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Controversies in cleft lip & palate: A literature review

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Abstract

Orthodontics is the branch where great pioneers exist and they come with varying opinions leading to controversies. Nasoalveolar molding (NAM) is a form of presurgical infant orthopedics (PSIO) whose goal is to reduce the severity of the cleft deformity and improve surgical outcomes. The data on efficacy and benefit of NAM are mixed. Those in favor of NAM report improved nasal symmetry and appearance, reduced overall costs, psychosocial benefit to family, and decreased need for early nasal revision. Those in opposition to NAM argue NAM causes increased caregiver burden, poor patient compliance, increased costs in the short term, relapse of nasal symmetry in the first year, and no conclusive data of improved outcomes.

For cleft palate repair controversy exists with timing of cleft repair, each with its own advantage and disadvantage with no conclusive results. These two controversies have been discussed here.

Keywords: Presurgical orthopaedics, nasoalveolar molding, cleft palate repair

Introduction

A controversy can prove unsolvable if the specialty fails to differentiate between real data and uninformed, self-serving opinions. Cleft lip and palate is one of the most common congenital craniofacial disorders. Management of CL+/- CP involves a multidisciplinary approach. Thus management of CL+/- CP involves many controversies.

Various controversies associated with CL+/- CP

- Presurgical orthopaedics – should be done or not
- Timing of Alveolar Bone Grafting and Palatal Repair.

Nasoalveolar moulding

In the late 1990s, Grayson and Cutting described a presurgical appliance that added additional nasal molding prongs [1]. The NAM technique uses acrylic nasal stents attached to the vestibular shield of an oral molding plate to mold the nasal alar cartilages into more normal form and position during the presurgical period.

- It non-surgically improves congenital deformity by taking advantage of plasticity of infant cartilage and increased level of estrogen in maternal estrogen in 6 starting 6 months of life.
- It expands the columellar length by tissue expansion by gradual expansion of nasal stent.

The process begins immediately after birth; a custom-made alveolar mold is worn full time with weekly or biweekly adjustments until the alveolar cleft is narrowed to 5 mm or less, achieving the goals of bringing the lip segments together, reducing the alar base width, and relaxing tension on the splayed alar rim. A nasal stent extending from the intraoral plate to the cleft nostril is then added to elevate the alar rim into a more symmetric and convex position. NAM is used at least occasionally by 38% and greater than half the time by 25% of surgeons. This increase in use in the past 20 years raises the question, Is NAM effective?

Proponents

Clinicians favoring NAM suggests aesthetic improvement, psychological benefit to the parents and reduced overall cost. To evaluate the psychological benefits for the patients a prospective longitudinal study was performed which included 118 caregivers divided into 2 groups. Group 1 treated with NAM plus traditional care & group 2 with traditional care only. Qualitative and quantitative results, however, indicated caregivers of NAM-treated infants experienced more rapid declines in anxiety and depressive symptoms and better coping skills over time than caregivers whose infants had traditional care. Frequent visit for NAM adjustments reduce caregiver's anxiety and increases strength [2].

Studies in support of nam

A study from India by Sabarinath and colleagues examined pre-NAM and post-NAM study models of 10 unilateral cleft lip-palate infants. Results from this study showed that NAM was effective in reducing the severity of the initial cleft deformity mainly at the anterior portion of the maxillary arch, because it increased the arch perimeter and intertuberosity distance while decreasing the width of the alveolar cleft and anterior arch length and width [3].

Bennun and colleagues performed a prospective controlled trial of unilateral cleft lip and palate patients. They compared 44 patients managed with an occlusal prosthesis plus NAM to 47 patients who had an occlusal prosthesis alone as well as 48 noncleft controls. The results at 6 years showed the nasoalveolar group exhibited greater nasal tip protrusion, columellar length, and columellar width compared with the non-NAM group, and tip protrusion was similar to that of non cleft controls. Both the NAM and non-NAM groups had greater nasal width than the non cleft controls [4].

Another retrospective study on unilateral cleft lip-palate by Chang and colleagues compared primary rhinoplasty alone, NAM alone, NAM plus primary rhinoplasty, and NAM plus primary rhinoplasty with overcorrection. Photography taken 5 years after surgery showed no difference in nostril height, nostril width, nostril area, or nostril height-to-nostril width ratio. There was a significant difference seen in the NAM groups showing improved symmetry in the medial nostril height and nasal sill height [5].

Studies not supporting nam

Although there are some studies that indicate a positive trending effect with NAM, there are many studies with mixed or negative results. Clark and colleagues retrospectively compared 20 patients who underwent primary lip repair after NAM to 5 patients who underwent primary lip repair alone. Neither group had primary nasal repair at time of surgery, and analysis was performed at 5 years and 6 years, respectively. Study patients showed no difference in the measures of philtrum, lip scar, nasal anatomy, dental arch, nasal measurements, lip measurements, and maxillary arch analysis [6].

Opponents of NAM argue that studies show no significant effects on maxillary growth, dentition, or occlusion. Lee and colleagues assessed the effects of NAM and gingivoperiosteoplasty in 20 unilateral cleft lip-palate patients and found that mid face growth in sagittal or vertical planes (up to the ages of 9–13 years) were not affected by presurgical alveolar molding and gingivoperiosteoplasty [7]. Adali and colleagues studied the effect of NAM on arch circumference and arch forming 75 unilateral cleft lip palate patients and found that presurgical orthopedics produced no

statistically significant mean change in any arch form variable when compared with the non-PSO group. Lip repair produced greater change in arch form than did presurgical orthopedics, reducing the mean alveolar cleft width by 4.45 mm compared with 0.69 mm by NAM [8].

Effect of nasoalveolar moulding on speech development

Effect of presurgical orthopedics on speech development has shown inconclusive results. Suzuki and colleagues evaluated 17 patients with unilateral and bilateral cleft lip and palate. Analysis of tongue movements was by ultrasound and speech therapist analyzed articulation approximately 4 years after palate repair. They found that infants with cleft palate could not create negative pressure in the oral cavity, even with the presurgical orthopedics. Continuous use of the orthopedics up to the time of palatoplasty seemed effective for the postoperative articulatory, inhibiting irregular movements of the tongue, and possibly preventing "palatalized articulation [9]."

In a prospective randomized clinical trial (Dutch cleft), patients with complete unilateral cleft lip palate were followed-up longitudinally at 2 years, 2.5 years, 3 years, and 6 years. One group of 6 infants was treated with IO in the first year of life; the other group of 6 did not receive this treatment. Phonological skills, such as number of acquired consonants, order of phonological development, use of phonological processes, and occurrence of nasal escape, were evaluated. At age 3, the children in the IO group had acquired more initial consonants, suggesting children treated with IO during their first year of life followed a more normal path of phonological development between 2 years and 3 years of age [10]. The trial also examined receptive language skills and expressive language skills. Children treated with IO during their first year of life produced longer sentences than non-IO children at the ages of 2.5 and 3 years. At 6 years of age, however, both groups presented similar expressive language skills. Hence, IO treatment did not have long-lasting effects on language development [11].

Current view

Several review articles have been written on this topic but with inconclusive results. The Eurocleft and Amercleft studies attempted to pool data to come up with some conclusions but also produced mixed results. The Eurocleft study was a longitudinal cohort study between 5 different cleft centers in Northern Europe that examined patient results at 9 years, 12 years, and 15 years of follow-up. They examined many different aspects of cleft care, including nasolabial appearance. In this study, they found that the 2 centers with less favorable ratings had a more complex treatment program, including presurgical orthopedics. They also cited difficulty, however, with the rating system, such as low inter examiner agreement and variability in the quality of the photographs. The Amercleft study compared data from 4 North American cleft lip and palate centers at patients ages 6 and 12. In this study, they found no significant differences in patient appearance despite differences in protocols, including presurgical orthopedics.

This study had better inter examiner agreement but was also limited by the consistency of the photography. There is poor agreement between studies, with some showing positive results, whereas others show negative or mixed results. This may in part be attributed to variations in protocol and technique of NAM application. Between studies, there is variation in timing, duration, adjustments, and differences in

appliances. Before NAM is applied, there is divergence in pretreatment protocols, with some institutions using taping and others using NAM as the first intervention. Regarding timing, studies varied in when the patients first obtained the NAM appliance, beginning as early as 7 days old and up to 12 weeks of age. This relatively advanced age might have altered the results of these patients because the cartilages lose postnatal malleability during this time. Duration of device use varies between studies with time frames ranging from 2 months to 8 months. There is no standard protocol for adjustments, and the frequency of adjustment also varies. It is unclear if patients who had weekly visits and adjustments have better outcomes than those who had less frequent appointments. Furthermore, there are likely differences in the appliance used as well as variation in the skill and technique of the dentist and orthodontist. Finally, there may be a selection bias in many studies because the inadequate reporting of indications and the process of selection for NAM

Controversy for cleft palate repair

Present day corrective procedures involve surgery, the type and timing of the operation depending on whether the surgeon is a von Langenbeck soft tissue descendant, with or without presurgical maxillary orthopedics, or a Brophy “steel-clamp and silver wire” bony closure man. There is a basic philosophical conflict between the two major groups. Some surgeons employ staphylorrhaphy for closure of the cleft palate, a variation of the von Langenbeck, Furlow, or Veau-Wardell Kilner V-Y pushback procedure using mucoperiosteal flaps. They may or may not use vomer flaps to line the surface of the mucoperiosteal flap. The soft tissue is repositioned over what many surgeons consider a normal bony framework (with the exception of the cleft area) that had failed for some reason to unite in the midline. Today, most surgeons are convinced that aberrant embryonal and fetal influences force the lateral halves of the maxilla apart, making the intramaxillary width excessive. In the past, it was Brophy, today it is many McNeil disciples who utilize presurgical maxillary orthopedics, believing that the first step in habilitation is to restore what they believe to be a normal segmental relationship at a very early age. Each school of thought has intense convictions that there is only one correct approach – its own.

Four surgical approaches are commonly used to close the palatal cleft

1. Early complete palate repair (3–9months). Rationale: to achieve maximum speech results with a possible chance of inhibiting midfacial growth and creating severe dental occlusion. This approach favors speech above facial growth and development.
2. Delayed complete palate repair (12–24months). Rationale: speech results are nearly as good as with earlier repair and the facial growth disturbance is less.
3. Late complete palate repair (2–5 years). Rationale: to prevent facial-palatal growth inhibition, accepting the poorer speech results. Use of a palatal obturator is needed in most cases.
4. Early lip and soft palate repair (2–9months) and delayed hard palate repair (5–9 years). Rationale: to avoid facial and dental deformity but perhaps still achieve good speech with the aid of an obturator.

Surgery to the hard and soft palate with temporary disruption of the blood supply does not, by itself, cause damages to the

underlying bone. Most surgeons and orthodontists believe that the principal growth inhibitor seems to be the quantity and distribution of scar tissue that is created after surgery. When evaluating the effects of surgery on maxillary growth and development, it is necessary to consider that clefts of the palate can differ greatly in size and form at the same age due to the amount of osteogenic deficiency in the hard and soft palate and lip. The great individual variation in the relationship of the size and form of the cleft space relative to the size of the palatal segments is responsible for the differences in the amount of scar tissue formed even when the same surgical procedure is performed by the same surgeon. A review of cleft palate surgical history clearly shows that a single mode of surgery for all cases invariably resulted in severe palatal and midfacial deformities, as well as poor speech development. Unfortunately, the same poor results still occur despite the timing of surgery and the skill and experience of the plastic surgeon. This is so because of failure to define the criteria for the timing of palatal surgery and failure to agree on which surgical procedures interfere with normal growth and development of the structures involved. Poor results were understandable when there were no standardized methods for estimating success or recording the effects of surgery on speech and facial growth and development; these shortcomings no longer exist. Nonetheless, some present-day surgical reports still do not adequately describe the original deformity; thus, the efficacy of the surgical effort cannot be evaluated.

Mapes and coauthors [14], Robertson and Fish [15], and Berkowitz (unpublished data) all concluded that nontraumatic palatal surgery accelerated the growth rate of the maxilla, helping it reach more normal dimensions in the following years. Berkowitz and associates demonstrated that, in the patient with complete bilateral cleft lip and palate, after conservative palatal surgery the palatal surface area doubled from birth to the age of 1 1/2 years. Also, in an isolated cleft palate of a patient with Pierre Robin Sequence, there was a 50% increase in the palatal surface area from birth to 1 year of age. The acceleration in growth tapered off after palatal surgery in both instances. Palatal growth accelerates 6–12months post surgery in some cases [16].

Conclusion

It is absolute impossible to answer which is better as no conclusive results exists. Even the literature review does not give definite protocol for presurgical orthopaedics or timing of palate repair. Both varies depending on the treating group, type of defect. Each has its own advantage and disadvantage. The clinician has to judge the benefit over disadvantage and decide accordingly about the treatment protocol.

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