Original Article

Nasoalveolar molding of bilateral cleft of the lip and palate infants with orthopaedic ring plate

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Abstract

Objective: To introduce a simple, self-retentive and cost effective presurgical infant orthopaedic plate with anterior ring to retract and align the grossly protruded and deviated pre-maxilla and to perform the nasoalveolar moulding in order to facilitate initial lip repair.

Method: The study was conducted in the orthodontic department of dental section of CH & ICH Lahore. The nonprobable purposive sample consisted of 35 subjects (27 males, 8 females) with a median age of 7 days (1-13 days). Nonsyndromic patients with bilateral complete cleft lip and palate were included in the study, patients were treated with custom made orthopaedic plate with an acrylic ring around the protruded premaxilla for around 3 months. Anthropometric readings and cast impressions were used to calculate the pre and post treatment differences, the data was tabulated and analysed using SPSS v 12.

Results: The results showed highly significant (<.005) difference between the pre and post treatment records of variables except the intercanine distance where the results are significant i.e. (< 0.05).

Conclusion: The orthopaedic plate causes significant retraction of the premaxillary segment without applying extra oral forces. It produces columellar elongation and increase in prolabium length facilitating the primary cheiloplasty and rhinoplasty to be precise and without additional scarring (JPMA 60:527; 2010).

Introduction

Orofacial clefts comprise a range of congenital facial deformities. The prevalence of orofacial cleft is reported as one in every 500 to 1000 births.[1] Golalipour et al,[2] referenced oral cleft prevalence as 1.34 per 1000 in whites, 0.41 per 1000 in African Americans, 0.34 per 1000 in Africans and 2.13 per 1000 in Japanese. Calzolari et al[3] reported that the cleft lip and palate is a common congenital anomaly with significant variation depending upon geographical location, racial and ethnic background and socio economic status. These 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 BCLP is unknown. Watanabe A 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 Raphel[5] described bilateral cleft lip and palate 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 bilateral cleft lip/palate 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 premaxilla 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.

The protruded segment renders the surgical repair and alignment of the pre-maxillary segment very difficult and is a challenge for reconstructive team. The determinant of difficulty for cheiloplasty in infants with bilateral cleft lip and palate (BCLP) is the extent to which premaxilla is protruded[7] and is the most troublesome malalignments of the alveolar arch in such patients. Cheiloplasty with markedly protruded and deviated segment may not end up in good results.

The rationale behind "Maxillary Orthopaedics" as described by McNeil, Burstone and Rosenstein is that early segment alignment will allow the maxillary halves to develop normally even though a normal union is not present.[8]

Since then a number of active and passive appliances have been reported in the literature. The appliances used to align the protruding premaxilla include intraoral appliances[9-11] and combination of intraoral and extraoral appliances.[7,12-15] Santiago et al[14] demonstrated that the use of pre-surgical infant orthopaedic plate prior to initial lip repair helps in achieving alignment of premaxillary segment and elongation of the columella attributing to increased nasolabial mucosal lining surface area. Grayson and Cutting[16] described that the pre-surgical nasoalveolar molding and pre-maxillary alignment are known to facilitate initial surgical repair and reduce post surgical scarring. During the initial 2 to 3 months of life the plasticity of cartilage and mouldability of the alveolar bone segments especially the premaxilla is high that helps in achieving premaxillary retraction, derotation and approximation of the cleft alveolar segments. This high plasticity is due to the high levels of hyaluronic acid, a component of the proteoglycan intercellular matrix[17] and maternal oestrogen circulating in the neonates.[16]

In recent years devices of various designs have been used as nasoalveolar molding appliances for active alignment of the cleft alveolar segments prior to initial lip repair. Such appliances are usually consisting of an intra-oral palatal plate that obturates the oronasal defect and extra-oral attachments which mainly consist of combinations of straps, tapes and springs.[18] With the introduction of soft acrylic the Pre-Surgical Infant Orthopaedic Plate (PSIOP) was designed which is self retentive and does not require any extra-oral means of retention. A custom made self retentive orthopaedic plate with anterior ring was designed for nasoalveolar molding of the neonates born with bilateral cleft of the lip and palate.[19] Idea behind this innovative device with anterior acrylic ring that surrounds pre-maxilla was to retract and align the protruded and rotated pre-maxillary segment without combination of extra oral attachment.

Patients and Methods

The present study is a Quasi experimental, intervention type and was conducted in the orthodontic department of dental section of CH & ICH Lahore with the permission of hospital ethical board. The non probable purposive sample consisted of 35 subjects. Out of this sample size, 27 subjects were males and 8 were females. Median age of the patient used in this study was 7 days (1 Day - 13 Days).

Patients with complete bilateral cleft lip and palate were included in the study, while patients with incomplete bilateral cleft lip and palate and syndromic patients were excluded.

Consent was taken from the parents and a detailed history proforma was prepared for each patient, comprising relevant information and a demographic data was obtained. The chair side clinical pre treatment records were obtained in the form of extra oral/intra oral photographs (Figure-1a,b and c), anthropometric measurements of the columella and prolabium with finally an initial impression with rubber based impression material (Elite H-D+, Putty Soft Normal Setting, Vinylpolysiloxane Impression Material, Z hermack) was taken and cast prepared in stone plaster.

Patients were treated for a period of almost 3 months, with custom made orthopaedic plate incorporating a self-cure acrylic ring around the protruding premaxilla (Figure-1d). The plate was made self retentive by the addition of soft acrylic (GC Corporation Tokyo, Japan) on its palatal surface, filling the cleft area (Figure-1d,e). This innovative self retentive plate was made up of acrylic which comprised of two parts; the palatal plate covered the palatal defect and served as a passive obturator and the anterior part of the plate extended as a ring around the protruded and deviated premaxilla (Figure-1d,e).

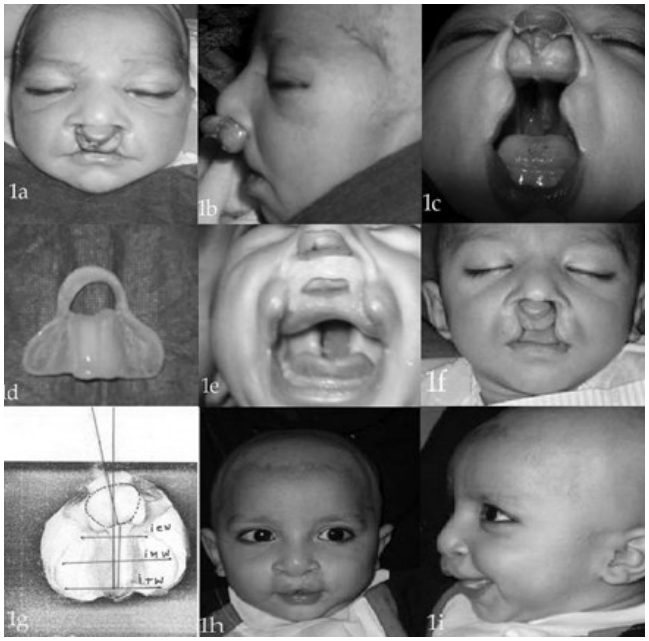

Figure: (1a) Pre treatment front view. (1b) Pre treatment profile. (1c) Pre treatment Intra oral view. (1d) PSIOP with anterior ring. (1e) Appliance in situ. (1f) Post molding front view. (1g) Cast photocopy analysis. (1h) Post surgical front view. (1i) Post surgical profile.

This ring acted as an active part to align and retract the malposed premaxillary segment. Taking care of the air passage, palatal cleft gap was filled with soft acrylic to attain retention. Later on weekly activation of the ring by 1mm addition of hard self cure acrylic along the inner surface and removal from the outer surface was undertaken to maintain uniform thickness of the ring. For correction of pre-maxillary rotation, more than 1 mm acrylic was further added against the area to be depressed. The protruding premaxillary segment was aligned and retracted to desired position (Figure-1f) where surgical repair of the lip would be more feasible, leaving fewer scars (Figure-1h,i). Anterior edge of the palatal plate within the margins of acrylic ring was trimmed slightly more than the addition of acrylic ring on each activation. This was done to facilitate retraction of the premaxillary segment. The palatal plate was adjusted every two weeks by adding self cure ortho acrylic on the palatal surface, followed by insertion and adaptation of the plate. On curing, the same amount of acrylic was removed along its outer and oral surface.

Photocopies were taken for both pre and post molding casts for recording linear and angular measurements (Figure-1g). In this study, a total of 8 variables were used comprising 6 linear measurements and 2 angular measurements (Table).

Results were based on the measurements from pre and post moulding records. Results were tabulated and analyzed by using SPSS 12.

The linear and angular measurements from the cast photocopies are: Retraction of premaxilla, Rotation of premaxilla, Septum deviation, Intercanine width, Inter molar width.

The linear measurement from the cast: Downward movement of premaxilla (with the help of soft vinyl scale)

Anthropometric measurements: (Directly from the skin with the help of soft vinyl scale) included Columellar length and Prolabium length.

Results

The average age of mothers of the studied patients was 26 years (19-33). Percentage of the mothers who had taken some form of medication (pain killer, antibiotics and herbal medicine) during their first trimester of pregnancy was 29%

Table: Results of variables for intervention.

Paired Samples Test

		Paired Differences					t	df	Sig. (2-tailed)
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference Lower	Upper			
Pair 1	Retraction of premaxilla pre-treatment - Retraction of premaxilla post-treatment	3.8000	1.30158	.22001	3.3529	4.2471	17.272	34	.000
Pair 2	Rotation of premaxilla pre-treatment - Rotation of premaxilla post-treatment	5.4143	7.40080	1.25096	2.8720	7.9565	4.328	34	.000
Pair 3	Deviation of septum pre-treatment - Deviation of septum post-treatment	1.6714	1.48494	.25100	1.1613	2.1815	6.659	34	.000
Pair 4	Downward movement pre-treatment - Downward movement post-treatment	-2.9286	2.33938	.39543	-3.7322	-2.1250	-7.406	34	.000
Pair 5	Intercanine width pre-treatment - Intercanine width post-treatment	-.7857	2.04467	.34561	-1.4881	-.0833	-2.273	34	.029
Pair 6	Intermolar width pre-treatment - Intermolar width post-treatment	-1.0286	1.44478	.24421	-1.5249	-.5323	-4.212	34	.000
Pair 7	Columellar length pre-treatment - Columellar length post-treatment	-2.2429	.74134	.12531	-2.4975	-1.9882	-17.899	34	.000
Pair 8	Prolabium length pre-treatment - Prolabium length post-treatment	-1.8571	.67051	.11334	-2.0875	-1.6268	-16.386	34	.000

P < 0.005 considered significant.

while those subjected to accidental radiation exposure was 1%. However, most of the mothers belonged to the socio economically compromised class. The percentage of consanguineous marriages was remarkably high being 75%. Three of the affected neonates presented with positive family history of cleft deformity, where one father, one sister and two first cousins were reported to have oral clefts.

The results of variables for intervention were retraction of pre-maxillary segment to be 3.80 ± 1.3 mm. Derotation of the pre-maxilla was recorded as 5.41 ± 7.4 degrees, whereas septum deviation was 1.67 ± 1.48 degree. The increase in columellar as well as prolabium length recorded was 2.24 mm and 1.85 mm respectively. Intercanine width was increased by 0.78mm after molding. The inter-molar width however showed an increase of 1.02mm .The downward movement of premaxilla measured from the cast was 2.92 ± 2.3 mm (Table).

Discussion

The objective of presurgical orthopaedics is to normalize anatomy as much as possible. Grayson[16,18] introduced devices for nasoalveolar molding of the neonates born with unilateral and bilateral cleft of the lip and palate. Santiago et al[14] described nasoalveolar molding as a non surgical method of reshaping the premaxilla, lip and nostrils before cleft lip and palate surgery, lessening the severity of the cleft. Pre surgical nasal and alveolar molding includes active molding and repositioning of the nasal cartilages and alveolar processes as well as lengthening of the deficient columella.[18] The process usually takes approximately three months because of maternal estrogen levels. A significant decrease in distance between the premaxilla and the lateral segments occur. Surgery is performed after the molding is complete, approximately three to six months after birth. Using active preoperative orthopaedics to reposition the protruding premaxilla of bilateral cleft palate is a reasonable and effective approach.[14,16] Bitter[20] described three different approaches for correcting the protruding premaxilla; surgical closure of soft tissues, a passively working appliance, and an actively working appliance. Complications of the traditional surgical approach were reported as growth arrest of the nasomaxillary complex.[9] Grayson and Santiago[21] demonstrated that the conventional surgical technique had other limitations, including lingual inclination of upper incisors due to lingually displaced pre maxilla, nasal airway obstruction, and flat face etc. Some of the problems that the traditional surgical approach failed to solve are described as deformity of the nasal cartilages in unilateral as well as bilateral clefts of the lip and palate and the deficiency of columellar tissue in infants with bilateral clefts. Johansson and Ohlsson[22] described the use of lip adhesion before primary bone grafting as an intervention. Demerits of lip adhesion included the risks of an additional surgical procedure, scarring of the involved tissue, and dehiscence of the surgical site. Bitter[20] also described that surgical closure alone and passively working appliance did not align the premaxilla in perfect position. The use of NAM technique has eliminated surgical columella reconstruction and the resultant scar tissue.

Among the active devices various extra-oral and combination appliances including facial strapping[7,12-15] and intra-oral pinned appliances[9,11] have been used. These appliances have been using straps or adhesive tapes and pin fixation systems as means of retention and to generate an active force to retract the premaxilla. Such means of retention and pins are reported to be cumbersome and traumatic to the sensitive facial skin of the newborn and premaxillary segment. Potential damage to the tooth buds has also been described in the literature.

Our custom made ring plate, contrary to the above mentioned appliances, does not use traumatic or cumbersome means. The soft acrylic is used to fill the cleft gap for retention and it exerted the pressure to act as nasal stent. The anterior ring made up of acrylic is merely an anterior extension of the intra oral palatal plate around the pre-maxilla that acts as an active component for retraction. No strapping or extra oral tapes are used to retract the pre-maxillary segment. Instead sequential reshaping of the ring by addition of acrylic along the inner aspect and removal from the outer surface is undertaken for active retraction. In this device, retraction force is generated by the functional activity of the orofacial musculature. The tongue being placed at the back exerts distalizing force during function, like suckling and swallowing. The retraction or molding of the premaxillary cleft segment becomes easier due to the mouldability of the tissues, which in turn is because of the raised levels of hyaluronic acid[17] and maternal estrogen circulating in the neonates.[18] The activation of acrylic ring was done on weekly basis during the initial three months after birth. In order to correct rotation of the deviated pre-maxilla, more acrylic was added against the part to be depressed. The palatal adjustment was done to follow growth of the alveolar segments as well as to ensure retention of the device.

In our experience with this simple and cost effective intra-oral appliance we achieved significant retraction and alignment of the deviated pre-maxilla after orthopaedic treatment. The columella and prolabium showed an adequate increase in length because of the partial stenting effect from the soft acrylic filling the undercut area of cleft. Our findings show a positive effect of molding therapy on the soft and alveolar tissues and are supported by the research studies of various other investigators.[11,14,16,18,20,21] However, a previously conducted study at the same centre involving ten neonates with BCLP who presented immediately after birth and given the same ring plate device, showed greater premaxillary retraction. This finding could be ascribed to highest maternal oestrogen levels, immediately after birth.[17]

Improvement of the deviated nasal septum was also observed besides marked alignment of the premaxillary segment in this study. The combination of presurgical nasoalveolar molding and non-surgical columella elongation allows bilateral cleft lip and nose correction in a single stage. This has been shown to result in an improved aesthetic outcome and reduced need for surgical revision before the age of secondary bone grafting. Most of the infants, who were provided surgical repair by the plastic surgeons after receiving moulding therapy by our ring plate, underwent the same experience. Grayson and Cutting[16] described that proposed benefits of traditional intraoral presurgical orthopaedics are improved feeding, prevention of palatalized articulation, normalization of tongue position, growth guidance, development of palatal segments, minimization of treatment at a later age, better speech of the patient and positive psychological effect on the parents, savings in cost to the patient and society through the reduction in number of surgical hospital admissions. Finding of the current procedure, lasting over the period of seven years, are supported by these statements.

The effect of pre surgical orthopedics on maxillary growth has been a matter of debate. A 10 year follow-up study,[23] concluded better facial growth in patients treated with pre surgical orthopaedics when compared to control subjects. Our study too, has drawn the same conclusion. Another study[24] has demonstrated that pre surgical infant orthopaedics enables a more precise repair with less tension and if surgery is thereby improved, this is a powerful incentive to adopt pre surgical infant orthopaedic procedures. The co-ordination between the parents, orthodontist, and plastic surgeon leads to the desired outcomes of pre-surgical nasoalveolar molding.

Conclusion

The study proved that the orthopaedic self retentive ring plate is comfortable to wear and simple to use and cost effective. Maxillary growth is facilitated with this device and thus functional activity. It causes significant retraction of the premaxillary segment without applying extra oral forces, produces columellar elongation and increase in prolabium length without additional scarring.

The primary cheiloplasty and rhinoplasty becomes more precise with this appliance by making the lip tension free and maintaining the symmetry of nares.

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References

1. Durning P, Chestnutt IG., Morgan MZ, Lester NJ. The relationship between orofacial clefts and material deprivation in wales. Cleft Palate Craniofac J 2007; 44: 203-7.
2. Golalipour MJ, Mirfazeli A, Behnampour N. Birth prevalence of oral clefting in northern Iran. Cleft Palate Craniofac J 2007; 44: 378-80.
3. Calizolari E, Bianchi F, Rubini M, Ritvanen A, Neville AJ, EUROCAT Working Group. Epidemiology of cleft in Europe: implications for genetic research. Cleft Palate Craniofac J 2004; 41: 244-9.
4. Watanabe A, Akita S, Tin NT, Natsume N, Nakano Y, Niikawa N, et al. A mutation in RYK is a genetic factor for non-syndromic cleft lip and palate. Cleft Palate-Craniofacial Journal 2006; 43: 310-6.
5. Morand B, Raphael B. Bilateral cleft lip and palate. Anatomic and clinical characteristics and therapeutic results. Orthod Fr 2004; 75: 243-51.
6. Mulliken JB, Wu JK, Padwa BL. Repair of bilateral cleft lip: review, revisions, and reflections. J Craniofac Surg 2003; 14: 609-20.
7. Mishima K, Sugahara T, Mori Y, Minami K, Sakuda M. Effects of Pre surgical orthopaedic treatment in infants with complete bilateral cleft lip and palate. Cleft Palate Craniofac J 1998; 35: 227-32.
8. Singla S, Kaur M. Cleft palate habilitation. Indian J Pediatrics 2008; 75: 703-8.
9. Georgiade NG, Latham RA. Maxillary arch alignment in the bilateral cleft lip and palate infant using the pinned coaxial screw appliance. Plast Reconstr Surg 1975; 56: 52-60.
10. Papay FA, Morales L Jr, Motoki DS, Yamashiro DK. Presurgical orthopaedic premaxillary alignment in cleft lip and palate reconstruction. Cleft Palate Craniofac J 1994; 31: 494-8.
11. Delgado MD, Marti E, Romance A, Romero M, Lagaron E, Salvan R, et al. Presurgical orthopaedic treatment for cleft lip and palate. Cir Pediatr 2004; 17: 17-20.
12. Hotz MM, Gnoinski W. Comprehensive care of cleft lip and palate children at Zurich University: a preliminary report. Am J Orthod 1976; 70: 481-504.
13. Georgiade NG, Mason R, Riefkohl R, Georgiade G, Barwick W. Preoperative positioning of the protruding premaxilla in the bilateral cleft lip patient. Plast Reconstr Surg 1989; 83: 32-40.
14. Santiago PE, Grayson BH, Cutting CB, Gianoutsos MP, Brecht LE, Kwon SM. Reduced need for alveolar bone grafting by pre-surgical orthopedics and primary gingivoperiosteoplasty. Cleft Palate Craniofac J 1998; 35: 77-80.
15. Wang T. Wang DZ, Xu HF. Effect of orthopaedic premaxillary retrusion on craniofacial development in animal model. Zhonghua Kou Qiang Yi Xue Za Zhi 2004; 39: 313-5.
16. Grayson BH, Cutting CB. Presurgical nasoalveolar orthopaedic molding in primary correction of nose, lip and alveolus of infants born with unilateral and bilateral cleft. Cleft Palate-Craniofacial J 2001; 38: 193-8.
17. Matsuo K, Hirose T, Tonomo T, Iwasawa M, Katohda S, Takahashi N, et al. Nonsurgical correction of congenital auricular deformities in the early neonates, a preliminary report. Plast Reconst Surg 1984; 73: 38-50.
18. Grayson BH, Maull D. Nasoalveolar molding in infants born with cleft of the lip, alveolus and palate. Clin Plast Surg 2004; 31: 149-58.
19. Ijaz A. Management of complete bilateral Cleft of the lip and palate with modified pre Surgical infant Orthopaedic plate. Pak Oral Dental J 2003; 23: 131-6.
20. Bitter K. Latham's appliance for presurgical repositioning of the protruded premaxilla in bilateral cleft lip and palate. J Craniomaxillofac Surg 1992; 20: 99-110.
21. Grayson BH, Santiago PE. Pre Surgical Orthopaedics for Cleft Lip and Palate. Grabb and smith's Plastic Surgery, 5th ed. Philadelphia: Lippincott Raven Publishers; 1997; 20: 237-44.
22. Johanson B, Ohlsson A. Bone grafting and dental orthopedics in primary and secondary cases of cleft lip and palate. Acta Chir Scand 1961; 122: 112-24.
23. Robertson NR. Facial form of patients with cleft lip and palate. The long term influence of presurgical oral orthopaedics. Br Dent J 1983; 155: 59-61.
24. Ross RB, MacNamera MC. Effect of pre surgical infant orthopedics on facial aesthetics in complete bilateral cleft lip and palate. Cleft Palate Craniofac J 1994; 31: 68-73.