THE LONG TERM STABILITY AND HISTOLOGY OF ANTERIOR MAXILLARY DISTRACTION OSTEOGENESIS IN HYPO-PLASTIC MAXILLAE IN CLEFT LIP & PALATE PATIENTS

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By

DR. R. GUNASEELAN

Department of Oral & Maxillofacial Surgery

Rajan Dental Institute

Ragas Trust Dental College & Hospital,

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Dedicated to

My Patients

My Teachers

&

My Students

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INTRODUCTION

INTRODUCTION

Cleft lip and cleft palate are congenital malformations resulting in non-fusion of lip and palate during development *in utero.* These clefts of the lip and palate are some of the more commonly occurring malformations observed in 1:800 live births. Apart from the obviously detrimental cosmetic effects, these malformations contribute to feeding difficulties right from birth, persistent recurring ear infections, speech difficulties, dental problems and psychological challenges.

The management of these patients requires a team of specialists (Pediatricians, Oral & Maxillofacial Surgeons, Plastic surgeons, Pedodontist, Orthodontist, Otolaryngologist, Speech and Language therapists, Anesthetists and Psychologists) with a treatment plan from birth to adulthood directed towards ensuring the child's ability to eat, drink, speak, hear and a normal facial appearance.

Surgical corrections are undertaken between 10 weeks to 3 months of age for cleft lip and between 6 and 18 months of age for cleft palate.

Surgery on the upper lip, alveolus and maxillae seems to lead to an interference with growth of the mid face. Skeletal maxillary growth is known to be affected due to such surgery and leads to hypo plastic maxillae. This produces a concave facial profile leading to poor facial esthetics. The hypo-plastic maxillae also frequently lead to dental crowding due to tooth size arch length discrepancy.

During early dentition period or during the mixed dentition period, the skeletal hypoplasia can be corrected by maxillary osteotomies or by maxillary distraction. Osteotomies are done after completion of facial growth. Surgery usually performed is Le-Fort 1 osteotomy with anterior advancements and sometimes also with inferior repositioning to gain facial height. The surgical advancement especially if involving significant magnitude of movement can lead to surgical relapse, instability and worsening of velo-pharyngeal dysfunction (VPD), especially in patients with a pre existing borderline VPD. This can also worsen the individual's speech.

Distraction of the maxillae and palatine bones by means of internal or external distractions at the level of the Le-fort -1 became popular due to limitations of surgery. The advantages claimed include stability and the possibilities of operating even during the growth period.

External distractors were often cumbersome and had poor patient compliance. Internal distractors were less obtrusive, but still had significant morbidity and still require surgery for their removal.

Long term results with complete maxillary distractions were however, not very promising with significant long term relapse being reported.

Segmental distraction of the maxillae seems to be the answer. This procedure entails Anterior Maxillary Osteotomy followed by gradual distraction. These distractors were either bone borne or teeth borne. Tooth borne distractors produce significant amount of maxillary distraction and increase the alveolar volume in the distraction site which can be used for orthodontic de-crowding or for dental implant placement. This also helps keep the distraction segment is stable. The morbidity for the patient is far lesser than for full jaw distraction. The present study is to observe and report:

1) Long term stability of Anterior Maxillary Distraction which is done with tooth borne device.
2) Study the histology of the distracted soft tissue.
3) Study if speech is affected by the procedure.

AIMS AND OBJECTIVES

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Since skeletal maxillary hypoplasia is common in operated cleft lip and palate patients and there is no uniformly recommended procedure for a stable surgical correction, the anterior maxillary distraction procedure was done with the following aims and objectives.

1) To study the stability of this procedure by :-
   - i) Pre and post operative radiographic records;
   - ii) Pre and Post Operative Photographs
2) To study the histology of soft tissues in the distraction zone.
3) To do a pilot assessment of speech before and after distraction surgery.

REVIEW OF LITERATURE

REVIEW OF LITERATURE

ANATOMY

The palate is formed by hard and soft palate forming the roof of the mouth and the floor of the nasal cavity. The palatine processes of the maxilla and horizontal plate of the palatine bones form the hard palate. It is supplied by the greater palatine artery, which passes through the greater palatine foramen and are innervated by the anterior palatine and nasopalatine nerves. The soft palate is a fibro-muscular structure attached posterior portion of the hard palate. It separates the nasopharynx from the oropharynx by tensing and elevating during swallowing. The soft palate consists of the tensor veli palatini, the levator veli palatini, the musculus uvulae, the palatoglossus, and palatopharyngeus muscles. CN V supplies the tensor veli palatini, while CN IX and CN X innervate the others. The levator veli palatini is the primary elevator of the palate.

EMBRYOLOGY

The primary and secondary palates are delineated according to embryological development. The primary palate or premaxilla is a triangular area of the anterior hard palate extending from anterior to the incisive foramen to a point just lateral to the lateral incisor teeth. It includes that portion of the alveolar ridge containing the four incisor teeth. The secondary palate consists of the remaining hard palate and all of the soft palate. The primary palate forms during the 4th to 7th weeks of gestation as the two maxillary swellings merge and the two medial nasal swellings fuse to form the intermaxillary segment. The intermaxillary segment is composed of a labial component (forms the philtrum), a maxilla component (forms alveolus and 4 incisors), and palatal component (forms the triangular primary palate). Normally during development of the primary palate, a cleft does not exist (unlike the secondary palate in which cleft formation occurs as a natural stage of development). The secondary palate forms during the 6^{th} to 9^{th} weeks of gestation, as the palatal shelves change from a vertical to horizontal position and fuse. The tongue must migrate away from the shelves in an antero-inferior direction for palatal fusion to occur.

CLEFT FORMATION

In general, patients with clefts have a deficiency of tissue and not merely a displacement of normal tissue. A cleft lip occurs when an epithelial bridge fails, due to lack of mesodermal delivery and proliferation from the maxillary and nasal processes. Clefts of the primary palate occur anterior to the incisive foramen. Clefts of the secondary palate are due to lack of fusion of the palatal shelves, and always occur posterior to the incisive foramen. The secondary palate closes 1 week later in females, which may explain why isolated clefts of the secondary palate are more common in females. A cleft of the lip increases in probability of a cleft palate developing. The cleft of the lip occurs earlier and inhibits tongue migration, which may then prevent horizontal alignment and fusion of the palatal shelves. In the unilateral cleft lip, the floor of the nose communicates freely with the oral cavity, the maxilla on the cleft side is hypoplastic, the columella is displaced to the normal side, and the nasal ala on the cleft side is laterally, posteriorly, and inferiorly displaced. The lower lateral cartilage of the nose is lower on the cleft side, its lateral cruz is longer, and the angle between the medial and lateral cruz in more obtuse. The muscles of the orbicularis oris do not form a complete sphincter but instead are directed superiorly to the ala nasi laterally and the base of the columella medially. In the bilateral cleft lip, the central portion of the alveolar arch is rotated anteriorly and superiorly. The medial or prolabial segment of skin contains no muscle or vermillion. In palatal clefts, the muscles of the soft palate are hypoplastic and insert in the posterior margin of the remaining hard palate rather than the midline raphe. Associated dentition abnormalities include supernumerary teeth (20%), dystrophic teeth (30%), congenitally missing teeth (50%), and malocclusion (almost 100%)[25].

GENETICS

Non-syndromic inheritance of facial clefting is multifactorial. Familial inheritance of both cleft lip and palate occurs with varying frequency, depending on whether a parent or sibling is affected. For cleft lip with or without cleft palate, the risk rate for future offspring is 2% with only one parent affected, 4% with only one sibling affected, 9% with two siblings affected, and 10-17% with one parent and one sibling affected. For cleft palate alone, the risk rate for future offspring is 7% with only one parent affected, 2% with only one sibling affected, 1% with two siblings affected, and 17% with one parent and one sibling affected.[12, 46, 61] Chromosome aberrations such as trisomy D and E have increased incidence of clefts. Facial clefts are associated with a syndrome 15-60% of the time. More than 200 recognized syndromes may include a facial cleft as a manifestation. Common syndromes with cleft lip and/or palate include Apert's, Stickler's, Treacher Collins, Van der Woude's and Waardenberg's syndromes.[25]

HISTOLOGY OF DISTRACTED TISSUE:

The histology of the distracted site has been observed and reported in literature[2,4,25,30,38] and the process of bone formation at the site of distraction has been likened to that of interrupted healing of a fracture site. Briefly, the process of distraction creates a tension which promotes angiogenesis and increased cellularity which leads to collagen fibers being laid along the long axis of the callus at the fracture site. This region undergoes mineralization and remodeling during the consolidation phase. It is not difficult to understand that the process of distraction osteogenesis must exert some influence on the soft tissues overlying the site of distraction. In this context and to my knowledge, only three reports of such studies describe the mucosa of the distraction site [11,35,37] . This study aims to add to the existing knowledge in this aspect.

COMPLICATIONS ASSOCIATED WITH CLEFT LIP AND PALATE:

- **Feeding Problems:**

Occasionally, nasal regurgitation during breast feeding leads to detection of cleft palate in these patients. The soft palate, a soft tissue shelf, separates the nasal cavity from the oral cavity during swallowing. Among cleft palate patients, this seal is absent, leading to poor sealing and subsequent leakage of milk. In cleft lip patients a lack of circum oral seal is observed resulting in poor suckling efficiency. In these patients breast and/ or bottle feeding are therefore difficult necessitating alternate feeding techniques with/ without the use of a custom made palatal obturator may be recommended. It also requires training the care giver/ parent so that the patient does not lack for nourishment.

- **Speech Disorders :**

Errors in articulation are common in cleft palate patients, especially those involving affricates and fricatives. Other errors include stop, glides, and nasal semivowels. Velo-pharyngeal incompetence is associated with an audible escape of air from the nose during production of pressure sounds and is termed nasal emission or snort. It is estimated that 75% of patients have velo-pharyngeal competence following primary cleft palate surgery, and this can be increased to 90-95% with directed secondary procedures. Velo-pharyngeal competence is the most important determinant of articulation performance and listener understanding of speech in cleft palate patients. Others factors include dentition, associated hearing loss, and muscular and neurologic deficits. Velo-pharyngeal competence can be estimated by direct examination of the nasopharyngeal depth, palatal length, and palate movement during phonation. Flexible fibro-optic nasopharyngoscopy has the added advantage of direct visualization of palatal motion and pharyngeal wall motion with both single sounds and connected speech

- **Ear Disease :**

  Patients with an isolated cleft lip have an incidence of hearing loss similar to that in the normal population. In contrast, cleft palate is very often associated with eustachian tube dysfunction and a resulting conductive hearing loss. Eustachian tube dysfunction in these patients is due to an abnormal insertion of the levator and tensor veli palatini muscles into the posterior margin of the hard palate. In addition to middle ear effusion, the patients also appear to have an increased incidence of cholesteatoma (7%). With increasing age, the incidence of eustachian tube dysfunction decreases and in many cases normal eustachian tube function develops by mid adolescence. Otologic goals in the cleft palate patient are to provide adequate hearing, maintain ossicular continuity and adequate middle ear space, and prevent deterioration of the tympanic membrane.

- **Airway Problems**

Airway problems may arise in children with cleft palates, especially those with concomitant structural or functional anomalies. For example, Pierre-Robin sequence is the combination of micrognathia, cleft palate, and glossoptosis. Affected patients may develop airway distress from their tongue becoming lodged in the palatal defect.

- **Dental Problems**

Clefts of the lip and/ or palate can affect the dentition by changes in number, size, shape and position of both deciduous and permanent teeth. The teeth most commonly affected are those in the area of the cleft, the lateral incisors. The lateral incisors maybe missing, may exhibit twinning or show abnormalities of crown shape at the site of the cleft. These patients may also exhibit misaligned teeth and require orthodontic treatment.

FACIAL GROWTH IN CLEFT PATIENTS

Zbynek Smahel et al[77] in 1994 used cephalometry to study facial growth and development in 32 males with complete cleft lip and palate. They underwent primary periosteoplasty and were examined at 10 and 15 years. The results were compared with the development of the face in 20 males with the same type of cleft who were operated with a primary bone graft. The series with periosteoplasty showed more marked proclination of the upper anterior dentoalveolar component with the restoration of positive overjet, as compared to a persistent crossbite in males with bone graft. The lower jaw showed a larger protrusion, while individuals with bone graft were characterised at the ages of 10 and 15 years with marked posterior rotation of the mandible. The growth of the maxilla did not differ between the two series. In both series, there was a marked reduction in the growth of the maxillary depth and of upper lip height, while the highest growth rate showed parameters of nasal prominence. An increasing mandibular protrusion and maxillary retrusion resulted in an impairment of sagittal jaw relation and in flattening of the face, both of which occurred in almost all patients. The rotation of the lower jaw was not correlated with either the convexity of the face, sagittal jaw relations, or an overjet. Fixed appliances promoted a more marked proclination of upper incisors and the alveolar process than removable appliances, but they exerted no direct action on the other facial parameters studied.

Yildiz Ozturk et al[75] in 1996 conducted a study to make cephalometric evaluation of possible differences in craniofacial morphology of Turkish children with and without unilateral cleft lip and palate (UCLP). Twenty UCLP patients with a mean age of 10.75 years were compared with a control group of children without UCLP who were matched for age and sex. No patient had received orthodontic treatment. Linear and angular variables were measured from tracings of lateral cephalometric radiographs and the resulting data was evaluated statistically. Compared with children in the control group, children in the cleft group demonstrated greater flattening of the cranial base, a more retrognathic and posteriorly inclined maxilla with decreased length, a larger mandibular plane and large gonial angle, large anterior facial height and decreased posterior and upper posterior facial height.

Stellzig et al[1] in 1999 conducted a study to a) investigate whether growth increments till 6 months of age are influenced by particular factors b) to analyze whether anterior cleft reduction is dependent on the extent of the cleft width at birth and c) to examine the correlation between maxillary measurement at birth and the anterior cleft width at 6 months of age. The study design was retrospective and longitudinal and the records of 34 patients with complete unilateral cleft lip, alveolar ridge, and hard and soft palate were included in the study. All patients were treated with the same protocol and were assessed at 0 months and at 6 months of age. Maxillary plaster casts of the patients were analyzed using a computer controlled 3-dimensional digitizing system. No statistically significant differences were found between maxillary growth changes and increases in width and length. There was no significant interaction between the extent of the alveolar cleft width at birth and its reduction prior to lip closure. In contrast, significant growth differences of maxillary growth increments were found between male and female patients. The results of the study concluded that gender plays a certain role in growth changes within the first 6 months of age.

Bert Braumann et al[6] in 2003 studied the patterns of maxillary alveolar arch growth changes of infants with Unilateral Cleft Lip and Palate (UCLP). Consecutive casts of the maxilla (1 week and 3, 6, and 12 months of life) of 15 patients with complete unilateral cleft lip and palate (cUCLP) and 13 patients with incomplete unilateral cleft lip and palate (iUCLP) were studied. All patients were treated with passive palatal plates. Cheiloplasty was performed at 6 months of age. No primary osteoplastic surgery was carried out. Following digitizing with a three-dimensional laser scanner, all cast surfaces were computer reconstructed, aligned and superimposed. Distances between the surfaces were determined and expressed graphically. Computer-aided determination of defined maxillary dimensions was performed. The volumes of segmented surfaces were determined and compared. Within the first year of life, decreased sagittal but increased transverse alveolar growth for patients with iUCLP was found. The increase in alveolar crest length in patients with iUCLP was 50% less within the first year of life than in patients with cUCLP. In the same patients, the volumes of the molar segments were, on average, larger at each registration stage and the increase in these volumes larger within the first year of life. Conclusions regarding the direction and extent of growth could not be drawn from the visible level of severity of the malformation.

Hermann et al[26] in 2004 analyzed craniofacial morphology and growth in children with bilateral complete cleft lip and palate (BCCLP) and compared it with a control group with unilateral incomplete cleft lip (UICL), before any treatment as well as 20 months after lip closure. The children were drawn from a group representing all Danish children with cleft. Sixty-four children were included in the study (19 BCCLP and 45 UICL). The ages were 2 and 22 months at examinations 1 and 2, respectively. The method of investigation was infant cephalometry in three projections. The craniofacial morphology was analyzed using linear, angular, and area variables. Growth was defined as the displacement vector from the coordinate of the corresponding landmark in the x-ray at examination 1 to its coordinate at examination 2, corrected for x-ray magnification. The BCCLP group differed significantly from the UICL group. The most striking findings in BCCLP were an extremely protruding premaxilla; markedly increased posterior maxillary width; increased width of the nasal cavity; short maxilla with reduced posterior height; short mandible; bimaxillary retrognathia; severe reduction in the size of the pharyngeal airway; and a more vertical facial growth pattern. The study findings indicated that a facial type including a wide and posterior short maxilla, short mandible, and bimaxillary retrognathia might be a liability factor that increases the probability of developing cleft lip and palate.

Shetye et al[60] in 2004 reviewed the facial growth of adults in unoperated clefts. The maxilla in unoperated patients is normally positioned or protruded. The protrusion of maxilla in the unoperated adult cleft patients is limited to the non-cleft side, contributing to hemifacial maxillary prognathism. Surgical scar tissue is known to interfere with the growth of the face. Palatal surgery has more significant influence on the growth of the midface than lip surgery: therefore, it is important to reduce the effect of surgery by delaying the timing of the palatal surgery. Delaying palatal surgery until maxillary growth is complete is desirable but could lead to poor speech development. Therefore, it is essential to perform the palatal surgery before speech development. The other variables that affect the midface are the genetic makeup of the child, the amount of tissue deficiency, the timing of surgery, surgical technique and the skill of the surgeon. Surgeries continue to have some inhibitory effect on maxillary growth but it is essential to recognize and perform those surgical procedures that have the least effect on the growth of the maxilla. This will help minimize extensive orthodontic treatment and eliminate major secondary orthognathic surgery for correction of the abnormal growth of the maxilla.

Servet Dogan et al[59] in 2006 investigated the craniofacial morphology of Turkish children with unilateral complete cleft lip and palate who had operations to close the cleft lip when they were 3 months old and to close the palate at 12 months. They were not given orthopaedic or orthodontic treatment. 42 patients with unilateral complete cleft lip and palate (UCCLP) with 45 control children without UCCLP were compared. The children with UCCLP had considerable morphological deviations compared with the matched children without clefts. They had significantly shorter and more posteriorly positioned maxillas. There was also an increase in cranial base angle, mandibular plane and gonial angle. There was a reduction in the posterior facial height and an increase in the anterior facial height. The profile of the soft tissue was more convex and the upper lip was thinner than in the children in the control group, and their noses were placed relatively further backwards and downwards.

Geraedts et al[23] in 2007 carried out a retrospective, mixed longitudinal study to assess the long-term outcome of early secondary closure and premaxilla osteotomy in 40 bilateral cleft lip and palate patients who underwent early secondary osteotomy of the premaxilla and bone grafting at the age of 8-12 years. The study revealed that a trend towards maxillary growth retardation existed which was partially compensated by orthodontic and dental treatment. Since the results are comparable to those reported for the Oslo group with regard to maxillary growth retardation, the surgical protocol followed does not require revision. Considering the benefits, i.e. closure of alveolo-palatal cleft, continuity of dental arch, eruption of canine in the graft and closure of oro-nasal communications, this mode of treatment should be continued.

Wolford et al[39] in 2008 evaluated the long-term effects of orthognathic surgery on subsequent growth of the maxillomandibular complex in the young cleft patient. 12 young cleft patients, who underwent Le Fort I osteotomies, with maxillary advancement, expansion, and/or downgrafting, by use of autogenous bone or hydroxyapatite grafts, when indicated, for maxillary stabilization were evaluated.

Before surgery, all patients had relatively normal growth. After surgery, cephalograms showed statistically significant growth changes with decreasing maxillary depth and increased proclination of upper anteriors. Post surgically, maxillary growth was predominant in the vertical vector with no anteroposterior growth. No significant difference was noted in the effect on growth in patients with unilateral clefts versus those with bilateral clefts. The presence of a pharyngeal flap was noted to adversely affect growth, whereas simultaneous mandibular surgery did not. They concluded that orthognathic surgery may be performed on growing cleft patients when mandated by psychological and/or functional concerns. The surgeon must be cognizant of the adverse postsurgical growth outcomes when performing orthognathic surgery on growing cleft patients with the possibility for further surgery requirements. Performing maxillary osteotomies on cleft patients would be more predictable after completion of facial growth.

Hiroshi Iwasaki et al[28] in 2009 conducted a study to determine intrinsic effects of congenital cleft palate on craniofacial morphology by retrospectively comparing craniofacial features between children with unoperated submucous cleft palate and noncleft children with normal occlusion in prepuberty. Twelve Japanese children (7 girls and 5 boys) with unoperated submucous cleft palate at age 9 were examined cephalometrically. In cleft children, anteroposterior length of the maxilla was significantly short, and the posterior part of the maxilla was more in the anterior position compared with noncleft children. Also, the anterior parts of the maxilla tended to be slightly retruded in cleft children. In the current study, characteristic differences in the craniofacial morphology between the unoperated submucous cleft palate children and the noncleft children in prepuberty were recognized and showed that the craniofacial deviation in the cleft children can be defined as the intrinsic effects of congenital cleft palate itself.

Ye et al[73] in 2010 investigated the effects of the cleft itself and palatoplasty on the development of dental arch morphology. 30 adult patients with operated unilateral complete cleft palate and 30 adults with unoperated cleft palate were included in the study. The result showed that all maxillary arch widths and anterior arch length of the operated group were significantly smaller than those of the unoperated growth. The influence of cleft palate on the development of maxillary arch is limited to the vicinity of the cleft in the anterior region only. Palatoplasty is a main cause resulting in the construction of the maxillary arch, while inhibiting the sagittal development of the anterior arch

Ysunza et al[76] in 2010 studied the maxillary growth in a group of cleft palate patients operated on around 4-6 months of age, and receiving further orthodontic treatment. The controversy about timing of cleft palate repair has been focused on early closure for improved speech versus delayed repair for enhancing maxillary growth. Early palatal repair enhances phonological development decreasing the frequency of articulation disorders associated with velopharyngeal insufficiency (VPI). In contrast, it has been described that early surgery adversely affects maxillary growth. In this study, a group of 20 cleft palate patients, who were subjected to early minimal incision palatopharyngoplasty around 4-6 months of age, were followed for a minimum of 10 years (range: 10-14 years). All patients received the same orthodontic management, starting at 4 years of age. None of the patients had orthognathic surgery or alveolar bone grafting. SNA, SNB, ANB, and WITS cephalometric measures were compared. A non-significant difference was found in all measurements between the two groups. The authors concluded that early cleft palate repair enhances phonological development. Although maxillary growth was affected in cleft palate patients, appropriate orthodontic treatment could achieve normal maxillary growth as measured during adolescence.

STUDIES ON CLEFT:

Houston et al[29] in 1989 studied the surgical and postsurgical changes in maxillary position following transpalatal osteotomy at the Le Fort I level in 30 patients with clefts of lip and palate and the results were evaluated cephalometrically. By superimposing on natural reference structures, a more accurate and detailed evaluation of change in maxillary position was possible than by using conventional cephalometric analyses. The mean horizontal advancement was 9 mm, with a mean vertical change of 3 mm. While it is customary to express postsurgical relapse as a percentage of surgical change, the most remarkable finding to emerge from this study was the variability in surgical and postsurgical change which would be obscured by concentrating on descriptive statistics. Postsurgical change was related to the amount of surgical change, but the correlation coefficients are quite low, and so other factors must be responsible for a significant proportion of any relapse.

Posnick et al[53] in 1993 said that for the cleft patient presenting in adolescence with a jaw discrepancy and malocclusion, misinformation and limited available surgical and dental expertise often prevents a favorable facial reconstruction and dental rehabilitation. A major advantage of the modified Le Fort I osteotomy is its ability to simultaneously close cleft dental gaps, resolve oronasal fistulas, manage skeletal defects, stabilize dentoalveolar segments and correct jaw deformities. When a thoughtful staging of reconstruction is undertaken, individuals born with cleft lip and palate can reach adolescence after undergoing only a limited number of operations and interventions, without negative attention being drawn to their original malformation. The adolescent with bilateral cleft lip and palate (BCLP) undergoing orthognathic surgery may have multiple residual clefting problems, including a mobile, dysplastic premaxilla and hypoplastic lateral maxillary segments, with each segment misaligned in three dimensions. These problems are commonly compounded by residual oronasal fistulas, bony defects, soft-tissue scarring from previous surgery, and the congenital absence of the maxillary lateral incisor teeth with resulting cleft-dental gaps. They described modifications of the Le Fort 1 osteotomy that allow for the simultaneous routine and safe management of these deformities. Results of this surgery on 22 consecutive patients were reported, with findings of follow-up ranging from 1 to 5 years. The long-term parameters reviewed included closure of residual oronasal fistulas, stabilization of the premaxilla, cleft-dental gap closure, maintenance of attached gingiva at the cleft site, maintenance of a positive overjet and overbite, the need for prosthetics to complete dental rehabilitation, and surgical morbidity.

Posnick et al[54] in 1995 reviewed the complications and long-term results of a consecutive series of adolescents (67 males, 49 females; age range 15 to 25 years; mean 18 years) born with a cleft who underwent primary repair in childhood and later developed a jaw deformity and malocclusion that required orthognathic surgery. Orthognathic procedure included a Le Fort I osteotomy; simultaneous sagittal split osteotomies of the mandible; and 87 underwent osteoplastic genioplasty. The preoperative clinical examination varied according to cleft type and individual variation, but all patients had maxillary hypoplasia. Additional cleft-related deformities included residual oronasal fistula and bony defects, clefted alveolar ridges that retained dental gaps and mobile premaxilla that lacked union to the lateral segments. The long-term maintenance of overjet and overbite measured directly from the late (> 1 year) postoperative lateral cephalometric radiograph indicated that 97 percent of patients maintained a positive overjet and 89 percent maintained a positive overbite; 5 percent shifted to a neutral overbite. The methods used to manage jaw deformity, malocclusion, residual oronasal fistula, and bony defects in adolescents born with a cleft are safe and reliable and offer the patient an enhanced quality of life. They also provide a stable foundation in which final soft-tissue lip and nose revisions may be carried out.

Molina et al[47] in 1998 stated that in patients with cleft lip and palate, normal growth of the maxilla may be impaired by early cleft repair, and many of them do not respond to orthodontic procedures alone. In the last few years, distraction techniques have been used successfully to correct the hypoplastic human mandible. Maxillary distraction is an alternative technique to correct maxillary hypoplasia during mixed dentition. The procedure was performed in 38 patients aged between 6 and 12 years. Photographs, postero-anterior and lateral cephalograms, and dental models are obtained preoperatively (as well as an orthopantomogram) to locate the tooth buds. Using a facial mask and an intraoral fixed appliance system as an anchorage, distraction was initiated on the fifth postoperative day. A combination of forward and downward distraction forces can be used to achieve simultaneous advancement and elongation of the hypoplasic maxilla. The aesthetic results are excellent, and the nasolabial angle is increased, including a more anterior projection of the upper lip. Nasal breathing is improved as well as the air flow and patency of the nasal airway. Velo-pharyngeal function remains unchanged after the procedure. The follow-up in this series varied from 6 months to 3 years. No relapses were observed.

Duffy et al[21] in 2000 examined the facial surfaces of cleft children and unaffected children aged 8–11 years with the aim of identifying and assessing differences in their facial surface morphology. The investigation was carried out using an Optical Surface Scanner, an instrument that utilizes laser light to construct and archive a three-dimensional image of the face suitable for linear measurement and direct surface comparisons. Thirty-nine cleft lip and palate (CLP) patients and 25 unaffected subjects were studied and a range of linear facial measurements was compared. Three dimensional differences between the cleft subgroups and the control group were visualized by superimposition of averaged cleft scans over the averaged control group images. Statistically significant dimensional differences in interocular width, nose base widths, mouth widths, and nose base/mouth width ratios were found between the cleft group and the control group. Qualitative differences over the whole of the face were readily demonstrated between the groups by superimposition. Face width and submandibular area depth differed consistently between the groups, the cleft face appearing narrower with a deeper submandibular area. The study concluded that significant differences existed between the facial surface morphology of CLP patients and control subjects.

Claudia Zuniga et al[9] in 2000 conducted a study to compare craniofacial relationships, position, and curvature of the cervical spine between children with cleft lip and cleft palate who had been operated on and children without clefts. This study was performed in 28 children with mixed dentition. They were divided into two groups. The study group included 14 children with unilateral operated cleft lip and cleft palate, ranging in age from 6 to 12 years, who clinically presented with a short upper lip, abnormal lip seal, and inhibition of sagittal development of the midface that was radiographically assessed. The control group included 14 children without clefts, ranging in age from 8 to 11 years. All of them had normal lip seal, nasal breathing, and a clinically normal body posture. A lateral craniocervical radiograph in a self-balanced natural head position in an erect posture, and without using a head holder, was taken for each child of both groups, with the mandible in maximum intercuspation and lips in habitual posture. The true vertical was marked on all the films. Specific angular and linear dimensions were used to assess the craniocervical relationships, as were the position of the cervical spine, its curvature, or both. The study group presented a significant increase in the extension of the head on the neck, forward position of the cervical spine, and a decrease in the curvature of the cervical spine in comparison with the children without clefts. These results were more relevant considering that the study group also presented higher significant values of lower facial height than children without clefts.

Bert Braumann et al[5] in 2002 evaluated accuracy, precision, and validity of a newly developed 3-D digital computer-aided procedure to visualize and metrically analyze the growth of the edentulous maxilla of infants with CLP. The method was applied to 10 infants with complete unilateral CLP. Consecutive casts of the maxilla (1 week and 3, 6, and 12 months) of each patient were optically measured with a 3-D laser scanner. Following digitizing, the casts were computer reconstructed, aligned, and superimposed using specialized computer software. The distances between the surfaces were measured. Additionally, the surfaces were segmented perpendicular to the alveolar crest, the reference points being C1, C1', C2, C2', and I. The volumes of the resulting segments were determined and compared with one another. The newly developed analysis enabled visualization of the extent and direction of morphological changes in the maxilla of infants with CLP. With this method it was possible to quantify these changes of the volume of defined alveolar segments. The 3-D analysis developed is an ideal tool for the examination of 3-D morphological changes in the edentulous maxilla of patients with CLP. The results will serve as the starting point for a longitudinal study on the efficacy of different methods, not only of presurgical infant orthopedics but also of surgical procedures.

Tadashi Yamanishi et al[65] in 2009 conducted a study to evaluate the palatal morphology of patients with complete unilateral cleft lip and palate after early 2-stage palatoplasty (ETS) consisting of soft palate closure by a modified Furlow palatoplasty at 12 months of age and hard palate closure at 18 months of age. They compared the result obtained with the palatal morphology obtained by Wardill-Kilner push-back palatoplasty (PB) at 12 months of age with that of children with noncleft palate. In the present study the authors investigated whether ETS can result in better palatal development than conventional PB. Thirty subjects were treated by ETS and 42 underwent PB. At 4 years of age, the anteroposterior palatal length of ETS was significantly longer than that of PB and the transversal palatal width of ETS was also markedly wider than that of PB at every point measured. Furthermore, ETS showed potential catch-up growth in the anteroposterior palatal length from 12 months to 4 years of age. These results demonstrate that ETS has a considerable benefit for the palatal development of patients with complete unilateral cleft lip and palate compared with PB.

TIMING OF SURGERY:

DeLuke et al[18] in 1997 reviewed the literature on timing and technique for primary repair and reports on the outcome for a consecutive group of patients treated by a single surgical protocol at a Cleft Palate Clinic. Twenty-eight patients treated by a standardized clinical protocol from infancy through adolescence were evaluated with respect to the need for orthognathic surgery to correct jaw size discrepancy. For each patient, data was collected regarding type of cleft deformity, total number of surgical procedures from infancy, surgeon performing the primary repair, and the need or indication for orthognathic surgery. Twenty-five percent of patients treated by this protocol required orthognathic surgery because of anteroposterior jaw size discrepancy. The number of prior operations was not a significant factor. The need for orthognathic surgery was seen in all types of CLP deformity. The results of this study paralleled other larger cohort studies with respect to the percentage of patients requiring orthognathic surgery. The number of prior operations does not significantly affect the later need for orthognathic surgery.

Da Silva Filho et al[17] in 2003 studied the influence of lip repair on craniofacial morphology in patients with complete bilateral cleft lip and palate (BCLP). They compared two groups of adult male patients with complete bilateral cleft lip and palate on the basis of lateral cephalometric radiographs. The first group comprised of 13 unoperated BCLP patients and the second group comprised of 14 patients who had been operated on the lip prior to 2 years of age. The results suggested that lip repair had a significant influence on certain areas of the craniofacial complex, mainly the premaxilla and the upper incisors. The most significant findings consequent to lip repair consisted of reduction of the pre-maxillary anterior projection and lingual tipping of the upper incisors. Retropositioning of the premaxilla, especially in the alveolar part is a desired effect of the lip repair in complete BCLP. Such effects on the projected premaxilla is usually beneficial, except when the exceedingly severe lip pressure, unfavorable growth pattern, or both retropositions the midface profile beyond acceptable sagittal limits.

Cohen[10] in 2004 stated that every effort should be made to achieve the best possible results at the time of lip and palate repair. Appropriate and extensive evaluation, short- and long-term planning with optimal timing for each procedure, close cooperation with the members of the craniofacial team, selection of the most appropriate technique, careful execution, and close follow-up are prerequisites for success. Additional surgical procedures or revisions are required to improve appearance and function and to manage unfavorable results of previous interventions. Such procedures should be also planned carefully, taking into consideration all aspects of the deformity to provide patients with superior habilitation.

Liao et al[40] in 2006 investigated whether timing of hard palate repair had a significant effect on facial growth in patients with unilateral cleft lip and palate (UCLP). Retrospective longitudinal study design was used and 104 UCLP patients who had hard palate repair by 13 years were included in the study. They concluded that timing of hard palate repair significantly affects the growth of the maxilla in patients with

UCLP. Late hard palate repair has a smaller adverse effect than does an early hard palate repair on the growth of the maxilla. This timing primarily affects the anteroposterior development of the maxillary dentoalveolous and is attributed to the development being undisturbed before closure of the hard palate.

STUDIES ON DISTRACTION VS SURGERY:

Yeow et al[74] in 1999 presented a case of midface distraction in a bilateral cleft lip and palate patient. The patient was a 10-year-old who underwent a high LeFort I osteotomy followed by placement of the rigid external distraction halo. Distraction was commenced on the fifth postoperative day at a rate of 1 to 1.5 mm per day until a total of 17 mm of maxillary advancement had been achieved. There were no complications and the patient was followed up 9 months post distraction. Results showed that the patient had improved facial aesthetics and dental occlusion which was overcorrected to a Class III relationship. Velopharyngeal function was unaffected. Distraction osteogenesis of the midfacial skeleton in cleft patients offers the possibility to remodel not only the underlying bony skeleton but also the soft tissues of the face and palate.

Wen – Ching Ko et al[70] in 2000 conducted a study to evaluate the soft tissue profile changes after maxillary advancement with distraction osteogenesis (DO). Sixteen subjects who had maxillary advancement with rigid external distraction after a high Le Fort I osteotomy were included. The subjects included UCLP, BCP & CP. Pretreatment and post treatment lateral cephalograms were compared to evaluate the changes in soft tissue profile. The preoperative facial concavity was reduced by 15.59

degrees, and the nasal tip moved 3.75 mm forward and 2.05 mm upward. These changes were positively correlated with the change of ANS position. The soft-tissue-to-hard-tissue ratio was 0.53:1 for nasal tip and ANS & the ratio was negatively correlated with the age of the patient. Maxillary DO improved the soft tissue profile by increasing nasal projection, normalizing the nasolabial angle, and making the upper lip more prominent. More upper anterior tooth show in the rest position was obtained, but the upper lip length did not change. The concave facial profile became convex, with improved facial balance and aesthetics.

Wiltfang et al[71] in 2002 studied the long term results of distraction osteogenesis of the Maxilla & Midface. Eight patients were treated by osteodistraction to correct hypoplasia of the maxilla and midface of various origins. Among them were five patients who were treated by high LeFort I osteotomies and insertion of subcutaneous intraoral distraction devices in the malar region. In the remaining three patients, extraoral distraction devices were applied after LeFort II and III osteotomies. Distraction osteogenesis was successful in all cases. All patients were kept under orthodontic supervision before, during, and after osteodistraction. Long-term cephalometric and clinical evaluation after a mean follow-up period of 24 months in the intraoral distraction group and 12 months in the extraoral distraction group showed stable results concerning the skeletal and dental relations.

Kumar et al[36] in 2006 studied the usefulness of LeFort 1 internal distraction osteogenesis as an alternative to one step orthognathic advancement. The advantages of distraction osteogenesis were gradual lengthening and earlier treatment in growing patients. Patients with maxillary deficiencies were divided into three groups: group

1, mild to moderate deficiency (< 10 mm.) treated with conventional orthognathic procedure; group 2, severe deficiency (> or = 10 mms.) treated with conventional orthognathic procedure and group 3, distraction procedure for severe deficiency. Results demonstrated that LeFort 1 internal distraction for severe cleft maxillary deficiency leads to better dental occlusion, less relapse and better speech results.

Cheung et al[8] in 2006 published a meta analysis to provide evidence based data on the choice between distraction osteogenesis or conventional osteotomies for Cleft lip and Palate patients. The study concluded that distraction osteogenesis tends to be preferred to conventional osteotomy for younger CLP patients with more severe deformities. In such cases, it is feasible to use distraction to correct moderate to large defects of the maxilla by either complete or incomplete LeFort 1 osteotomy, and a concurrent mandibular osteotomy was less frequently required. Intra operative and post operative complications were uncommon with either technique, and some of the traditional ischemic complications related to conventional osteotomy were replaced by infection of the oral mucosa due to prolonged retention of the distractor. Both distraction osteogenesis and conventional osteotomies can deliver a marked improvement in facial aesthetics.

Cheung et al[8] in 2006 conducted a randomized controlled study aiming to compare the postoperative clinical morbidities in cleft lip and palate patients treated with distraction osteogenesis versus conventional orthognathic surgery. 29 cleft lip and palate patients with moderate maxillary hypoplasia requiring a maxillary LeFort I advancement of 4 to 10 mm were randomized into 2 groups for either internal maxillary distractors or immediate fragment transposition using miniplates and screw fixation. Clinical morbidities were recorded using standardized questionnaires. Two of the 15 patients in the distraction group developed infection around the distractor and one patient had occlusal relapse. In patients with conventional surgery, the complications included intraoperative hemorrhage, plate exposure and occlusal relapse. They concluded that there were no major differences in the clinical morbidities between the osteotomy and distraction groups. Distraction provided better skeletal stability, whereas there was a significant amount of skeletal relapse in the first 12 weeks after conventional cleft maxillary osteotomy.

Gulsen et al[24] in 2007 conducted a study to evaluate the results of maxillary advancement by using internal LeFort I distractors on 6 patients with unilateral cleft lip and palate who had maxillary hypoplasia. Lateral cephalograms were taken to evaluate 3 months before and 12 months after distraction. It was concluded that effective and easy distraction is possible with internal LeFort I distractors in cleft lip and palate patients who require maxillary advancement.

Baek et al[7] in 2007 compared treatment outcome and relapse between maxillary advancement surgery with LeFort I osteotomy and maxillary distraction osteogenesis in patients with cleft lip and palate with maxillary hypoplasia. The sample consisted of a maxillary advancement surgery with LeFort I osteotomy group (group 1, N= 14, mean age, 21.7 years) and a maxillary distraction osteogenesis group (group 2, N = 11, mean age, 16.3 years). Although the amounts of forward movements of point A, upper incisor, and upper lip were greater in group 2, there were no significant differences in the amounts of relapse between the two groups. Counterclockwise rotation of the palatal plane was observed in group 2 as a result of downward movement of posterior nasal spine (PNS), whereas group 1 had clockwise rotation of palatal plane because of downward movement of anterior nasal spine (ANS). The amounts of relapse in vertical movements of PNS and upper incisor were significantly different between the two groups. The amount of required maxillary advancement, vector control of palatal plane, and vertical position of upper incisor would be important factors when planning a surgical treatment in patients with cleft lip and palate with midface hypoplasia.

Chua et al[14] in 2010 compared the long term stability of distraction osteogenesis (DO) and conventional orthognathic surgery (CO) in patients with cleft lip and palate (CLP). CLP patients requiring maxillary advancement were randomized and assigned to either CO or DO. Results demonstrated that in the CO group, the maxillae relapsed backwards and upwards, whereas in the DO group, it advanced more forward and downward over 5 years. Hence it was concluded that distraction of the cleft maxillae can achieve better long term skeletal stability in maintaining its advance position than CO.

STUDIES ON INTERNAL AND EXTERNAL DISTRACTERS

Block et al[44] in 1994 carried out a pilot study to apply the principle of distraction osteogenesis for advancing the anterior maxilla of a dog. After an anterior maxillary osteotomy, the anterior segment was advanced 10 mm in 10 days. The authors found that soft and hard tissue formation across the distraction gap resulted in complete healing without a soft tissue defect.

Block et a4[35] in 1995 used the principle of distraction osteogenesis to advance the anterior maxilla of the dog using a totally tooth supported distraction device. After an anterior maxillary osteotomy, the distraction device was activated 0.5mm every 12 hours to advance the anterior segment 10 mm in 10 days. Radiographic measurements indicated that a tooth borne maxillary distraction device will result in significantly greater dental movement than skeletal movement and that skeletal fixation may be needed for appliances used to advance the maxilla.

Polley et al[52] in 1998 said that patients with severe maxillary hypoplasia secondary to congenital facial clefting present numerous challenging problems for the reconstructive surgeon. Traditional surgical/orthodontic approaches for these patients often fall short of expectations, especially for achieving normal facial aesthetics and proportions. They presented their clinical experience and cephalometric results with the use of rigid external distraction for the treatment of patients with severe maxillary deficiency. Eighteen consecutive orofacial cleft patients with severe maxillary hypoplasia were treated with maxillary distraction osteogenesis. Criteria for patient selection included severe maxillary hypoplasia with negative overjet of 8 mm or greater, patients with normal mandibular morphology, and patients with full primary dentition or older. There were 10 unilateral cleft lip and palate patients, 6 bilateral cleft lip and palate patients, and 2 patients with severe congenital facial clefting. A maxillary splint was prepared for each patient, and all patients underwent a high Le Fort I maxillary osteotomy. In all face mask distraction patients, the initial maxillary hypoplasia was under corrected. Maxillary distraction osteogenesis with rigid external distraction permits full correction of the midfacial deficiency, including both the skeletal and soft-tissue deficiencies. Rigid external distraction in patients with severe maxillary hypoplasia allows full correction of the deformity through treatment of the affected region only. It offers the distinct advantage of correcting these severe deformities through a minimal procedure. Rigid external distraction had dramatically improved their treatment results for patients with severe cleft maxillary hypoplasia.

Mommaerts[43] in 1999 reported on a bone-borne titanium device with interchangeable expansion modules that used a callous distraction policy. Conventional devices used for surgically assisted rapid palatal expansion were tooth-borne. Dental fixation entails a number of possible drawbacks such as loss of anchorage, skeletal relapse during and after expansion period, cortical fenestration and buccal root resorption. To overcome these short comings, a new none borne device was described. Conventional devices used for surgically assisted rapid palatal expansion are tooth-borne. Dental fixation entails a number of possible drawbacks such as loss of anchorage, skeletal relapse during and after the expansion period, cortical fenestration and buccal root resorption. A bone-borne titanium device with interchangeable expansion modules, used with a callous distraction policy, was presented.

Swennen et al[64] in 1999 stated that Cleft lip and palate patients can present with a maxillary retrusion with tendency to Class III malocclusion after cleft repair. Maxillary distraction osteogenesis is a technique that provides simultaneous skeletal advancement. They reported on six nonsyndromic cleft lip and palate patients, ages 12 to 16 years (mean, 13.8 years), who underwent maxillary distraction; four with unilateral and two with bilateral cleft lip and palate. After an incomplete LeFort I osteotomy; a latency period of 3 days was respected. On Postoperative Day 4, distraction was initiated through anterior traction on a Delaire facial mask using distraction forces of 900 gm. The aesthetic improvement obtained by maxillary distraction osteogenesis during the permanent dentition to correct maxillary retrusion in these cleft lip and palate patients was impressive. Skeletal advancement varying from 1 to 3.5 mm (mean, 1.7 mm) was found. However, significant dentoalveolar compensations occurred in three patients. This was due to the dental anchorage of the distraction device and can be avoided only by the use of skeletal fixation.

Hierl et al[27] in 2000 stated that distraction osteogenesis of the midface offers new possibilities for the treatment of large sagittal discrepancies between the upper and lower jaws. The use of an extraoral halo-borne distractor, which allows free three-dimensional vector control, may cause problems in the connection between the midface and the distractor. To overcome these difficulties, they presented a new modular retention system to gain bone anchorage whenever a tooth borne appliance was not suitable. Distraction osteogenesis with an extraoral appliance is therefore possible even in edentulous elderly patients.

Swennen et al[63] in 2000 reported DO in Class III malocclusion. Two 13-year-old patients, born with non-syndromic cleft lip and palate underwent maxillary distraction--one had a bilateral, the other a unilateral complete cleft lip and palate. Maxillary distraction osteogenesis is a challenging technique to treat severe maxillary retrusion. Maxillary advancement by distraction has the advantage to provide new bone in combination with simultaneous expansion of the soft-tissue functional matrix. Cleft lip and palate patients can present with severe maxillary retrusion. Maxillary advancement in these two patients was performed using an external distraction device in combination with titanium miniplates as a skeletal maxillary anchorage. After a complete LeFort I osteotomy with pterygomaxillary dysjunction, a latency period of 3 days was respected. On the fourth postoperative day, distraction was initiated at the rate of 1 mm/d. Preoperative clinical photographs, dental casts, lateral cephalograms, and panoramic radiographs were taken. Further lateral cephalograms were obtained after the latency period, after completion of the active period of distraction, at the completion of the consolidation period, and at 6 and 12 months postoperatively. The aesthetic outcome was excellent and skeletal advancement of 8 and 7 mm was measured without dentoalveolar compensations.

Kebler et al[51] in 2001 said that the use of distraction osteogenesis in the hypoplastic maxilla and midface was still controversial. 25 patients treated with osteodistraction technique were included. Among those, were four patients who were treated by high LeFort I osteotomies and insertion of a newly developed subcutaneous distraction device in the malar region. Distraction osteogenesis was successful in all four cases resulting in a mean sagittal bone gain of 12 mm at the level of distractor fixation. All patients were kept under orthodontic supervision during osteodistraction. The final occlusal relation was satisfactory. Cephalometric measurements after distraction showed an anterior rotational movement of the midface region. As the question of relapse and further growth is still not clear, Delaire masks were used to stabilize the surgical result after removal of the distractor. The importance of long-term follow-up was stressed in their study.

Tong et al[68] in 2003 presented a case report on the use of interdental distraction osteogenesis for correction of maxillary hypoplasia and orthodontic tooth alignment. The principle of distraction can be applied to the dentoalveolar region. Vertical height augmentation of the alveolus and creation of new edentulous ridge were some of the applications of distraction. The application of interdental distraction osteogenesis to the management of a case of maxillary hypoplasia with severe dental crowding was presented to illustrate the feasibility of utilizing this principle. New bone was created which was used for tooth alignment and simultaneous correction of maxillary hypoplasia.

Karakasis et al[20] in 2004 presented a case report on the use of internal distraction for the advancement of the anterior maxilla. They said that several techniques of distraction osteogenesis have been applied for the correction of compromised midface in patients with cleft lip, alveolus and palate. They presented a technique of callus distraction applied in a specific case of hypoplasia of the cleft maxilla with the sagittal advancement of the maxilla thus not affecting velopharyngeal function. The authors concluded that application of distraction osteogenesis for the advancement of the anterior maxillary segment in cleft patients offered many advantages.

Gateno et al[31] in 2005 conducted a pilot study to test a new Le Fort 1 internal distraction device. The device was used in 3 patients with cleft lip and palate and severe maxillary hypoplasia who needed maxillary advancements in excess of 12 mm. The distractors were pre-bent and installed on the stereolithographic model and activated to advance the maxilla. Surgery was performed in a conventional manner, and distraction was started after a 7-day latency phase at the rate of 1 mm/day and continued until the presurgical plan was achieved. The distractor was removed after a 3-month consolidation phase. This new Le Fort I internal distraction device successfully distracted the maxillae as planned in all 3 patients. A clockwise rotation of the maxilla was observed with a tendency to a posterior open bite. Postoperative radiographs also showed that the actual distraction vectors differed from the planned vectors. Excellent new bone formation at the osteotomy sites was evident on radiographs. The study also showed that the actual distraction vector differed from the planned vector. This discrepancy was caused by a clockwise rotation of the maxilla during the distraction. Finally, the study showed a variable relapse rate not previously reported in maxillary distraction.

Xiao-Xia Wang et al[72] in 2005 reported on internal midface distraction for the correction of severe maxillary hypoplasia secondary to cleft lip and palate. 10 patients with severe maxillary hypoplasia secondary to cleft lip and palate were selected. They were treated with three kinds of internal distraction devices. Successful maxillary advancement ranging from 5 to 15 mm was measured from preoperative and post operative cephalograms. Orthodontic therapies were adopted before and after midface distraction. After the consolidation period, dense new bone was found to have formed in the distraction gap. During the follow up period, the position of the maxilla and the final occlusal relationship were stable and acceptable and no obvious relapses were seen. They concluded that midface distraction was an ideal choice for the correction of severe maxillary hypoplasia secondary to cleft lip and palate.

Jayade et al[13] in 2006 noted that maxillary distraction osteogenesis delivers excellent results, particularly in patients with clefts. In the past, devices such as the conventional facemask and the rigid external distraction device have been used to correct maxillary hypoplasia after a Le Fort I osteotomy. They described a new device, the Glasgow extra-oral distraction device. They found that the extent of skeletal and dental stability of corrections achieved in 10 patients with maxillary hypoplasia associated with clefts was satisfactory. The device costs little, can be produced in developing countries, and provides effective treatment for severe secondary deformity associated with clefts.

Van Sickels et al[33] in 2006 reviewed the workup, experience, and preliminary results with the use of internal distraction osteogenesis for maxillary hypoplasia. 10 patients in whom maxillary distraction osteogenesis was done with internal distractors were included. Follow-up of 6 months or more was available for 8 patients. Latency prior to the start of distraction was 3 to 7 days and varied with the age of the patient. Distraction occurred at approximately 1 mm per day with an average distraction length of 8.5 mm. Excellent occlusal results were obtained in 5 patients. Major complications including nonunion and failure to achieve acceptable occlusal results were observed in 3 patients. Minor complications including pain and loosening of the distracter devices were observed in 2 patients, but did not appear to affect the esthetic and functional results. Distraction osteogenesis is a useful alternative to traditional orthognathic surgery to treat maxillary hypoplasia. Internal distractions are attractive to patients, but are more difficult to place and can cause discomfort to patients when trying to achieve an ideal primary vector of distraction. Stereolithographic models can help with placement of the device. Changes in design of distractors may help with patient discomfort.

Gunaseelan et al[55] in 2007 reported on tooth borne palatal distractor for anterior maxillary distraction. Traditionally, tooth borne palatal distractors have been used for rapid maxillary expansion in children and surgically assisted maxillary expansion in adults. The use of palatal distractor in the anteroposterior direction to advance a retruded maxilla after an alveolar osteotomy was already reported. The authors modified this and presented an alternative technique involving distraction of the entire segment of the cleft anterior maxilla by a tooth borne palatal distractor. The technique of usage and the results obtained were illustrated with 2 cases of cleft lip and palate.

Distraction osteogenesis of the Le Fort I segment is advocated for patients who require significant advancement of the maxilla or who have a soft tissue envelope compromised by scar tissue. David Kahn et al[12] in 2008 presented a technique for maxillary distraction using an interconnecting intraoral device anchored to the malar prominences above the osteotomy and either the maxilla and/or the dentition below the level of the osteotomy. Ten patients with nonsyndromic cleft lip and palate, underwent Le Fort I maxillary distraction osteogenesis for management of maxillary hypoplasia. A Le Fort I osteotomy was performed and an Intraoral Midface Multi-Vector Distractor was placed leaving a 1 mm to 2 mm distraction gap. After a 2 to 4 day latency period, distraction was performed at a rate of 1 mm a day. Once the desired occlusion was achieved, the device was left in place for a minimum of 2 months for consolidation. Sella – Nasion – A point & overjet values increased considerably between the pre-treatment & post treatment. The results remained stable at a follow up of 30 months. This device design allowed the forces of distraction to be shared across a larger surface area delivering a uniform and reliable vector of distraction with increased stability.

Scolozzi et al[58] in 2008 presented a systematic review of literature reporting the use of external distraction osteogenesis (DO) and internal DO in the treatment of severe maxillary hypoplasia in cleft lip and palate patients. This review demonstrated that external and internal DO in the treatment of severe maxillary hypoplasia in cleft lip and palate patients (i) is a reproducible and valuable alternative to standard orthognathic procedures, (ii) allows for a global improvement in facial aesthetics (iii) allows a maxillary correction in patients during the period of mixed dentition and (iv) allows either for an unchanged or better velopharyngeal function.

Wang et al[69] in 2009 evaluated the feasibility of anterior maxillary segmental distraction to correct maxillary hypoplasia and severe dental crowding in cleft lip and palate (CLP) patients. 7 patients with maxillary hypoplasia, shortened maxillary dental arch length and severe anterior dental crowding secondary to CLP were selected for the study. After anterior maxillary segmental osteotomy, 3 patients were treated with internal distraction devices and 4 with rigid external distraction devices. An average of 10.25 mm anterior maxillary advancement was obtained in all patients after 10-23 days of distraction and 9-16 weeks of consolidation. The sella-nasion-pointA angle increased from 69.5^0 to 79.6^0. Midface convexity was greatly improved and velopharyngeal competence was preserved. The maxillary dental arch length was greatly increased. Dental crowding and malocclusion and were corrected by orthodontic treatment. The results showed that anterior maxillary segmental distraction can effectively correct the hypoplastic maxilla and severe dental crowding associated with CLP by increasing the midface convexity and dental arch length while preserving velopharyngeal function and crowding can be corrected without requiring tooth extraction.

Okcu et al[49] in 2009 conducted a study to determine the relative movement of the teeth and bone after premaxillary distraction with a tooth borne device. The effect of this device on the anterior segment and teeth, the space formed between the anterior and posterior segments of the maxilla was evaluated and was measured on computed tomography from 10 patients. The average differences were 3.5 mm at the apex level, 5.5 mm at the alveolar ridge level and 7.4 mm at the crown level. The ratio between the movement at the apex and crown levels was 46%. These results showed that the tooth borne distractor was able to distract the anterior segment of the maxilla but it also caused anchorage loss of the maxillary incisors.

El-Sayed et al[22] in 2010 said that alveolar bone grafting is a standard method for treating alveolar cleft. To ensure the best outcome, improving the arch form as well as soft tissue quality in the area around the cleft was recommended. In this study, 11 patients who presented with alveolar cleft and collapsed maxillary arch were treated with transpalatal distraction osteogenesis followed by soft tissue surgery in some cases and by cancellous bone graft. In all cases, transpalatal distraction osteogenesis successfully corrected the transverse maxillary deficiency. One case showed a complete loss of the bone graft. Other minor complications were reported but they did not affect the final outcome.

Seda Gursoy et al[56] in 2010 conducted a study to determine the long-term outcomes of maxillary distraction osteogenesis (DO) on skeletal and dental structures of growing children with cleft lip and palate. Severe maxillary deficiencies treated with a rigid external distractor device followed by a consolidation period were included. Preoperative and postoperative orthodontic treatment lasted a mean of 14 months and

16 months, respectively. During DO, the maxilla was horizontally advanced and moved downward as indicated by the significant changes at the SNA and ANB angles and at maxillary points A, ANS, and PNS. The increase in the divergence between the maxilla and mandible (ANS-PNS/Me-Go) was found to be significant. The mandible also moved downward and backward significantly because of mandibular autorotation. The overjet increased and the overbite decreased significantly. The advancement of the upper incisors and upper molars was slightly more than the skeletal points. In a long-term follow-up of 5 years, the ANB angle and horizontal overjet continued to decrease but both values remained positive, indicating a Class I relationship. This cephalometric study of young adolescents with cleft lips and palates found great improvement in dentofacial structure after maxillary DO and stability in maxillary skeletal advancement. During a 5-year follow-up, the achieved dentoskeletal treatment outcome was partly diminished. The extreme need for maxillary advancement or facial correction because of psychosocial stress and providing an easier approach for finalizing osteotomy are the two major indications for DO treatment.

INFLUENCE ON SPEECH:

Okazaki[50] in 1993 studied the influence of maxillary advancement by osteotomy on speech in 10 patients with cleft palate. Ages at the time of surgery ranged from 16 to 26 years. Preoperatively and postoperatively, hypernasality, nasal emission on pressure consonants, and articulation disturbances were evaluated perceptually, and velopharyngeal function was evaluated by lateral cephalographic and nasopharyngoscopic studies. 8 patients showed increased hypernasality after surgery.

Nasal emission showed a similar tendency. Articulation errors were not improved postoperatively. Lateral cephalograms recorded from the patients with increased hypernasality showed increases in the shortest palatopharyngeal length and in the soft-palate-length to pharyngeal-depth ratio. Also, deterioration in velopharyngeal closure was noted postoperatively by nasopharyngoscopy in majority of the patients with increased hypernasality.

Ko et al[34] in 1999 evaluated the static velopharyngeal anatomic changes on lateral cephalograms in patients who underwent maxillary advancement through distraction osteogenesis (DO) with a rigid external distraction device and to correlate these changes with clinical speech data. The effect of maxillary advancement on speech may have benefits on articulation improvement but could compromise velopharyngeal (VP) closure by increasing the nasopharyngeal distance. Twenty-two patients who underwent maxillary advancement through DO utilizing a rigid external distraction device (age, 5.2 to 25.7 years) with various diagnoses were included. Lateral cephalograms of preoperative, immediate post distraction, and 1-year post distraction were obtained for analysis. Speech evaluation was performed preoperatively, immediate post distraction, and then at 6-month intervals, and included assessment of air pressure flow, hypernasality, and articulation with an average amount of 8.9 mm maxillary forward advancement, 14% of patients presented deterioration in hypernasality. However, 57% of patients (12 of 21) demonstrated improvement in articulation. The cephalometric analysis demonstrated an increase in nasopharyngeal depth and velar angle. The length of the soft palate remained unchanged. The deterioration of hypernasality was related to the amount of forward distraction, especially in patients without a preexisting pharyngeal flap (PF). Speech evaluation is an important aspect concerning treatment planning for maxillary distraction. The increase in nasopharyngeal depth may compromise VP closure. The increase in velar angle was considered to be part of the compensation in the VP mechanism. An adverse effect of a preexisting PF on maxillary distraction was not observed; however, it prevented postoperative hypernasality.

Steve Bureau et al[62] in 2001 conducted a prospective study to evaluate the outcome of speech after complete closure of oronasal fistulas with bone grafts and to determine the possible relationship between outcome of speech and the size and location of the oronasal fistulas. Ten unilateral cleft lip and palate patients with postoperative oronasal fistulas, ranging in age from 7 to 14 years, underwent secondary alveolar cleft repair and closure of the oronasal fistulas with an iliac bone graft were included. All patients underwent videofluoroscopic evaluation of the velopharyngeal valve, audiologic assessment, and speech evaluation preoperatively. The examinations were repeated 3 months postoperatively. Six patients had preoperative velopharyngeal competency. Of the 4 patients with slight to mild velopharyngeal incompetency preoperatively, 2 developed velopharyngeal competency postoperatively. All patients had satisfactory audiologic function preoperatively. Every patient also was intelligible before and after surgery. (80%) showed nasal emission before surgery and most of them improved postoperatively ($P < .01$). Nine patients had articulation errors before surgery, with no significant improvement postoperatively. Nasal resonance was significantly improved in selected sequences. All patients had variable levels of nasality preoperatively. The results were not related to location or size of the oronasal fistulas. A significant improvement in speech was noticeable after closure of oronasal fistulas. Early oronasal fistula closure might prevent permanent speech distortions acquired by the cleft palate patients at an early age.

John Janulewicz et al[32] in 2004 conducted a study to evaluate how advancing the maxilla would affect the speech and articulation disorders of cleft patients. The study was carried out retrospectively to evaluate speech of 54 cleft lip and palate patients who underwent maxillary advancement. This was a retrospective study in which the speech scores of 54 cleft lip and palate patients who underwent maxillary advancement was compiled and evaluated. Although 34 individuals underwent an isolated Le Fort I advancement, 20 patients had a combined Le Fort I advancement/mandibular setback operation. The following variables were recorded from both preoperative and postoperative speech evaluations: presence of a pharyngeal flap at the time of surgery, oronasal fistulas, nasality, 7 different articulation errors, velopharyngeal function assessment, and overall speech score. A decrease in competent velopharyngeal function mechanisms was noted postoperatively, increased borderline incompetence, and complete velopharyngeal insufficiency. Speech scores deteriorated significantly, whereas articulation defects insignificantly improved after surgery, with those related to the anterior dentition showing the greatest change. The frequency of hyponasality decreased after surgery. The number of cases of mild to moderate hypernasality increased. This study confirmed previous findings that patients with clefts of the lip and palate or palate alone are predisposed to velopharyngeal function alteration after maxillary advancement, particularly with borderline function preoperatively. However, the results show that surgical correction of skeletal relationships and occlusion may translate into improvements in certain aspects of speech disorders.

Anthony Stephan et al[3] in 2006 compared the effect of a cranial-based pharyngeal flap on the speech of children born with a unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), cleft palate (CP), or primary velopharyngeal insufficiency (VPI) without cleft. A total of 234 children born with clefts and 22 children born with primary VPI were evaluated. 74 children underwent pharyngeal flap surgery for VPI. The mean follow-up period was 7 years. There were significant differences in outcome among the 4 groups. The positive effect on speech of a cranial-based pharyngeal flap is greater in children born with a UCLP or CP than in those born with a BCLP. In children born with primary VPI, this operation has only a slightly positive effect on speech that shows compensatory misarticulations; in such cases, alternative surgical choices or secondary procedures may be indicated. This information should be clearly conveyed to the parents in pre-surgical consultation so that they know what to expect from the procedure and postoperative adjuvant therapy.

STUDIES ON STABILITY:

Mehra et al[42] in 2001 conducted a study to evaluate the stability of maxillary advancement using bone plates for skeletal stabilization and porous block hydroxyapatite (PBHA) as a bone graft substitute for interpositional grafting in cleft and non-cleft patients. The records of 74 patients who underwent Le Fort I maxillary advancement using rigid fixation and PBHA interpositional grafting were evaluated retrospectively. All patients also underwent simultaneous sagittal split mandibular ramus osteotomies. Patients were divided into 2 groups for study purposes: group 1 consisted of 17 cleft palate patients and group 2 consisted of 57 non-cleft patients.

Maxillary advancement with Le Fort 1 osteotomies using rigid fixation and interpositional PBHA grafting during bimaxillary surgery was a stable procedure with good predictability in cleft and non-cleft patients, regardless of the direction of vertical maxillary movement.

Thongdee et al[67] in 2005 conducted a study to evaluate the long-term three-dimensional stability of Le Fort I maxillary osteotomy in patients with unilateral cleft lip and palate (CLP) who had preceding alveolar bone grafting. Thirty patients with unilateral cleft lip and palate were included in the study. Cephalometric and study cast analyses using pre- and postoperative records (3, 6, 12, 24, and 36 months) was performed. Evaluation of surgical movement and postsurgical change at all above time intervals was carried out to determine stability of surgical maxillary movement in the horizontal and vertical planes and to identify rotational and transverse relapse. Total relapse of surgical movement was 31% in the horizontal plane and 52% in the vertical plane, as well as 30% rotational. Relapse correlated with extent of surgical movement, and most relapse occurred in the first 6 months after surgery. Alveolar bone grafting prior to osteotomy stabilizes the transverse dimension of the dental arch, but does not improve horizontal, vertical, or rotational relapse, which remains significant. Correlation of relapse with extent of surgical movement does suggest that planned over-correction is a reasonable option.

Saleh Al-Daghreer et al[57] in 2008 conducted a systematic long-term review on skeletal stability after craniofacial distraction osteogenesis. Several electronic databases key words used in the search were “distraction,” “osteogenesis,” “craniofacial,” “maxillofacial,” “stability,” “relapse,” and “recurrence.” only 6 articles were finally selected. These 6 articles reported long-term stability after craniofacial distraction osteogenesis. Sample sizes were small, and the methodological quality of the studies was poor. Although, based on the selected studies, craniofacial bone distraction osteogenesis appeared to show long-term stability; limitations of the studies merit caution in interpreting these findings. Some early relapse occurred in the first 3 years post distraction, but stability was maintained thereafter. Some methodologically sounder studies are needed to confirm the present findings.

Takahiro Kanno et al[66] in 2008 assessed the long-term skeletal stability of the repositioned maxilla, midface in patients who underwent maxillary advancement using distraction osteogenesis (DO). The study included 19 non-growing patients with maxillary hypoplasia with a Class III relationship, a normally developed mandible, and follow-up after DO exceeding 2 years. Twelve patients had midfacial hypoplasia associated with a cleft lip and palate (CLP), and 7 patients had developed noncleft-related hypoplasia. The surgical treatment included modified Le Fort I osteotomy in combination with intraoral (5 cases) or extraoral (14 cases) distraction devices. Distraction was started after a latency period of 5 to 7 days and continued until the proper convexity was obtained. Midfacial DO was successful in all cases, resulting in a mean change obtained at point A of 10.3 mm (8.4 mm horizontally, 4.7 mm inferiorly). Point A underwent a moderate amount of skeletal relapse [0.4 mm (5%) horizontally and 0.6 mm (13%) superiorly], with a mean of 8% (0.6 mm) horizontally and 19% (1.0 mm) superiorly over the mean 2.8-year (2.0-4.8 years) follow-up. After long-term follow-up, the maxillary advancement with DO was stable in both CLP and non-CLP patients with maxillary hypoplasia. In addition, our original technique using a rigid external device provided the most reliable results in terms of skeletal stability.

This retrospective study showed that DO of the maxilla gives a very stable midface, offering a promising treatment alternative for patients with maxillary hypoplasia.

Muge Aksu et al[48] in 2010 evaluated skeletal and dental stability in adult cleft lip and palate patients treated with a rigid external distraction system at the end of distraction and during the post distraction period. Lateral cephalograms of 7 patients were obtained before distraction, at the end of distraction and during the postdistraction period. The mean follow-up period was 37.3 ± 12.4 months. The assessment of findings showed that skeletal maxillary sagittal movement was achieved in a superoanterior direction. The maxillary depth angle and effective maxillary length increased significantly after distraction, whereas the palatal plane angle increased by 8°, resulting in an anterior movement of the maxilla with a counterclockwise rotation. During the postdistraction period, the maxilla showed a slight relapse of 22%. The effective maxillary length decreased and the palatal plane angle almost returned to its original position, showing 7° of clockwise rotation. The lower facial height remained stable. The upper incisors moved anteriorly and the upper first molars showed a significant mesioangular change during follow-up. After distraction, significant maxillary advancement was achieved with a counterclockwise rotation.

Daimaruya et al[15] in 2010 investigated the changes in and stability of the maxilla and soft tissue profile achieved after the application of distraction osteogenesis (DO) by use of rigid external distraction (RED) with a retention plate system in unilateral cleft lip and palate (UCLP) adult patients. 2 treatment methods in the management of maxillary hypoplasia were compared - Le Fort I osteotomy and DO. Six UCLP adult patients who underwent treatment with the RED retention plate system were examined (DO group). Changes in the positions of soft and hard tissue landmarks were calculated from lateral cephalograms taken before distraction, at the removal of the halo, and 1 year after surgery and were compared with those in 7 other UCLP patients who underwent Le Fort I osteotomy (LF1 group). The mean maxillary advancement was significantly larger in the DO group than in the LF1 group after distraction. During the follow-up period, the relapse rate of the maxilla was significantly smaller in the DO group. The DO group tended to have a higher soft tissue–to–hard tissue anterior movement ratio from the time of distraction to follow-up. The RED retention plate system improved the midfacial profile by advancement of soft and hard tissue and minimized the risk of injury to the upper lip. Using the RED system with retention plates prevented the undesirable labial inclination of upper incisors that was found in the LF1 group.

HISTOLOGY OF MUCOSA OVERLYING THE SITE OF DISTRACTION OSTEOGENESIS:

Cope JB, Samchukov ML and Muirhead DE[11] evaluated the effect of distraction osteogenesis on the gingival tissues to analyze the newly formed bone and gingiva during the consolidation period of mandibular osteodistraction using standard histologic techniques. Seventeen skeletally mature male beagle dogs were subjected to 10 mm of bilateral interdental mandibular lengthening. It was observed that mineralization began at the host bone margins at the end of the distraction period, followed by a progressive increase in bone surface area, with a concomitant decrease in fibrous tissue. The gingiva initially exhibited mild inflammatory and reactive changes during distraction and during the first few weeks of consolidation. The rate of bone formation gradually increased from the end of distraction to the fourth week of consolidation, at which time it remained constant until sometime before the eighth week, when it tapered off slightly as remodeling began. From the second through the eighth week of consolidation, regenerative changes and neohistogenesis were seen in the gingival tissues. The authors conclude that the gingiva responds favorably to increased length by regeneration rather than by degeneration. The authors' further state that though the results appear favorable evaluation in humans is required for further verification.

Kruse-Lösler B, Flören C, Stratmann U, Joos U and Meyer U[35] studied the histologic, histomorphometric and immunohistologic changes of the gingival tissues immediately following mandibular osteodistraction. The authors studies 48 rabbits which underwent mandibular osteodistraction using defined distraction protocols with physiologic, moderate and hyperphysiologic forces. The soft tissues overlying the distraction gap were harvested finally for histologic, immunohistologic and histomorphometric investigations. The authors observed that the control group without distraction showed the typical architecture and thickness of normal gingiva. In groups with distracted mandibles, an accelerating atrophy of gingiva depending on the degree of mechanical loading was obvious, characterized by decreasing thickness of epithelial layer, loss of rete ridges and disorganization of the different cell layers with a high number of apoptotic cells. In lamina propria collagen fibres were reduced and elastic fibres increased. Histomorphometric analysis revealed significant correlation between degree of distraction and atrophy in overlying soft tissues. The authors conclude by stating that the rabbit model of mandibular lengthening shows an accelerating atrophy in the covering soft tissues following hyperphysiologic distraction and that the atrophic changes observed may be of temporary nature.

Kunimori K, Maruoka Y, Sato M, Harada K and Omura K[37] investigated distracted keratinized epithelium to elucidate any proliferative and degenerated changes and to estimate the stability of the gingival tissues in mandibular distraction osteogenesis in a rabbit model. Twenty-two rabbits were subjected to unilateral vertical osteotomy. After a latency period of 4 days, devices were activated 3, 6, and 10 days at a rate of 1 mm/day. The authors also investigated the recovery of the distracted gingiva in consolidation periods for 3 weeks. The animals were examined by histologic and immunohistologic methods using proliferating cell nuclear antigen (PCNA), single-stranded DNA (ssDNA), and keratin. Atrophy of distracted gingiva was observed characterized by loss of rete ridges, acanthosis, vacuolation in the prickle cell layer, and cleavage of the keratin layer. Proliferating cell nuclear antigen–positive cells and ssDNA-positive cells were observed in the basal and prickle layers, respectively. During consolidation periods, slight recovery of rete ridges, thinning of the keratin layer, and immature epithelial layer was observed. The authors concluded that proliferative and degenerative changes occurred to compensate for cell death and distracted space. Thickness of gingival tissues was maintained by high mitotic activity and delay in the rate of cell maturation. Immature epithelial layer exhibited weak resistance against various stimulating factors, such as cleavage of the keratin layer among distracted gingival tissues.

HISTOLOGY OF BONE DURING DISTRACTION OSTEOGENESIS:

Histology at the distraction site has been reported in literature [2, 4, 30, 38] and histological changes in the different phases of distraction osteogenesis have been well described by Dheeraj et al[19]. The histology at the site of distraction may be described as one of interrupted fracture healing. The tension is responsible for the increased angiogenesis and fibroblastic proliferation. It has been observed that the fibrous tissue of the soft callus at the fracture site align along the long axis. Subsequently osteoblasts are recruited to lay down the osteoid matrix. Bone formation occurs along the vector of tension and is maintained by the growing apexes, known as "growth zone." Following distraction, during the phase of consolidation three phases are observed:

a) **Central zone**: where the tissue is composed of mesenchyme-like and spindle shaped cells in which many capillaries are dispersed. So, it is called as "mesenchymal or proliferative area."

b) **Paracentral zones**: seen on both the sides of central zone in which number of cells and capillaries are decreased gradually accompanied with intercellular matrix mainly consisting of collagen fibres. So,it is called as "fibroblastic or collagenous area."

c) **Proximal distal zones:** are the areas in direct continuation with old bony edges. Woven bone trabeculae are observed and hence it is called as "trabecular or mineralizaion area." The tips of the trabecular area recruit pre-osteoblasts from the collagen ridge distracted tissue. These are arranged concentrically around the tips of trabeculae and have high proliferation index. Then these preosteoblast mature into osteoblast and contribute to trabecular growth. After completion of this period a homogenous zone is again observed and the newly formed woven bone is composed of irregular trabeculae with deeply staining haphazard resting and reversal lines. Following remodeling by osteoclastic resorption, the woven bone is systematically replaced my mature lamellar bone.

SCOPE AND PLAN OF WORK

SCOPE AND PLAN OF WORK

When we started anterior maxillary distraction for cleft lip and palate patient, there was no long term data available in the literature regarding the stability of this procedure. Hence, we decided to do the study and observe our results over several months and then analyze the results.

Surgeries were performed by the author and his team of assistant surgeons under general anesthesia. Patients were from Ragas Dental College and Hospital and from Rajan Dental Institute.

The analysis of the skeletal and dental land marks were done by orthodontist independently.

Speech was assisted pre-operative and 3 months post operative by a qualified speech pathologist.

Histology of the distracted cleft maxillary tissue, both soft and hard tissue, was analyzed under H & E sections. This was done by Oral and Maxillo-facial pathologists. The possibility of distracting fibrous scar tissue of the palate and alveolus was assessed.

MATERIAL AND METHODS

MATERIALS AND METHODS:

All the patients were from Rajan Dental Institute, Mylapore, Chennai and Ragas Trust Dental College, Chennai. All the patients or their parents were explained the surgical procedure and informed consent was obtained from them.

A. The study was carried out in three parts:

i. **To study the stability of anterior maxillary distraction in hypoplastic cleft maxilla**

   8 patients in a mixed dentition period and adult dentition period were operated on to correct the hypoplastic cleft maxilla. There were six males and two females. Few patients had pre-surgical fixed orthodontics (straight wire) and others had post surgical fixed Orthodontics. The purpose of pre-surgical orthodontics was to obtain sufficient space between the roots of the teeth in the intended osteotomy site. Post-surgical orthodontics was done to align and level the arch using the distracted bone also as part of the alveolar arch.

   Pre-surgical records such as radiographs and study model and photographs were obtained and stored. Post-surgical records were obtained six months and one year after completion of distraction. These were then assessed by two orthodontists independently to ascertain the extent of distraction and the skeletal and dental stability of the anterior segment of the maxilla following the procedure.

ii. **To assess the effect of the segmental distraction procedure on the patients' pre-operative speech:-**

   Speech can be affected in many individuals with surgically repaired cleft lip and palate. Very little is known about the effect of segmental distraction surgery on speech and velo-pharyngeal defect in cleft individuals. Pre-surgical speech assessment was done in 3 patients and analyzed by speech pathologists. Speech was again evaluated using identical methods three months after completion of distraction and removal of the hyrax appliance. Speech assessment was again done by speech pathologists and compared to the pre-surgical evaluation.

iii. **Histology of distracted tissue** :-

Distraction of scar tissue has not been described before. It was decided to biopsy the distracted soft tissue in the surgical site. Only patients requiring implant supported space closure were chosen for histological analysis of mucosa over distracted site. Punch biopsies were made under local anesthesia and fixed in 10% formalin. Samples were dehydrated in a graded series of ethanol and embedded in paraffin. Finally, 5-µm-thick serial sections were cut for histological evaluation. The sections were deparaffinized and rehydrated in a graded series of ethanol. For histological evaluation, the sections were stained with hematoxylin and eosin (H&E). This was to determine the nature of tissue formation in a scar area which was distracted.

B. Pre-operative preparation

Separators were placed between the 1st molars and 2nd premolars, 2-3 days before surgery. The next day patient was recalled for molar banding and banding of 1 or 2 teeth anterior to the distraction site. Upper impression with Rubber base material was taken with molar bands and they were transferred to the impression and cast was poured and the AMD appliance was fabricated.

C. **Surgical method:**

- All the surgical procedures were done under general anesthesia with naso-tracheal intubation.
- After surgically preparing and draping the patient, a throat pack was placed.
- The distraction appliance was tried on to check for accuracy of fit.
- 2% lignocaine with 1:200000 adrenaline was infiltrated along the buccal vestibule of the anterior maxilla and palate.
- A horizontal circum vestibular incision was made in the unattached alveolar mucosa about 5 mms apical to the teeth apices extending from the first maxillary molar region to the opposite side first maxillary molar region to expose the anterior surface of the maxilla and the piriform aperture.
- Mucoperiosteal flap was raised with a Howarth's periosteal elevator or molts no 9. The nasal mucoperiosteum was elevated of the bony floor and walls with care being taken to detach the septal mucoperichondrium from its attachments to the maxillary crest and anterior nasal spine.
- Crevicular incisions was placed in the maxillary premolars and molar areas bilaterally, this was done on both labial and palatal aspects and a mucoperiosteal flap was elevated along with interdental papilla and tunneled to expose the interdental bone at the osteotomy site.
- A horizontal osteotomy beginning at the piriform fossa 5mms above the maxillary anterior teeth extending till the planned distraction site was done and vertical interdental osteotomy cut extended caudal from the horizontal osteotomy.

- The interdental osteotomy was performed on both the labial and palatal surfaces and extended to meet the horizontal limb in the anterior maxilla. The position of the interdental osteotomy was determined by the root inclination of the teeth in the vicinity of the osteotomy and the number of teeth in the proximal and distal aspect of the osteotomy which will be used for anchorage of the tooth borne appliance. The interdental osteotomies which was performed using a 701 drill was completed with a thin spatula osteotome.
- The interdental osteotomy was connected to the horizontal limb completing the desired osteotomy.
- The same osteotomy procedure was done in the opposite side.
- The septal attachments to the maxillary crest were removed with a guarded septal chisel.
- The lateral nasal wall was osteotomised with a guarded lateral nasal chisel.
- Care was taken to ensure that the osteotomy was completed on the palatal alveolar and the palatal vault which may prevent complete mobilization and the segment from distracting forward. A finger was placed in the palatal vault while the osteotomy is being done with an osteotome to give us a tactile feedback on the depth and direction of the osteotome. This ensured the completion of the osteotomy without injuring the greater palatine artery pedicle for the viability of the osteotomised segment. The same osteotomy procedure was done on the opposite side.

- The anterior maxilla was down fractured with finger pressure and ensured that the segment was completely mobilized and free to finger pressure. The wound was closed with 3-0 vicryl.
- The AMD appliance was fixed in the upper arch and a band pusher was used to achieve anatomically snug fit. The distractor was trial activated to confirm the completion of osteotomy and demonstrate the movement. The device was removed, cleaned and dried. The upper arch was isolated with gauze pack and dental air syringe. The AMD appliance was cemented with zinc phosphate cement due to the better tolerance to moisture.
- A single suture placed at the interdental site to ensure good approximation of the interdental papilla. The patient was given IV antibiotics and analgesics on the first post-operative day and was continued on oral antibiotics for a period of five days.

D. **<u>Post-Operative procedures</u>**

Orthopantomograph and Lateral cephalometric x-ray were taken on 1st post-operative day. The latency period was for five days and activation was started on sixth post-operative day and was continued till the desired result was achieved.

During the activation period the AMD appliance was activated at a rate of 1mm and rhythm of 2 (twice daily). The AMD appliance was sealed with light cure composite at the end of the activation period. A consolidation period of six months was maintained

RESULTS

RESULTS

I. Skeletal and Dental Changes of Anterior Maxillary Distraction

Maxillary Retrognathism which is measured as SNA angle was determined pre and post operatively. All the patients had improvement of SNA which showed that the maxillae had moved anteriorly. The range varied from a maximum of 9° to a minimum of 2°.

The skeletal class 3 pattern improved (ANB angle) for all patients and this varied from a maximum of 10° in one patient to a minimum of 2°. All other patients had values close to 7°.

The anterior movement of the anterior nasal spine (HP-ANS) was seen to occur in all patients. This again ranged from a maximum of 7 mms to a minimum of 2 mms. The posterior movement of the posterior nasal spine was also observed. This could be explained by either the loss of anchorage in the posterior teeth or by the presence of severe scarring of the palatal tissues due to previous surgery. This movement ranged from the maximum of 3 mms in 3 patients, 2mms in 3 patients and 1 mm in 2 patients.

The upper central incisors showed proclination following distraction of the anterior segment. This varied from 6° to 17°. Most of these patients did not have prior fixed orthodontics. In severe proclination, the appliance also dislodged in the final stages of distraction.

The maxillary incisors showed superior movement ranging from 2 to 4 mms.

The maxillary molars also moved superiorly but the movement was lesser ranging from 1 mm upto the maximum of 3 mms.

The naso-labial soft tissue angle decreased in all patients. The variation is from 5° to a maximum of 19°. The decrease in naso- labial angle was accompanied by increased fullness in the lower paranasal areas imparting projection in the under developed cleft maxillae.

The upper lip length increased in all patients. This was probably due to the thinning of the lip with the anterior movement of the upper anterior teeth and alveolus. This ranged from 1 mm to a maximum of 3 mms.

Superior movement of point A: This closely follows movement of the anterior nasal spine. In six of these patients, the movement of point A superiorly matched that of anterior nasal spine. For two patients there was a discrepancy of 1 mm.

Lower facial height: This was measured from the anterior nasal spine of the menton. This increased in all patients with one patient showing the increase of 10 mms. Most of the increase in facial height ranged between 4mms & 7 mms.

II) Histological changes:-

Post distraction, most of the patients were started on fixed orthodontics to align the crowded maxillary teeth. Some patient who had had previous extraction were left with an edentulous space. We planned to rehabilitate such patients with dental implants and a fixed prosthesis as a two stage procedure. The first stage involved preparation of the Titanium Implant fixture site. During this period the overlying soft tissues was removed with a tissue punch for a flapless technique of implant placement. This tissue was sent for histology examination.

This area represented the area of new tissue regeneration caused by the activation of the distraction appliance. This area corresponded to the area of the osteotomy cuts and hence the soft tissue regenerate in this region had to be necessarily only tissue developed by distraction.

The soft tissue histology section showed in microscopic examination of the hematoxylin and eosin stained soft tissue section under low power magnification revealed orthokeratinized stratified squamous surface epithelium with long rete pegs extending into the underlying mucosa. The basal layer of the epithelium showed the layer composed of closely packed cuboidal cells with intensely hematoxyphilic round nuclei. Interspersed within this layer were some clear cells. The connective tissue is composed of collagen fibers, cells and thin walled blood vessels – capillaries. The collagen fibre bundles are arranged regular, parallel wavy pattern adjacent to the rete pegs (papillary layer) and the reticular layer showed capillaries, spindle shaped fibroblasts and numerous collagen bundles interlacing with each other, but predominantly oriented parallel to the vertical axis. Some sections exhibited increased chronic inflammatory cell infiltrate composed of lymphocytes and plasma cells. The histological features were similar to those exhibited by normal gingival.

Some of these patients had heavily scarred palates due to prior surgery. Inspite of heavy scarring, the palatal soft tissue and gingival tissue showed significant distraction.

III. Speech in relation to Anterior Maxillary Distraction:-

Velo-Pharyngeal dysfunction and speech defects are recognized in operated cleft lip and palate patients.

The pre and post surgery speech of 03 patients who had undergone anterior maxillary distraction were collected. The recording protocol adopted was uniform for all the subjects. All the speech samples were recorded by a speech language pathologist in a sound treated room using a voice recorder. The participants were instructed to perform the speech tasks including repetition of words, sentences, number counting (from 1-10) and 60-70, in English) and general conversation. The sentence repetition task included repetition of ten phonetically loaded sentences in Tamil language.

The speech samples collected were perceptually rated independently by three speech language pathologists for articulation, resonance, speech understandability and speech acceptability. All the listeners had a minimum of three years experience in the assessment of speech in individuals with cleft lip and palate. The ratings were carried out based on the universal parameters for reporting speech outcomes in individuals with cleft palate. The listeners were blindfolded regarding details of the subjects to eliminate factors biasing the analysis. All the speech samples were randomized before presenting to the listeners. The speech samples were presented to all the 3 listeners at the same point of time.

The analysis of speech samples rated by 3 listeners was compared. It was observed that in all the three patients there was no difference in the rating of any parameter of speech pre and post surgery. The small sample size of the subjects in this study makes it difficult to draw any conclusions in reporting the outcomes of this particular procedure.

However, no deterioration of speech was noticed in any patient.

DISCUSSION

DISCUSSION

Superior movement of anterior nasal spine of the patients had a vector in a forward and cephalic direction thereby increasing the existing open bite or reducing the existing overbite. This movement varies between 5 mm and 2 mm. The reason for this could be the rigidity of the hyrax appliance. Since the anterior maxilla is completely sectioned from all its bony attachments and also from the cartilaginous nasal septum, there was no counter acting or restricting force for the vertical movement of the anterior segment.

1) **Skeletal and Dental changes in anterior maxillary distraction using Hyrax appliance:**

Skeletal hypoplasia especially in the antero posterior plane is common in cleft lip and palate patients who have had surgery earlier. This hypoplasia can be corrected by one of the following techniques.

a) Maxillary Osteotomy and advancement surgery.

b) Maxillary distraction (Full jaw with Le Fort Osteotomy and using external or internal distractors.

c) Anterior segmental distraction (Osteotomy in one or two stages and then using extra oral or intra oral distractors).

Most of the distraction was done by bone borne distractors. The use of a hyrax appliance as a tooth borne distractor has only been described recently but not much data has been reported. Our patients have all had anterior maxillary osteotomies followed by a latency period and distraction using the hyrax tooth borne distraction device. The anterior maxillary segment including anterior floor of the nose and the teeth in the anterior segment were all moved over several mms anteriorly by distraction. The anterior maxillary skeletal base moved forward with anterior nasal spine with point A moving forward as much as 7 mms. There was anchorage loss with certain amount of posterior movement of the posterior nasal spine and also the first molars. The effective increase in the palatal length was as high as 9 mms in one patient with an average increase of 6.5mm length. The skeletal movement was accompanied by forward positioning of the upper lip and also increased fullness of the para-nasal area. The reduction of naso-labial angle further enhanced facial aesthetics. This movement has been stable for more than 6 months follow-up in all the patients. Distraction of palatal soft tissue and alveolar soft tissue and buccal mucosal soft tissue were all documented.

Good volume of bone regenerate was noticed in the entire alveolar region with a slight concave pattern) near the maxillary sinus floor region (reminiscent of the hour glass appearance of bone regenerate).

Tooth borne distraction has several advantages compared to bone anchored distractors. Those include:

a) Ease of application
b) Less morbidity such as cheek & lip ulcers seen with bone anchored appliances.
c) Removal of appliance at the end of consolidation period is very simple and can be done without anesthesia
d) Economical
e) Less chances of infection since there are no fixation screws.

The procedure however does have a few disadvantages such as follows:

a) Vector control unidirectional only
b) Need for more diligent oral hygiene maintenance
c) Activation at home being difficult

The ease of anchoring the appliance and the ease of adaptation to the appliance by the patient especially during the consolidation phase is significant.

Stability of the procedure is also a great advantage. Distraction in cleft lip and palate has always had a certain degree of surgical relapse within the first 1 year. This relapse can be as high as 50% of the movement achieved. Maximum relapse happens within the first six months of surgery.[55] In Thongdee's study, total relapse of surgical movement was 31% in the horizontal plane and 52% in vertical plane as well as 30% in rotational.

In our series of patients relapse has been shown to be minimal. The follow-up period has ranged from 12 months to 36 months. The stability of the procedure is hence a great advantage in this technique.

The bone regenerate in the alveolar region is used to either decrowd the maxillary teeth with fixed orthodontics or used to anchor Titanium implants to support a fixed dental prosthesis. The consolidation period is atleast 3 months during which bone maturation occurs.

The paranasal fullness is contributed by the advancement of the piriform rim.

The procedure is not without **complications.** These include:

i) **Root Damage** :

Damage to the roots of teeth close to the interdental osteotomy site. This is more common when a bur or saw is used for the Interdental osteotomy through & through. It is also more common when the roots of the teeth are not divergent or if inter radicular space has not been created by pre-surgical orthodontics. The damaged tooth may remain asymptomatic or could become non-vital and require treatment such as root canal and crown

ii) **Anterior Open Bite:**

All of the patients developed a forward and slightly cephalic direction of anterior segment movement. This sometimes creates an anterior open bite situation.

iii) **Bur breakage**:

One of our patients had a No.701 drill bit breakage at the osteotomy site which was left in situ at the time of surgery. This was later removed under local anaesthesia after distracting the anterior segment which created space at the osteotomy site.

iv) **Appliance fracture:**

There were episodes of appliance fracture, especially at the soldered joint between the band and hyrax distractor in 2 of our patients. In such patients, the appliance was removed and alginate impression taken immediately for a self cure acrylic space maintainer.

This was placed with ligature wires until the new distractor was fabricated. The procedure of Band adaptation and distractor soldering was done in the similar pre-surgical fashion but in the new teeth position. The distractor was then placed back after removing the acrylic space maintainer and distraction routine followed as usual.

v) **Appliance dislodgement**:-

The appliance was dislodged from one of the teeth in 2 patients who reported to us immediately. The distractor was removed completely, cleaned and cemented back in the same position. The distraction routine was then followed as before.

2. Histology of Distracted tissue:

Distraction is a well-known phenomenon to expand bone. Invariably, soft tissue, vascular channels and nerves also can be distracted without loss of integrity and function. This distraction histogenesis can also extend to scar tissue as evidenced by distraction of the palatal scar tissue in these patients. Histology in the distraction site revealed connective tissue interspersed with fibroblast and new vascular channels. The bone quality was normal and in one instance could also take the load of an osseo-integrated dental implant with a ceramic crown. The findings of Kruse-Losler et al[35] and Kunimori et al[37] were those observed, respectively, during and immediately after the procedure when the overlying mucosa was subjected to the tension of distraction. Whereas Cope et al[11] stated favourable gingival response following the procedure; the findings did not report the presence of mature, normal gingiva. In the current instance, the mucosa of the gingival – epithelium and connective tissue, gave the appearance of mature normal structures. It could probably imply that the process of distraction does not have permanent long lasting effect on the mucosa and any changes that occur during the process are reversible.

3. Speech before and after anterior maxillary distraction:

Speech can be affected and Velo-pharyngeal deficiency is commonly seen in operated cleft lip and palate patients. When such patients have border line VPD, the possibility of them getting worse with a full maxillary i.e. Le Fort 1 maxillary distraction (which essentially mobilizes the maxilla anteriorly) is very high. When distraction is done as a segmental procedure we have shown that there is no deterioration in speech.

Furthermore, loss of anchorage moves the posterior nasal spine and the posterior maxillary teeth further towards the oro-pharynx. One patient was assessed as having speech improvement. This is an important advantage of anterior maxillary distraction. However, the entire VPD has not been studied and could be the basis of future work.

4. Stability of Skeletal Movement:

Increase in the palatal length ANS to PNS was monitored from the completion of consolidation period for a further minimum period of 6 months. The relapse varied between 0.5 to 1 mm absolute value with the % relapse varying from 10% to 25%. Literature shows that most of the skeletal relapse happens in the first six months period.[55] The anterior movement of the nasal spine again relapsed between 1 & 3 mms, the percentage change being between 21 & 33%. This shows that there is a certain degree of skeletal relapse and we need to build in some over correction in such distraction techniques. The posterior movement of the posterior nasal spine showed an average 30% relapse. (Range from 0% to 66%).

The superior movement of the anterior nasal spine showed a correction relapse between 25% and 50%. The other important change which is the superior movement of point A relapsed again between 25% and 50%. This can however, be controlled by some form of callus moulding technique one week after completion of distraction. The proclination of upper incisor reduced between 23% and 44%. This can also be altered to a more stable position by use of post distraction orthodontics. The soft tissue stability in terms of nasolabial angle also showed change in the immediate post consolidation period. The decrease in nasolabial angle relapsed about 30%, 6 months after consolidation. The detailed values of the relapse and the percentages are in the table given. (Appendix III).

The upper lip length showed a mild increase between 1 mm & 3.5 mms. This however, relapsed between 0 to 33%.

Stability of the segment which has been distracted adds to a long term success of the procedure in the form of function of aesthetics.

SUMMARY AND CONCLUSION

Summary & Conclusion

This innovative technique of distraction of a scarred, operated cleft palate by means of segmental distraction has shown to be more stable than the classical methods of Le-Fort 1 distraction.

The technique has no deleterious effects on speech when analyzed by speech language pathologists. This is a significant factor while considering surgical options in patients with pre-existing speech disorder or velo-pharyngeal dysfunction (VPD).

Histology shows that there is a hyperplasia of soft tissue in the region of the regenerate and that distraction is possible even in scar tissue.

The distracted regenerate bone was of adequate volume and quality to be able to have orthodontic realignment or support an osseo-integrated implant.

The complications seen were minor and easily rectifiable.

Patient adaptation and acceptance was good. However, patient's attenders have to be alert enough to activate the appliance correctly and note any complication early enough.

RECOMMENDATIONS

RECOMMENDATIONS

The technique should be reproducible in several centers to get a larger sample size and more extensive data collection and interpretation. The technique itself looks promising and also cost effective and hence should be taken up by centers which have limited resources. A larger sample size will also help better speech assessment. Though the findings of normal mucosa were confirmed by routine histology, further studies, probably ultrastructural, are needed to verify the 'normalcy' of the cells and tissues. This technique should also be tried in late mixed dentition period where orthognathic surgery cannot be performed. Appliance designed can be further enhanced with easier methods of activation such as self activating devices or miniature motorized devices.

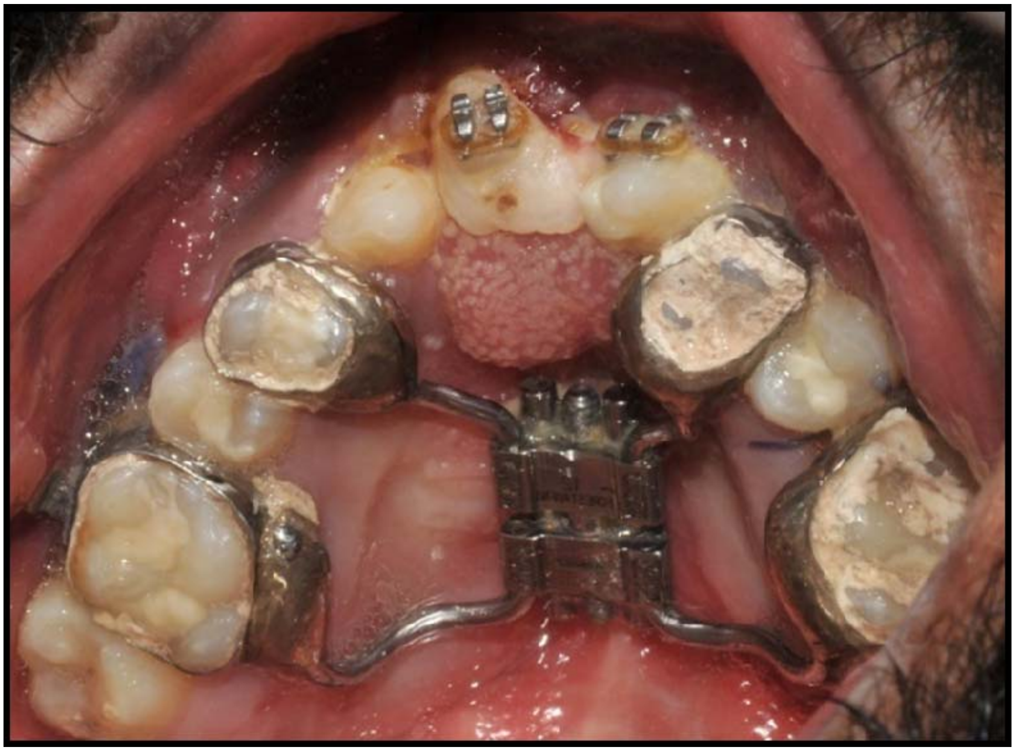
APPENDIX

SURGICAL STEPS

Anterior maxillary distraction cuts

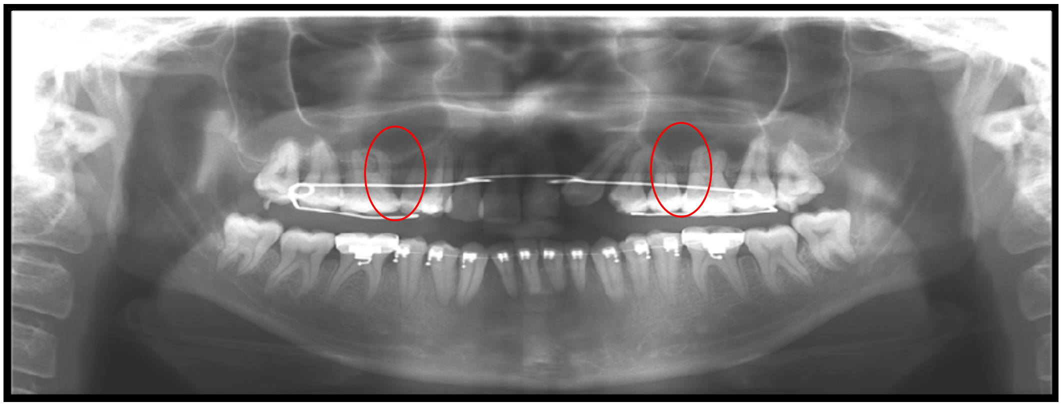
Distractor cemented with ZnPO4 cement

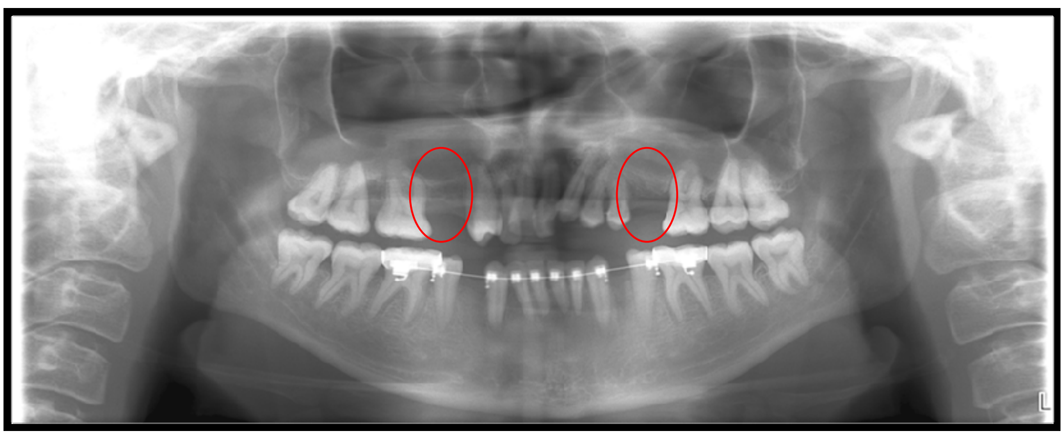
PRE & POST DISTRACTION: INTRA-ORAL



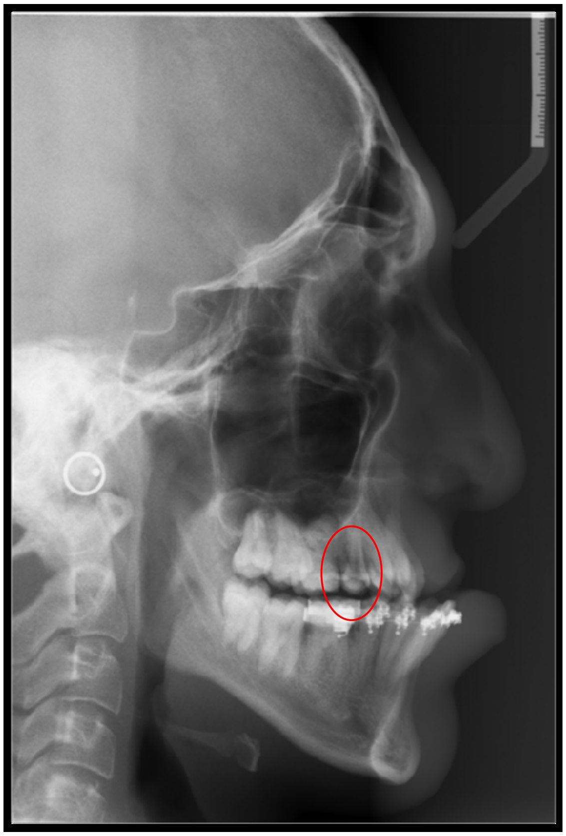
PRE & POST DISTRACTION: INTRA-ORAL

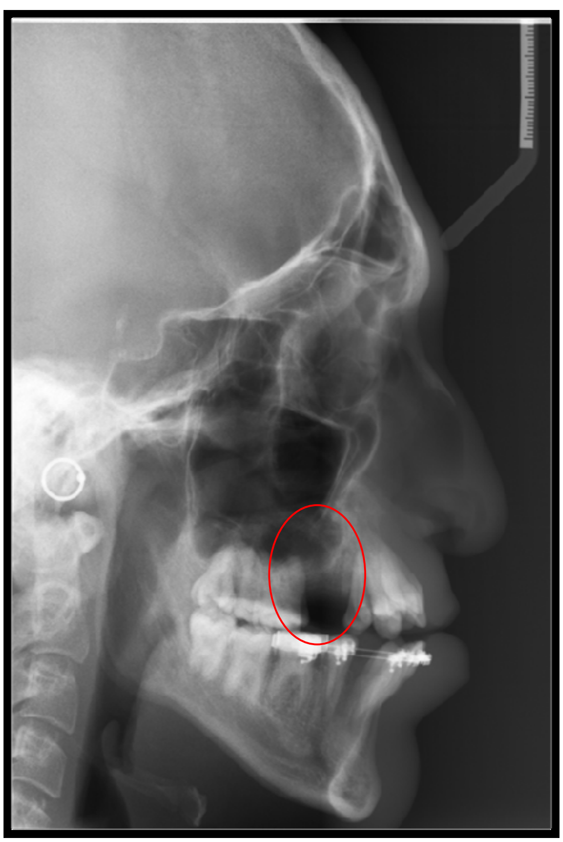
PRE & POST DISTRACTION: OPG



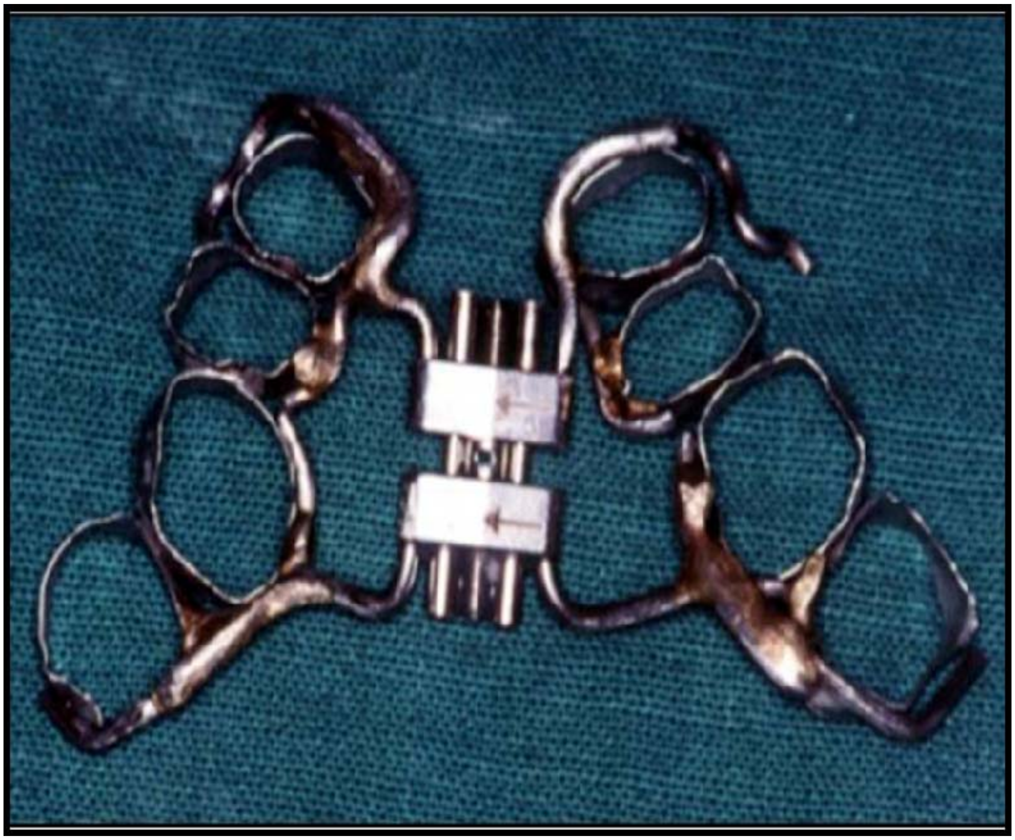


PRE & POST DISTRACTION:Lateral Cephalogram

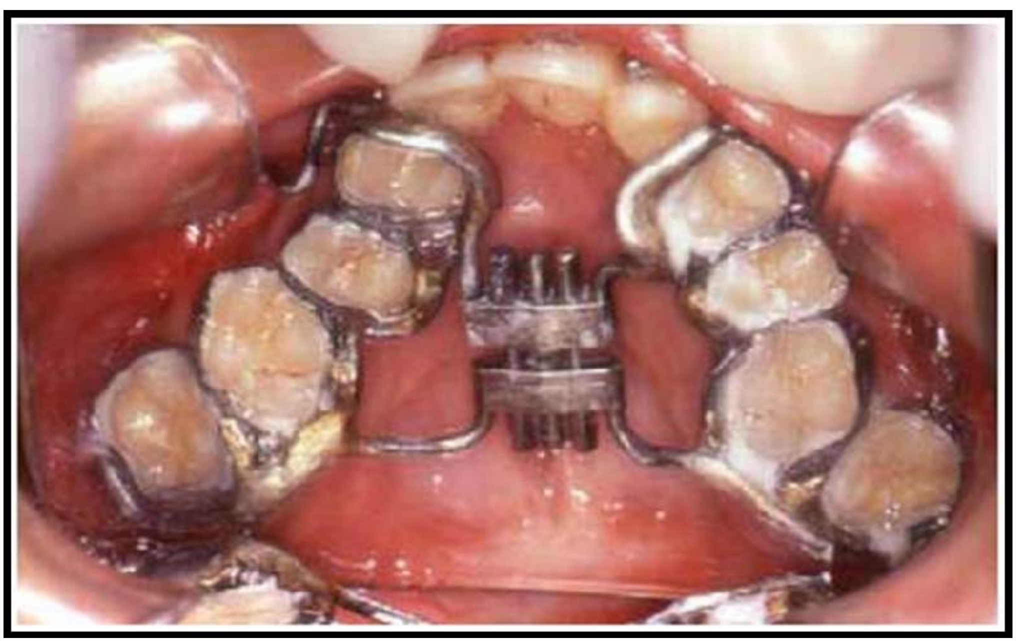




TYPES OF APPLIANCES



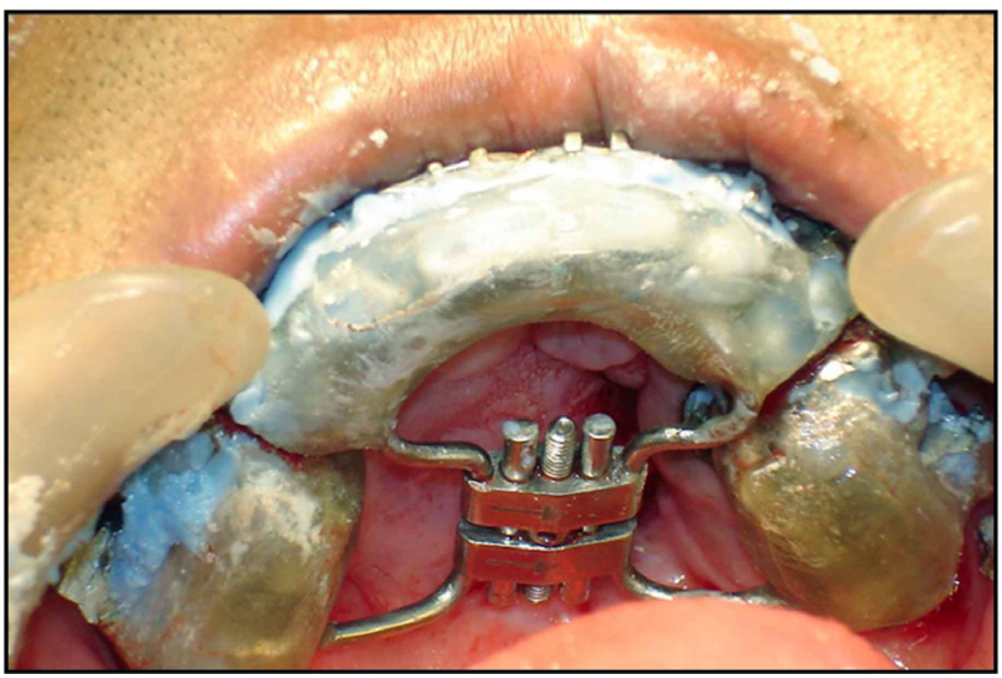
Hyrax Distractor



Hyrax distractor placed intra-orally

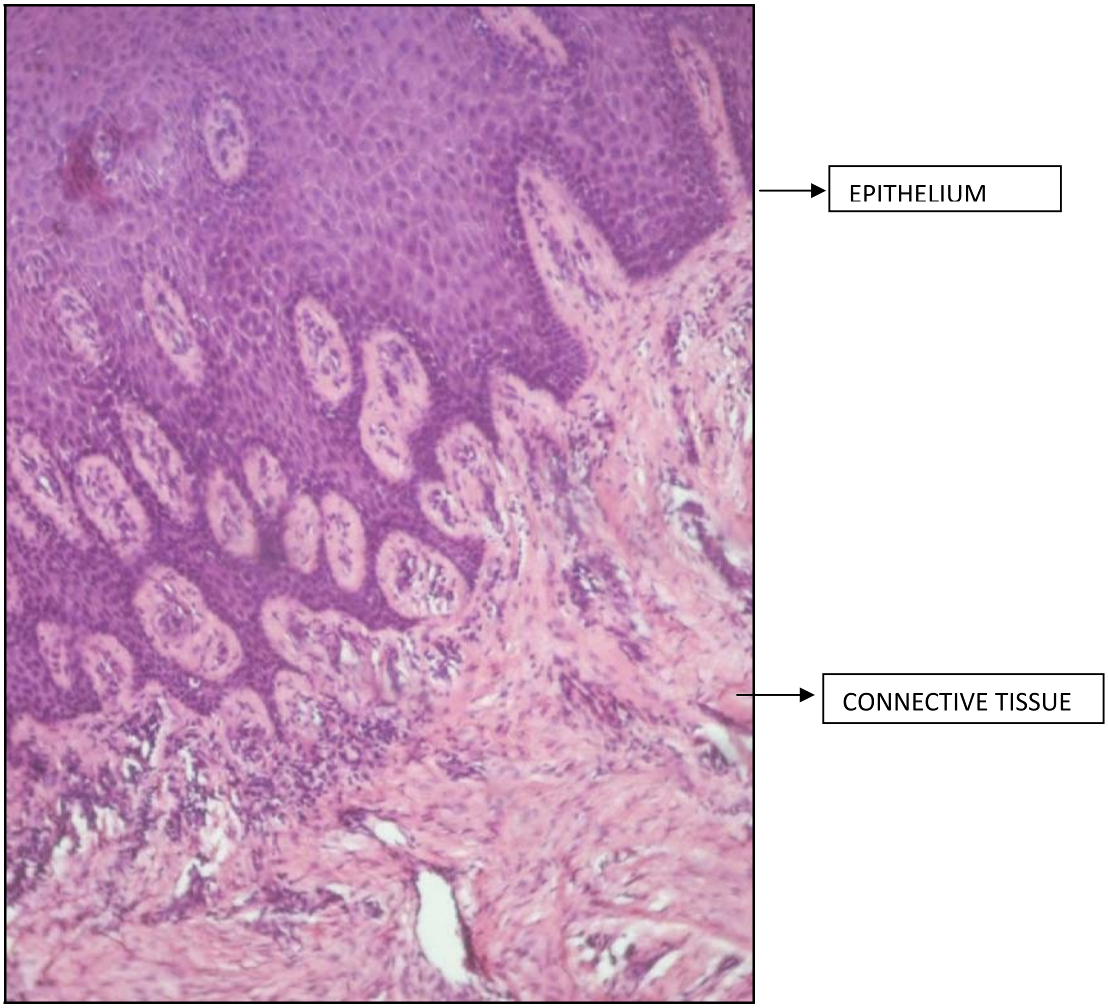
TYPES OF APPLIANCES

Hyrax Distractor with acrylic reinforcement

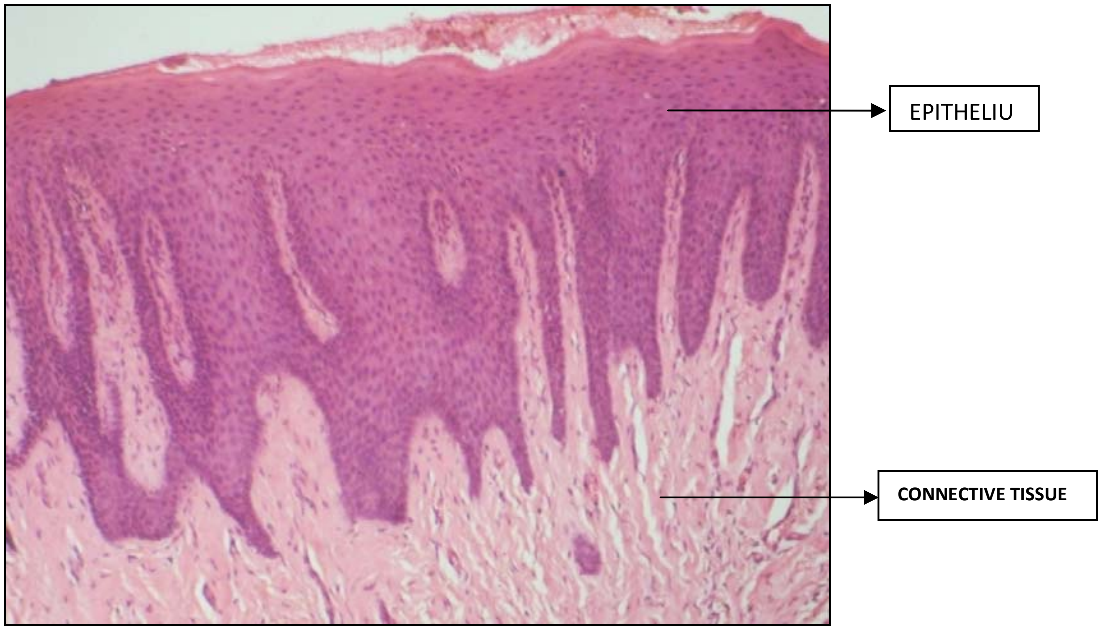


Hyrax Distractor with acrylic reinforcement placed intra-orally

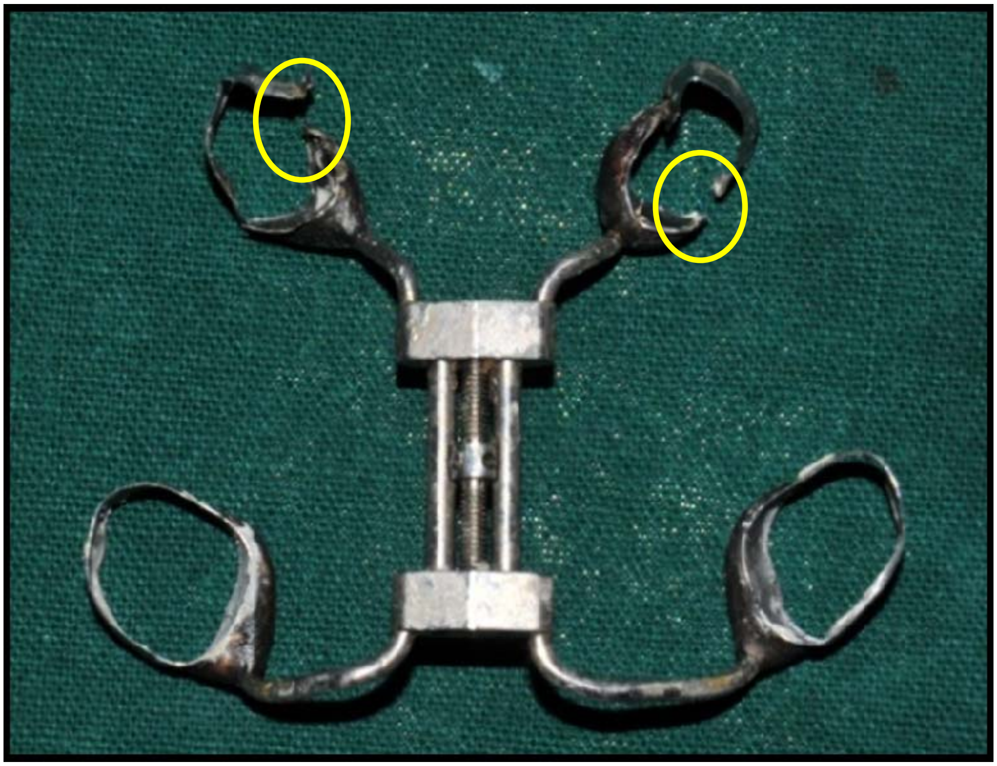
HISTOLOGY OF DISTRACTED REGION TISSUE



HISTOLOGY OF DISTRACTED REGION TISSUE

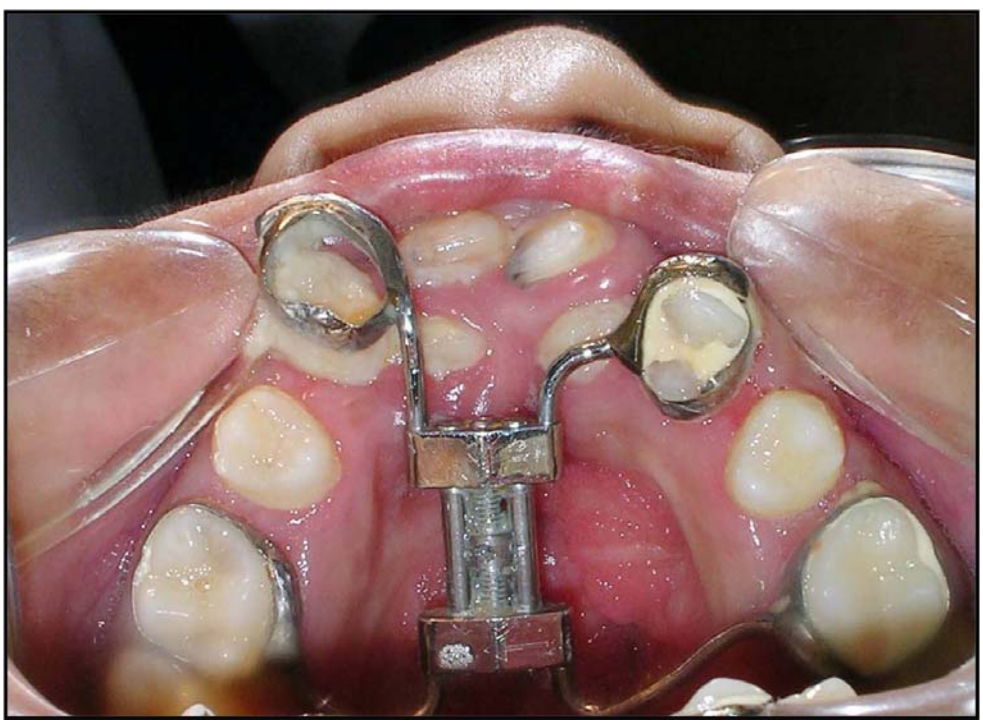


COMPLICATIONS - Broken Appliance

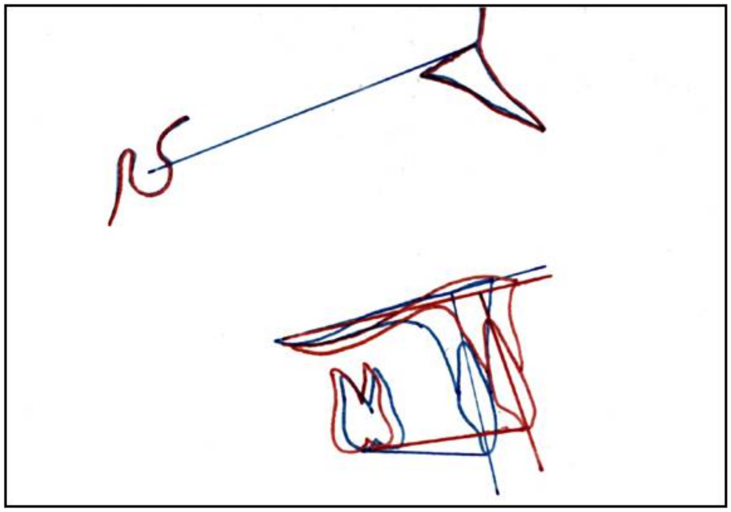


Space maintained with self-cure acrylic space maintainer
till the new distracter was fabricated

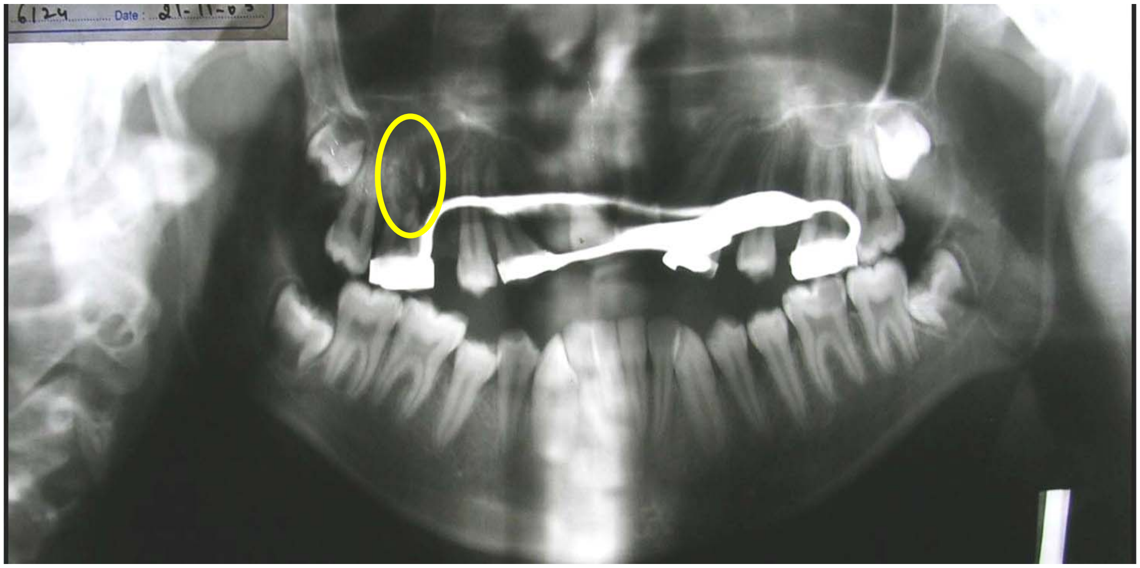
COMPLICATIONS - Dislodgement of appliance



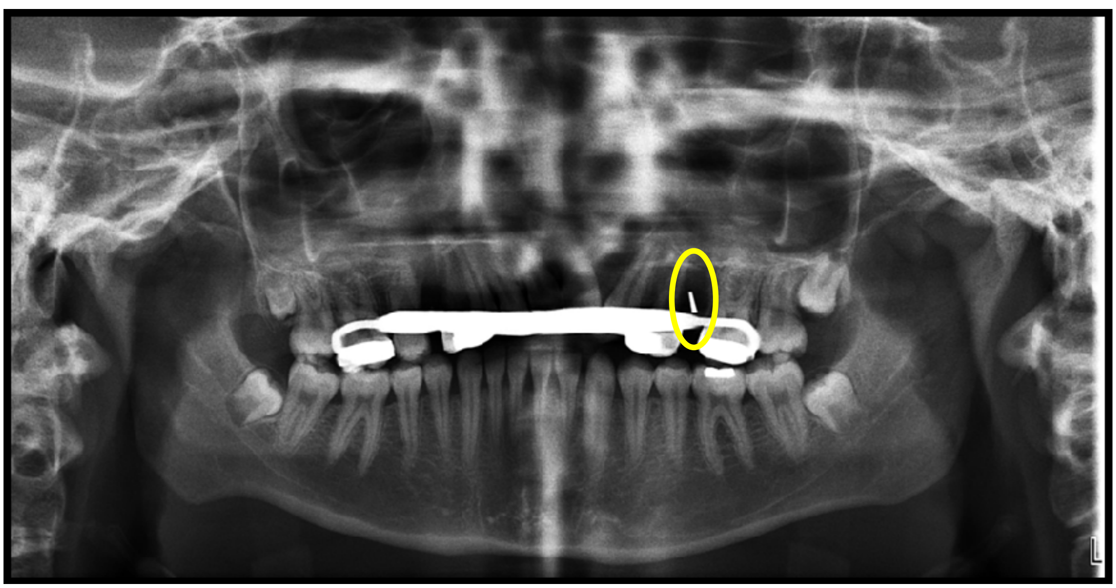
COMPLICATIONS – Anterior Open Bite



<u>**COMPLICATIONS**</u>

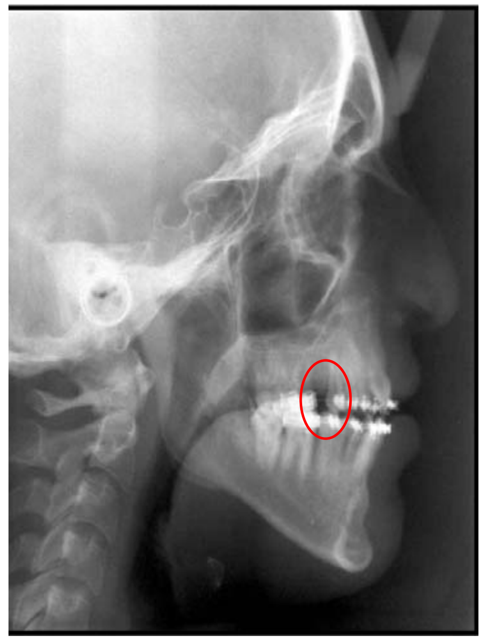


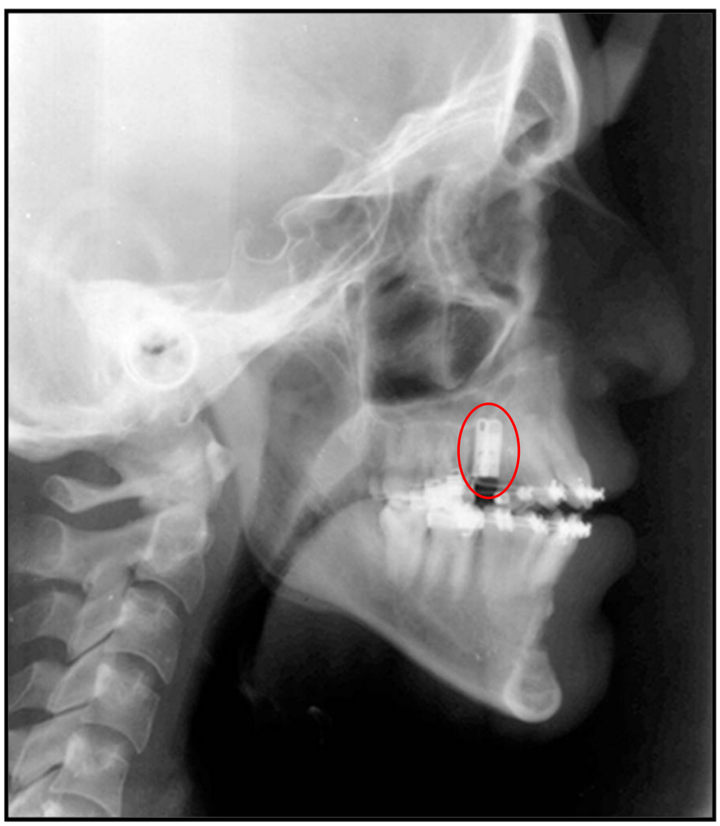
Root Damage



Bur breakage during surgery

DISTRACTION & IMPLANT PLACEMENT IN DISTRACTED REGION





Case No. 5 - Post Activation
Lateral Cephalogram

Case No. 5 - Pre Operative
Lateral Cephalogram

Case No. 4 - Post Activation
Lateral Cephalogram

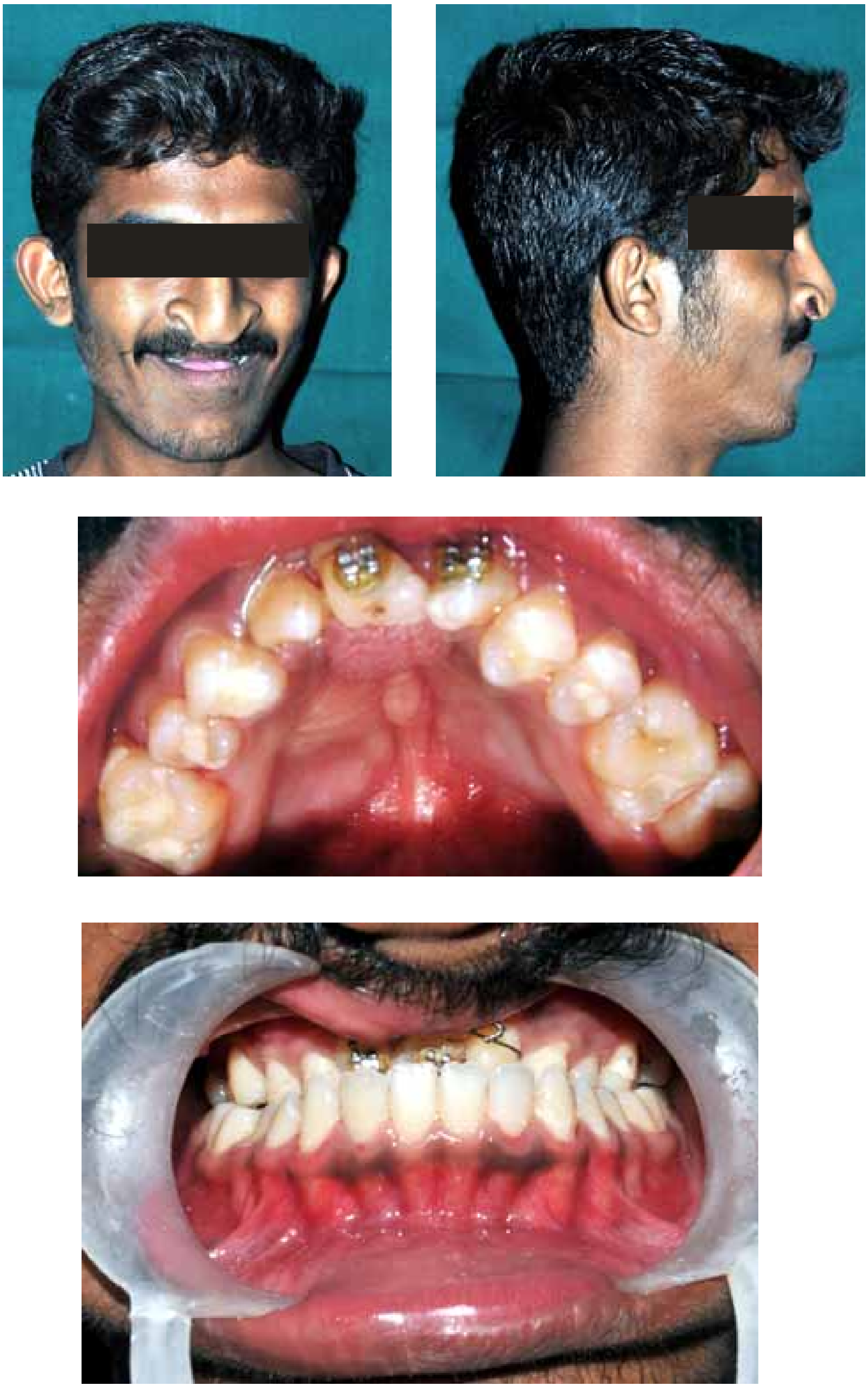
Case No. 4 - Pre Operative
Lateral Cephalogram

Case No. 1 - Pre Operative
Lateral Cephalogram

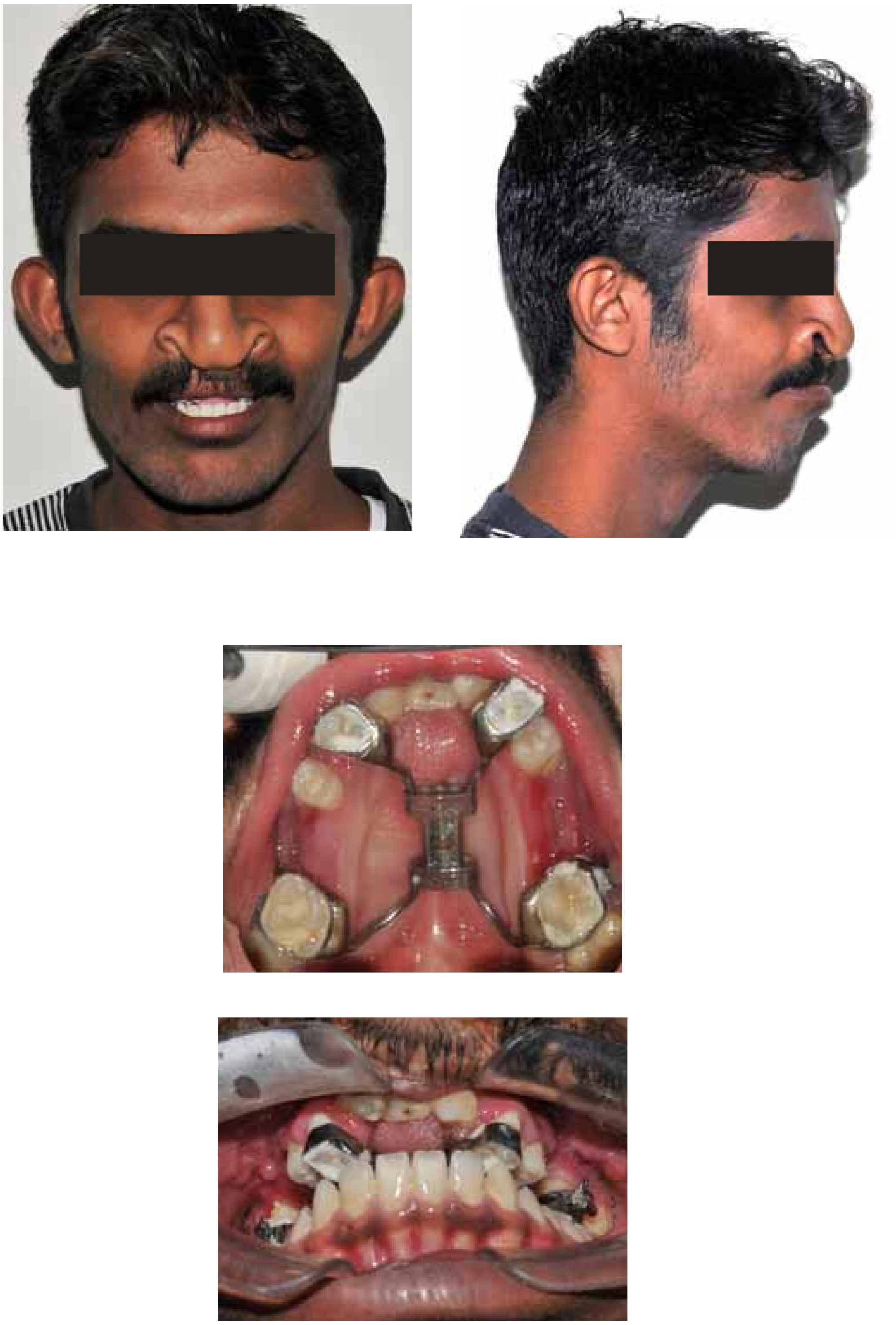
Case No. 1 - Post Activation
Lateral Cephalogram

Case No. 5 : Pre - Operative

Case No. 5 : Post - Activation



Case No.4 : Pre-Operative



Case No.4 : Post - Activation

Case No. 1 : Pre-Operative

Case No. 1 : Post-Activation
and Post Orthodontic Treatment

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