nasal symmetry	17 complete UCLP patients	1-year, 8±1 year, 15 years postoperatively	15 years follow-up	Hotz plate	Color photos of frontal and basal view taken serially 1-year, 8±1 year, 15 years postoperatively	The nostril height and width ratio and height of alar groove were significantly improved postoperative and maintained for 15 years
Long term effects of PSIO on speech							
Karling *et al.* (1993)[55]	Speech	84 UCLP, 45 (IO^+)*/39 (IO^-)*	13.7 years		T traction	Speech recording listener judgments	No significant differences in articulation or resonance found between groups
Konst *et al.* (2003b)[57] (Dutchcleft)	Speech	12 UCLP, 6 (IO^+)*/6 (IO^-)*	2, 2.5, 3, 6 years	6 years follow-up	Passive (passive appliance without extraoral strapping)	Speech recording, Reynell developmental language scales	PSIO had no long-lasting effects on language development
Suzukl *et al.* (2006)[56]	Speech	17 (IO^+)*, 91 (IO^-)*	4 years 1-month to 6 years 6 months	5 years 4 months after palatoplasty	Orthopaedic plate	Speech assessed by speech therapists	Continuous use of orthopaedic plate appeared to be effective in preventing palatal zed articulation

*Infant orthopaedics; PSIO: Presurgical infant orthopaedics; GPP: Gingivoperiosteoplasty; NAM: Nasoalveolar moulding; PNAM: Presurgical nasoalveolar molding; UCLP: Unilateral cleft lip and palate; BCLP: Bilateral cleft lip and palate; IO: Infant orthopedics

some differences in technique and timing, single layer cranially based vomer flap to close the anterior palate at the time of lip repair, no pre-surgical orthopedics, and delayed closure of hard palate at age 9. Conversely centres with lower ranking utilized active pre-surgical orthopedic treatment (extraoral strapping and nostril retraction, had many surgeons, utilized primary bone gragting and performed secondary revisions early. Similarly nearly identical conclusions were reached in the Americleft study[62] regarding the association of more favorable outcomes with standard, simpler, and less burdensome protocols without PSIO, without primary bone grafting, and fewer surgeons.

Very few studies evaluating the long term effects of PSIO in BCLP patients can be found in the literature due to the low prevalence of BCLP. Bilateral cleft lip – cleft palate NAM is designed for nonsurgical columella elongation, orthopedic retraction of pre-maxilla and molding of the posterior lateral alveolar ridges to an appropriate width to accept the pre-maxilla. 3 long term studies evaluating the efficacy of PSIO in BCLP patients have been included in the review out of which Ross and MacNamera[13] reported that conservative PSIO for BCLP patients does not have lasting effects on the esthetics of the lip and nose while Lee *et al*.[20] stressed that nasoalveoalr molding combined with retrograde nasal reconstruction improves the quality of reconstructive outcome and decreases the number of surgical procedures. Hak *et al*.[63] in a prospective longitudinal evaluation on 53 BCLP patients also showed that treatment with Hotz's plate until about 18 months of age prevented collapse of the pre-maxilla and supported the growth of dental arch length until the age of 5-year compared to non-cleft patients.

Thus unfortunately, at this time there is not a good body of evidence to support a statement either for or against using PSIO. However, a trend in the literature is being seen to support nasal moulding, but long-term studies are needed to analyse whether nasal moulding truly reduces the need for future nasal revision or other health care costs with age.

Limitations of the review

One of the greatest problems faced in PSIO research is small sample size, due to the relatively low prevalence of UCLP. Multicentre studies have aimed to overcome this by pooling different samples, thereby introducing additional problems with comparisons, especially regarding the heterogeneity of samples and variations in the number and experience of surgeons (Roberts-Harry *et al*.[64] Prahl *et al*.[16] The Euro cleft studies[44] have the longest follow-up in the literature (17-year), but direct comparison between ($PSIO^+$) and ($PSIO^-$) could not be made in the intercentre studies due to the differences in the treatment protocols. Thus, the effects of PSIO in adults remain unclear. Moreover, each study used a unique combination of population and treatment duration, which makes comparison of every single aspect of treatment impossible. In addition, this meta-analysis has not taken into account the different techniques of PSIO, timings of surgery and surgical techniques, which would have made comparison of the results even more difficult. All these aspects together have made conclusions of the results of the studies by means of the intended meta-analysis impossible.

CONCLUSION

The current review article is an attempt to critically analyse the literature regarding the effects of PSIO and to comment on the current status of PSIO. The documented studies report no beneficial effects of PSIO on maxillary arch dimensions and in the subsequent development of dentition and occlusion in UCLP patients. The studies on the effect of PSIO on speech are also inconclusive. Based on the literature review, nasal moulding seems to be more beneficial and effective with better long term results, however the effects on nasal and alveolar moulding needs to be studied further to assess the long term beneficial effects. In the future, well-designed RCTs with long term follow-up should be undertaken in order to provide additional evidence to confirm or reject PSIO effectiveness.

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