

Pre: Surgical orthopedic pre-maxillary alignment in bilateral cleft lip and palate patient

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Abstract

Pre-surgical orthopedic appliances are mainly used to retract and align the protruded and deviated pre-maxilla and to facilitate initial lip repair. This article presents a case report of a five year old male child patient with bilateral cleft lip and palate in whom a special custom made pre-surgical orthopedic appliance was delivered. Use of a special custom made presurgical orthopedic appliance for repositioning pre-maxilla in bilateral cleft lip and palate patient is discussed in this article.

Keywords: Bilateral cleft lip and palate, pre-maxilla, pre-surgical orthopedics

Introduction

Clefts of the lip and palate are the most common congenital deformities involving oro-facial region,^[1] with an incidence of one per thousand live births.^[2] Although cleft lip and palate are distinct and separate congenital abnormalities, they often occur concomitantly. Clefts may vary from minor notching of lip or bifid uvula to complete unilateral or bilateral cleft of the lip and palate. Exact cause of bilateral cleft lip and palate (BCLP) is unknown.^[3]

Watanabe *et al.*^[4] found that a missense mutation, 1355G>A, and one rare single nucleotide polymorphisms haplotype may play a role in the development of cleft lip and/or palate in the Vietnamese and cleft lip and/or palate and cleft palate only in the Japanese. Morand and Raphael^[5] described BCLP as the most serious anomaly because of the inherent disturbance of maxillary structures and because of the grave disturbances they inflict on the development of the middle third of the face.

Mulliken *et al.*^[6] reported that infants with complete BCLP appear different because they have nasolabiomaxillary hypoplasia and hypoplastic septum, small alar cartilages, deficient

basilar columella, underdeveloped contralateral philtral ridge, ill-defined Cupid's bow, thin vermilion-mucosa on both sides of the cleft, a more severely hypoplastic nasal tip, tiny prolabium and small/mobile pre-maxilla, which is displaced anteriorly and suspends from the tip of the nasal septum, whereas the nasal alae are stretching over the cleft. Because of the displacement, bilateral cleft anomaly is difficult to correct and camouflage.

Santiago *et al.*^[7] demonstrated that the use of pre-surgical infant orthopedic plate prior to initial lip repair helps in achieving alignment of pre-maxillary segment and elongation of the columella attributing to increased nasolabial mucosal lining surface area. Grayson and Cutting^[8] described that the pre-surgical nasoalveolar molding (NAM) and pre-maxillary alignment are known to facilitate initial surgical repair and reduce post-surgical scarring.

This case report aims at providing an overview of management of five-year-old bilateral cleft lip and palate patient using pre-surgical orthopedic appliance for positioning the pre-maxilla to a suitable position in order to facilitate initial lip repair.

Case Report

A 5-year-old male patient was referred to our department with parents complaining of difficulty in eating and also unhappy with the appearance of the child. Mother had a full term, normal uneventful pregnancy, and medical and dental history was non-contributory; family history revealed that parents were not of consanguineous marriage.

Extra-oral and intra-oral examination revealed complete bilateral cleft lip and palate with pre-maxilla protruded and deviated to the right side from the midline. Presence of primary dentition is seen; in the pre-maxilla, 3 anterior teeth were seen with missing primary lateral incisor on left side.

Consultation with the oral and maxillo-facial surgeon inferred that there were chances of failure of surgical lip closure due to tension if surgical procedure is carried out without retrusion

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of the protruding pre-maxillary segment. Hence, we planned for a custom made appliance that was used to retract the pre-maxillary segment in antero-posterior plane.

Appliance fabrication and design

Initial impression of the cleft lip and palate was made using fast setting alginate impression material. All the necessary precautions were taken during the impression procedure. A primary cast was prepared using dental stone, and undercuts were blocked. We prepared a custom-made orthopedic plate on the dental stone model incorporating an acrylic bulb covering the pre-maxilla. The pre-maxillary component of the acrylic was attached to the posterior component of the orthopedic plate by the use of two U loops of 21 gauges (0.7 mm) stainless steel wire, one on each side. The plate was made of self-cure acrylic resin of 2-3 mm thickness [Figure 1].

This innovative self-retentive plate comprised of two parts. The posterior part, palatal plate, covered the palatal defect and served as a passive obturator, it also covered canine and molar teeth completely in the posterior palatal halves on both the sides, which aided in anchorage, thus in retention of the appliance. The anterior part of the plate extended as a bulb around the protruded and deviated pre-maxilla. The acrylic bulb acted as an active part to align and retract the malposed pre-maxillary segment.

The appliance was fabricated, finished and polished, checked for any rough surfaces and then delivered to the patient [Figure 2]. On delivery appointment, parents were advised, instructed, and demonstrated for the removal and placement of the appliance and its cleaning. Parents were advised to ensure that the appliance is in place all the time.

A twice-weekly activation of the appliance was done by compressing the u loops about 1mm on either sides of the appliance. It was made sure that the pre-maxilla was not deviated inferiorly to the maxillary arch. Measurements were taken and tabulated at each appointment by recording fixed hard and soft tissue reference points to check the activation and movement of the pre-maxilla [Figure 3].

Considering deciduous 2nd molar to be constant in its position, the soft tissue reference points were taken from the soft tissue distal to 2nd primary molar to the prolabium to check the movement of soft tissue along with the pre-maxilla. The hard tissue reference points were taken from

1. Distal end of the 2nd primary molar to the distal end of lateral incisor on right and central incisor on left side, respectively, and
 2. From distal end of primary 2nd molar to midline and
 3. Mesial end of canine to distal end of lateral incisor on right and central incisor on left side, respectively, in pre-maxilla.
- This helped to know the movement of the pre-maxilla and closure of the alveolar cleft, respectively.

Over the treatment period, there was approximation and contact of the alveolar portion of the pre-maxilla and palatal vault on left side, thus the further movement of pre-maxilla is restricted [Figure 4]. The movement achieved was satisfactory and the patient was referred to department of oral and maxillo-facial surgery for further treatment [Figure 5]. Primary cheiloplasty and palatoplasty was done. Healing was uneventful [Figure 6]. Patient was planned for further follow up treatments.

The treatment was carried over a period of 3 months. During this period, incrementally force was applied and measurements were recorded [Table 1]. Five mm of posterior movement of the pre-maxilla was achieved on the right side and 6 mm on left side, respectively, in antero-posterior plane.

Discussion

Pre-surgical orthopedics concept was introduced at University of Glasgow by Kerr McNeil in 1954 as an adjunctive neonatal therapy aiming at non-surgical reduction of the size of the alveolar cleft. The rationale behind “Maxillary Orthopedics” as described by McNeil, Burston, and Rosenstein was that “early segment alignment will allow the maxillary halves to develop normally even though a normal bony union is not present”.^[4] In the present case, the patient had reported with a great delay for correction of defective lip and palate; however, on presentation the scope of surgical correction alone was minimal due to the deviated pre-maxilla on right side and possible high tension in the suture line if correction was attempted at this stage. The opinion of delaying surgical correction was supported by the findings of Sierra FJ^[9] where infants with wide unilateral cleft lip and palate or BCLP with a protrusive pre-maxilla were problematic for the surgeon due to the distance the tissue must be mobilized to close the defect. Hence, the pre-surgical infant orthopedics was advised in these cases which achieve alignment of the maxillary segments, presenting a more symmetrical platform and width reduction of the alveolar ridge cleft.^[3]

Bitter^[10] described three different approaches for correcting the protruding premaxilla; surgical closure of soft tissues, a passively working appliance, and an actively working appliance. Using active pre-operative orthopedics to reposition the protruding pre-maxilla of bilateral cleft palate is a reasonable and effective approach. Various appliances available are Bonnet and strap appliance, Micro foam tapes, Passive plates, custom made appliances with nasal stents, Ring covering premaxilla,^[11] Latham’s DMA appliance, Jackscrew appliance, and Nordin appliance.

The ideal time for the presurgical orthopedic treatment is from birth to 5 months of age,^[2] which will have the advantage of faster correction in less time. This case was referred to us at the age of 5 years because the parents were not aware of the available treatment options. In this case, we have used a special custom-made orthopedic appliance self-designed,

which was suitable in any clinical setting with a minimal material and instruments requirement.

This appliance was comfortable to wear, simple to use, and cost-effective. The working end of appliance was simple acrylic bulb to move the deviated pre-maxilla in-to a favorable position by application of intermittent and slow force, which in turn was controlled by the activation schedule of twice-a-week. This appliance was made and based on the principle of orthopedic appliance of force application and

force elimination, leading to favorable arch or jaw position and thus maintaining functionality.^[11]

The need for extra-oral forces was not required due to the maximum possible retraction of the pre-maxillary segment. In



Figure 1: Custom made presurgical orthopedic appliance



Figure 3: Taking measurements



Figure 2: Appliance delivered to the patient



Figure 4: Approximation of pre-maxillary with the palatal half in left side



Figure 5: After retraction of pre-maxillary



Figure 6: After primary cheiloplasty

Table 1: Distances between the hard and soft tissue reference points on both right and left sides measured in anteroposterior plane

Date	Right				Left			
	Soft tissue		Hard tissue		Soft tissue		Hard tissue	
	Soft tissue distal to 2 nd molar-midline (st)	2 nd molar (d)-lateral incisor(d)	2 nd molar (d)-midline	Canine (m)-lateral incisor(d)	Soft tissue distal to 2 nd molar -midline (st)	2 nd molar (d)-central incisor(d)	2 nd molar (d)-midline	Canine (m)-central incisor (d)
07/09/11	56	32	42	13	56	39	46	19
08/10/11	54	30	39	10	54	35	42	15
10/11/11	52	29	42	8	52	36	41	16
08/12/11	47	27	37	6	48	32	37	12

Values are reported in millimeters, d=Distal, st=Soft tissue, m=Mesial

this case, the pre-maxillary segment could not be approximated closely to the palatal segments due to the tissue contact of pre-maxillary stalk to the palatal segments medially on left side, and this can be attributed due to the uncontrolled, and deviated growth of pre-maxilla, which was not in accordance with palatal segments of both sides in a bilateral cleft lip and palate.

However, the primary cheiloplasty became more precise with this appliance by making the lip closure with less tension and maintaining the symmetry of nares. Hence, it would be possible to suggest that the pre-surgical orthopedics would be of great help in an infant; in these kind of delayed cases, the possible retraction and approximation of palatal shelves should be attempted first with pre-surgical orthopedics than a difficult surgical closure or intentional fracture of the premaxilla and closure of the lip which leads to a certain morbidity in the midface growth of the child.^[12]

Why this paper is important to pediatric dentists

This article provides information about a new custom-made appliance, which was successfully used in retraction of pre-maxilla in BCLP case. It also adds to the knowledge of pediatric dentists that a considerable retraction of the pre-maxilla is possible even after the ideal time for the repair in the treatment of BCLP with protruding pre-maxilla.

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