MANAGEMENT OF COMPLETE BILATERAL CLEFT OF THE LIP AND PALATE WITH MODIFIED PRE SURGICAL INFANT ORTHOPAEDIC PLATE

*ABIDA IJAZ, BDS, D. ORTH, MCPS. ORTH, MS. ORTH (Turkey)

ABSTRACT

Cleft of the lip and palate are the most common congenital facial deformities. Clefts may vary in severity from minor notching of the lip or bifid uvula to complete unilateral or bilateral cleft of the lip and palate. Treatment of complete bilateral cleft of the lip and palate had been a challenge in the past, because of protruding pre maxilla and deficient columella. The treatment of choice was excision of pre maxilla, which was later replaced, by surgical retraction of pre maxillary segment alone or the one followed by pre surgical infant orthopedics. The concept of Pre-Surgical Infant Orthopaedics (PSIO) was originated after disappointment from the results of existing techniques. An oral prosthesis, with extra oral attachments around babies head was used to retract pre maxilla. In the following years, this device was modified by making it self-retentive by filling the cleft part with acrylic similar to an obturator. In the recent past, however, introduction of soft acrylic has provided retentive means in the defect part. The device provides alveolar molding maintaining arch form and facilitates function. This article reports on pre surgical infant orthopedic treatment of a new born baby using modified orthopedic plate with anterior acrylic ring. The appliance comprised of an acrylic ring extending anteriorly from the existing orthopaedic plate. The device on insertion surrounded the protruded pre maxilla and exerted retraction force utilizing muscle activity. Acrylic ring was adjusted every fortnight by adding 1mm acrylic along the inner surface of the acrylic ring contacting labial surface of pre maxillary segment and trimming the same amount along the anterior margin of the plate. Duration of appliance wear was three months. The results showed a significant pre maxillary retraction of 3.5mm and correction of rotation by 12 degrees. The appliance proved most successful being used immediately after birth, making use of plasticity of the neonate cartilage due to maternal estrogen levels. Moreover, the appliance maintained arch form, facilitated function and rendered initial lip repair easy and tension free.

INTRODUCTION

Cleft of the lip and palate are the most common congenital facial deformities. Incidence varies according to the type of cleft, racial group and sex. Cleft lip with or without cleft palate ranges from 3.6/1000 births for Indians, to 0.5/1000 for Negroes and 1/1000 birth for Caucasians1.

Etiology of clefts of the lip and palate is still a matter of debate. Heredity is reported to play an important role. Amongst the environmental factors are drugs like corticosteroids, thalidomide, radiation, stress and infections like rubella virus etc.

The embryonic median nasal processes give rise to the tip of the nose, columella, prolabium and primary palate containing four incisors. Between 7th and 8th week of embryonic life, mesodermal invasion results in a merging of the lateral and median nasal process with the maxillary processes to form the upper lip. Failure of adequate mesodermal migration or proliferation can result in complete or incomplete cleft of the lip. The secondary palate develops from the inner surfaces of the maxillary processes during 6th week of uterine life. By the 8th week the tongue descends in the enlarging stomatodium and the palatal shelves swing up medially to achieve contact with one another, with the primary palate anteriorly and the nasal septum above. The epithelium at the sites of the contact breaks down and these processes fuse to form secondary palate between 8th to 12th weeks. Lack of elevation of palatal shelves at the critical time or failure to contact or inadequate epithelial break down, gives rise to a cleft of secondary palate.

Clefts may vary in severity from minor notching of the lip or a bifid uvula, to complete unilateral or bilateral cleft of lip and palate. A number of classifica-

* Head Department of Orthodontics, Children Hospital, Lahore
tions have been proposed. However, Veau’s classification was one of the earlier and most widely known.

In bilateral complete cleft, the pre maxillary segment is displaced anteriorly and suspends from the tip of the nasal septum. The prolabium lacks muscle tissue and is attached on the end of the short columella, while the lateral buccal segments are often well related to the lower arch.

Treatment of bilateral cleft of the lip and the palate deformities, in the past had been a challenge to the surgeons. The main obstacle to repair was the protruding pre maxilla and the deficient columella. During sixteenth, seventeen and eighteenth centuries, the surgical treatment involved excision of the pre maxilla followed by surgical union of the prolabium to the lateral lip segments. At a later age, prosthetic replacement of the anterior dentition was recommended to improve facial appearance.

In nineteenth century, surgeons noted that by excising pre maxilla, the lip became deprived of bony support, causing mid face deficiency, maxillary constriction, malocclusion, and an apparent mandibular prognathism. The focus was then based on preservation and retraction of the pre maxilla to achieve optimal lip repair. Two treatment modalities evolved: surgical correction alone and surgical correction following pre surgical infant orthopedics.

Surgical option for pre maxillary retraction included resection of part of the vomer or nasal septum, partial resection of the anterior portion of the pre maxilla, and full thickness vertical incision of the septum, that allowed the anterior and posterior segments to slide over each other. Although these techniques achieved the primary goal of retracting the pre maxilla they were associated with significant complications. Both long term clinical observation and animal studies have demonstrated that the resection of nasal septum produced growth arrest of adjacent bones. The technique had other limitations, including lingual inclination of upper incisors due to lingually displaced pre maxilla, nasal airway obstruction, and flat face etc.

Another surgical approach that attempted to achieve the pre maxillary retraction and lateral segment approximation was lip adhesion. In 1961 Johanson and Ohlsson described the use of lip adhesion before primary bone grafting. Disadvantages in the use of lip adhesion include the risks of an additional surgical procedure, scarring of the involved tissue, and dehiscence of the surgical site. In addition, the tension of the surgically adhered lip over the alveolar segments is an uncontrolled force that does not always align the segments in an ideal position, frequently causing collapse of the dental arches.

The concept of pre surgical infant orthopaedic treatment originated in 1500s, when excision of the protruded pre maxilla in bilateral cleft was the recommended treatment. Dissatisfied with the long-term results of this treatment modality, surgeons and dentists found new avenues to achieve more optimal results. In 1561, Franco described a head cap for extra oral therapy. Louis in 1768, Chaussier in 1776, and Desault in 1790 used bandages over the prolabium to stimulate muscle retraction, compressing the pre maxillary regions. In 1844, Hullihen, an American dentist, used facial adhesive strapping to prepare the cleft before surgery. He believed that closing of alveolar cleft prior to surgery during the first month of life was crucial in order to properly perform lip closure. Thiesch in 1875, using rubber bands; and Von Esmarch and Kowalzig, in 1892, employing an elastic band attached to a head cap, contributed to the development of this field.

The concept of modern pre surgical infant orthopedics (PSIO) started with the work of McNeil, who disappointed with the maxillary collapse created by the available techniques, utilized an oral prosthesis similar to an obturator to approximate the cleft alveolar segments. In his technique, a maxillary impression was taken of the newborn and an acrylic appliance was made from a plaster model that was cut and modified with the cleft gap slightly closed. By repeating this step and frequently modifying the appliance, McNeil was able to close not only the alveolar gap, but also the hard palate cleft by influencing bone growth direction.

Gnoinski devised another commonly used appliance that consisted of an alveolar molding plate made of a hard outer shell and a soft acrylic lining. By gradual alteration of the tissue surface of the acrylic plate, the alveolar segments were gently pressed to grow and mold into the desired shape and position.

A technique developed by Rosenstein (1963), Rosenstein and Jacobson (1967), and Monoroe (1968) consisted of an acrylic obturator extended into the alveolar undercut areas. The device allowed continued growth by a passive molding action without permitting medial movement of the buccal segments. Once the segments were in proper position, early lip repair and bone grafting could be performed.

One of the most controversial issues around the subject has been the effect of pre surgical orthopedics on maxillary growth. Ross (1987) showed in a major multicentre study that there is no difference in facial growth between cleft patients treated with or without pre surgical orthopedics. On the other hand, Robertson (1983) in a 10 year follow-up study by a single surgeon, demonstrated that better facial growth was achieved in patients treated with pre surgical orthopedics when
compared to control subjects. Ross and Mac Namera\textsuperscript{12} stated that one possible benefit of pre surgical infant orthopedics is that when the maxillary segments are repositioned so that the lip segments are in closer apposition, the lip surgery becomes easier, enabling a more precise repair with less tension and if surgery is thereby improved, this a powerful incentive to adopt pre surgical infant orthopedic procedures.

Pre surgical nasal and alveolar molding includes as its objectives the active molding and repositioning of the nasal cartilages and alveolar processes as well as lengthening of the deficient columella. A description of the protocol for treatment of the patients with bilateral cleft deformity was introduced in 1993 by Grayson, Cutting and Wood\textsuperscript{13}.

Matsuo, Hirose, and Tonomo\textsuperscript{14} postulated that the high degree of plasticity and lack of elasticity in neonatal cartilage is due to high levels of hyaluronic acid, a component of the proteoglycan intercellular matrix. As the estrogen level increases, the elasticity of the cartilage decreases\textsuperscript{15}. With the neonatal levels of maternal estrogen highest immediately after birth, the period of plasticity is slowly lost during the first month of postnatal life. It is during this first 2 to 3 months after birth when active soft tissue and cartilage molding plate therapy is most successful. A combined technique for nasal and alveolar molding is currently employed in managing infants with unilateral and bilateral cleft lip, alveolus, and palate by the cleft palate team of the institute of reconstructive plastic surgery at New York university medical center\textsuperscript{2}. This technique has been demonstrated to have a positive influence on the outcome of the primary nasal, labial, and alveolar repair.

The objective of pre surgical nasal and alveolar molding in the patients with bilateral deformity is to lengthen the columella, reposition the nasal cartilages towards the tip, and align the alveolar segments. Grayson and Santiago\textsuperscript{2} (1997) described the technique in which the everted pre maxilla was repositioned in alignment with lateral alveolar segments, using intra oral molding plate in conjunction with elastic bands that adhered to the cheeks or were attached to the head cap. Then the nasal stents were built from the anterior rim of the oral plate that entered the nasal apertures. This was reported to provide support and give shape to the dome and alar cartilages in the immediate newborn period. The nasal stents advanced the lateral alar cartilages into the nasal tip and provide stretch to the colmellar skin.

In the bilateral deformity, the columella at birth is often from 0 to 2.0 mm in length\textsuperscript{2}. In the technique described by Grayson and Santiago, it was pre surgically extended to 4.0 to 7.0 mm. The normal columellar, length for children at this age being 3.2 mm. Overcorrection of the columellar, length was intended to account for some postsurgical relapse. This procedure obviated the need for pre surgical lengthening of the columella, avoiding scar at the base of the nose. The goal of this less invasive surgical approach was to minimize the extent of scar tissue formation, enhancing nasal and facial aesthetics. The nasolabial complex is expected to grow more normally when free of the scar tissue that results from the conventional surgical lengthening of the columella.

The need for designing this appliance with anterior acrylic ring, surrounding the pre maxillary segment was to retract protruded pre maxilla, in the neonatal stage, with out the use of extra oral attachments.

This modified pre surgical infant orthopaedic plate was used in a 4 days old male baby. The child was otherwise healthy and was the first issue of his parents, who had no history of intermarriage. The clinical data of the patient was as given below;

**EXTRA ORAL EXAMINATION**

On extra oral examination, the pre maxillary segment was displaced anteriorly and suspended from the tip of the nasal septum Fig 1, 2. The nasal alae were stretching over the cleft. The prolabium lacked muscle tissues and was attached on the end of the short columella.

**INTRAORAL EXAMINATION**

The cleft extended backward involving the whole of hard and soft palate. The baby was classified as complete bilateral cleft of the lip and palate.

**TREATMENT PLAN**

Objectives of the treatment was to

- Retract and align rotated pre maxilla
- Facilitate function
- Maintain arch form
- Facilitate maxillary growth
- Facilitate initial lip repair

**APPLIANCE DESIGN**

Orthopaedic plate with anterior acrylic ring was fabricated for this baby. Special tray was prepared from self cure acrylic to record impression in rubber base material. Cast was poured in stone plaster. The defect part was boxed in wax and orthopaedic plate was made from self cure acrylic as usual, using pressure pot. Acrylic ring around the pre maxillary alveolar segment
was then fabricated and the plate was finished and polished for the next chair side procedure. The appliance was tried in the baby's mouth for adaptation and the defect part was then filled with soft acrylic for retention purpose Fig 3. The parents were demonstrated to use the appliance as full time wear in baby’s mouth and instructions were given for thorough cleansing of the plate after every feed, followed by antifungal drops to be administered over the top surface. As the child is fed on milk, fungal infection is common in cleft babies.

**TREATMENT PROGRESSION**

The appliance was adjusted by adding 1mm of acrylic along the ventral surface of the anterior portion of acrylic ring contacting labial surface of the pre maxillary segment and removing a little more along the anterior margin of the plate to accommodate distalizing pre maxilla. Treatment duration with this appliance was about 3 months Fig 4.

**TREATMENT COMPLETION**

The results were based on measurements from the pre and post treatment cast photocopies Fig 5, 6 & Table 1, 2. For pre maxillary retraction and derotation where as for columellar length measurements were recorded with the soft vinyl scale on the baby’s face at pretreatment, post treatment and post lip repair stage ( to see the relapse due to surgical scar) Fig 7, 8 & Table 3.

On the cast photocopies linear and angular measurements were recorded. The linear measurements were used to see the difference between pre and post treatment inter canine and inter molar width and the difference between the arch length that is pre maxillary alveolar ridge and tuberosity plane before and after treatment completion to determine retraction of pre maxillary segment. The pre maxilla showed 3.5mm retraction where as the buccal alveolar segments expanded in the canine region by 4mm and that in the molar region by 2.5mm Fig 6 & Table 1.

Angular measurements on the other hand were made to record mal alignment or rotation of the pre maxillary segment. Mid sagittal plane was used as the reference line and line from labial frenum attachment bisecting the reference line measured rotation in degrees. In this baby the pre maxilla showed 15 degree rotation prior to the use of this orthopedic plate, while post treatment reading gave 3 degree rotation interpreting correction of 12 degrees in inclination of the pre maxillary segment Fig 6 & Table 2. Columellar length measured in the first visit with vinyl soft scale was 5mm, unlike the usual length in such babies (0-2mm), reported by B.H. Grayson, whereas its length on completion of the treatment with this appliance was 8.5mm giving an increase of 3.5mm Table 3. However, relapse of 1mm after initial lip repair was observed because of scar formation Fig 7, 8.

**DISCUSSION**

Goal of pre surgical alveolar and nasal molding in bilateral cleft deformities is to retract protruded pre maxilla and align with buccal alveolar segments, to reposition alar cartilage towards nasal tip and to lengthen columella. By restoring the normal anatomy of the maxillary segments pre surgically, primary lip repair would be facilitated. One possible benefit of pre surgical infant orthopedics is that when the maxillary segments are repositioned so that the lip segments are in closer apposition, the lip surgery becomes easier, more precise and under less tension. This of course is a powerful incentive to adopt pre surgical infant orthopedics. This article describes the effect of modified pre surgical infant orthopaedic plate with anterior ring in a newborn (4 days) male baby, who presented with complete bilateral cleft of the lip and palate.

The idea behind this appliance design with acrylic ring surrounding the pre maxillary segment was to provide active molding of the pre maxilla taking advantage of the plasticity of the neonate cartilage which in turn was due to maternal estrogen levels. The more commonly used method comprises conventional orthopedic plate with extra oral attachments namely face bow attached to the head cap for retention purpose and adhesive tape or elastic bands attached to the cheeks or head cap for retracting pre maxilla. Appliance described by B.H. Grayson and P.E. Sangtiago also

<table>
<thead>
<tr>
<th>I.C.W</th>
<th>I.M.W</th>
<th>APL</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre</td>
<td>Post</td>
<td>Diff</td>
</tr>
<tr>
<td>22</td>
<td>26</td>
<td>4</td>
</tr>
<tr>
<td>Pre</td>
<td>Post</td>
<td>Diff</td>
</tr>
<tr>
<td>41</td>
<td>43.5</td>
<td>2.5</td>
</tr>
<tr>
<td>Pre</td>
<td>Post</td>
<td>Diff</td>
</tr>
<tr>
<td>41.5</td>
<td>38</td>
<td>3.5</td>
</tr>
</tbody>
</table>

**TABLE 2: DEROTATION OF THE PRE MAXILLARY SEGMENT IN DEGREES**

<table>
<thead>
<tr>
<th>Pre treatment</th>
<th>Post Treatment</th>
<th>Derotation</th>
</tr>
</thead>
<tbody>
<tr>
<td>15</td>
<td>3</td>
<td>12</td>
</tr>
</tbody>
</table>

**TABLE 3: LENGTH OF COLUMELLA IN MILLIMETER**

<table>
<thead>
<tr>
<th>Pre treatment</th>
<th>Post Treatment</th>
<th>Difference</th>
</tr>
</thead>
<tbody>
<tr>
<td>5</td>
<td>8.5</td>
<td>3.5</td>
</tr>
</tbody>
</table>
Fig 1. Front View

Fig 2. Lateral View

Fig 3. Modified Pre surgical infant orthopaedic plate with anterior ring

Fig 4. Treatment Completion (Step-1)

Fig 5. Pre Treatment Cast Photocopy

Fig 6. Post Treatment Cast Photocopy

Fig 7. Post Treatment Front view after lip repair

Fig 8. Profile view after lip repair
utilizes extra oral adhesive tapes or elastic bands to retrude pre maxilla, although cleft area has been used for retention purpose.

Strategy behind this modification with acrylic ring was to get rid of extra oral attachments for retention or retraction purpose. Skin of the new born baby is too delicate to bear all that (adhesive tape, face bow and head cap etc).

The defect area in this new plate was bridged with soft acrylic that not only provided retention to the appliance but also consolidated the appliance maintaining arch form during buccinator activity. Moreover, it partially served the purpose of nasal stents used in Grayson's appliance extending up in the nostrils. The new addition to this device was an acrylic ring, extending anteriorly from the plate around pre maxillary segment. This appliance proved to be a true functional device making use of the muscle forces. As the child suckles, muscle forces from the tongue are transformed on to the appliance in the form of retraction force, the tongue being positioned behind the protruded pre maxilla. Acrylic ring, however, was adjusted by adding 1mm of acrylic every 2 weeks along the surface contacting pre maxillary segment and trimming more than 1mm along the anterior margin of the plate to accommodate retraction pre maxilla. The appliance was entirely independent of extra oral forces.

As the pre maxilla distalized, it moved down along with columella that showed an increase in length of 3.5mm. Increase in length of the columellar tissue might have been facilitated by the soft acrylic holding the nasal structures. When the pre surgical phase of treatment is coordinated with the primary surgical repair this new device has been found to contribute to esthetic outcome of labial repair, minimizing the extent of surgery and additional scarring that occurs due to pre maxillary surgical retraction and elongation of columella.

CONCLUSION

The modified pre surgical infant orthopaedic plate comprising anterior acrylic ring was used in a new born male baby presenting with complete bilateral cleft of the lip and palate for a period of 3 months. Successful results were obtained with this device. On the basis of these results, following conclusions maybe drawn;

- The appliance being myo- functional in action was comfortable and easy to use. It produced significant retraction of the pre maxillary segment without the use of extra oral forces.
- It caused columellar elongation without additional scarring.
- It facilitated function
- The appliance being self retentive maintained arch form and inter arch relationship.
- Maxillary growth was facilitated with the use of this appliance.
- It rendered initial lip repair easy more precise and tension free.

ACKNOWLEDGMENT

I am thankful to Dr. Saeed Ashraf Cheema, Maxillofacial Plastic surgeon who gave me an incentive to design an appliance for bilateral cleft babies.

REFERENCES

(Old references are for historical interest)