Case Report

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Intricate Assessment of Facial Growth & Development in A Patient with Cleft Lip & Palate after Surgical Correction: A Clinical Report with Historical Perspectives

Sunil Pandey[1*] Brinda Suhas Godhi[2] Pragya Tripathi[3]

[1]Reader, Department of Pedodontics & Preventive Dentistry, Government Dental College, Raipur, Chhattisgarh, India.
[2]Reader, Department of Pedodontics & Preventive Dentistry, JSS Dental College & Hospital, Mysore, Karnataka, India.
[3]Reader, Department of Periodontology and Oral Implantology, Inderprastha Dental College & Hospital, Ghaziabad, India.

ABSTRACT

Background: Clefts of lip and palate usually vary in their clinical presentations from mild to severe, and may be related with deficient craniofacial development. Still the most practiced and refined methods of surgical repair of these defects are followed by scar contraction and fibrosis with subsequent skeletal defects, dental abnormalities, cosmetic disfigurement, and speech impairment. In recent times, enhancement of innovative visual technology has enhanced the development of operational techniques for feto-endoscopic surgery, which has been showed to be less invasive than the open approach. However in the future, such surgical intervention may become possible as fetal surgery becomes safer for the mother and foetus resulting in a major breakthrough in the field of cleft and craniofacial anomalies. Fetal cleft repair is a striking intervention for plastic surgeons as it affords the potential to provide a spotless repair that more closely approximates natural existing conditions. Till date, perhaps there are very few clinical studies have been conducted to evaluate the efficacy of facial growth and development following these repairs. Herein authors have genuinely explored this highly obscured field of craniofacial surgery.

Keywords: Foetal Surgery, Cleft lip and Palate, Facial Growth, Delaire Regime.

INTRODUCTION

Human face is a representation of healthy development & a mirror of personality of an individual. The perfect development leads to a symmetrical face; a beautiful projection in viewer's eyes. Deformities can lead to face far from esthetic composition & also undermine the personality of an individual. It can have functional esthetic & psychological implications. Cleft lip & palate is one such abnormality which has varied implication for an individual.[1] The development of face starts taking place from 4th to 9th week.[2] During this period the size of embryo increases from 3.5 to 30mm. The embryo is recognizable as human by this time. This marks the most important phase in human development because abnormalities occur most commonly during this phase of development. Cleft lip & cleft palate also develop during this period. The surgical techniques are available for correction of these anomalies & it is a team work which determines the outcome of correct measures. In case the patient does not receive care from the team work -the outcome may be unexpected.[3] The advancement in various diagnostic procedures determines the abnormalities or syndromes in the intrauterine life & if appropriate interventions are carried out during intrauterine life it may have profound positive effect on development of face. The growth pattern of surgically treated case of cleft lip & palate, their effect on maxillae, mandible, speech & psychology of child & other functions will

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*Correspondence Dr. Sunil Pandey.
Department of Pedodontics & Preventive Dentistry, Government Dental College, Raipur, Chhattisgarh, India.
Email: drsunilpandey@gmail.com

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be discussed.[4] Further we shall determine them to provide best possible management (modalities) of this patient in future. This article is an attempt to highlight the basic ideology, concepts with historical aspects of facial growth & development in a patient with cleft lip & palate after surgical correction.

Embryology

The primary palate arises from the fusion of two medial nasal prominences & medial growth of maxillary prominence that forms the intermaxillary segment, which develops towards the end of 5th week of intra-uterine life. It consists of two portions; a labial component (that forms the philtrum of the upper lip) and a triangular palatal component of bone (that includes the 4 maxillary incisor teeth). Palatal development begins towards the end of the 5th week of intra-uterine life & is complete at about 12 weeks. The critical period is from the end of the 6th week to the beginning of the 9th week. In normal palatal development, mesenchymal cells from the neural crest migrate to the primitive oral cavity forming the maxillary process in association with the craniopharyngeal ectoderm.[5] Development of intact secondary palate is a dynamic process which, consist of three stages. Stage-1 characterized by formation of the palatal shelves from maxillary process. These shelves are oriented vertically, either side of developing tongue. In Stage-2 these vertical palatal shelves elevate to a horizontal position above the dorsum of the tongue. This event occurs rapidly, possibly in a matter of hours. Stage-3 of secondary palate development involves fusion of the medial edge epithelium of the approximating palatal shelves with each other via numerous desmosome contacts to form a midline palatal seam. Some workers offered the possibility that the medial edge epithelium cells migrate into the body of the mesenchyme & transform into mesenchymal cells.[6] This process is known as epithelial-mesenchyme transformation (EMT). In vitro organ culture studies have suggested that TGF-β1 (transforming growth factor-β) & TGF-β2 accelerate the palatal shelf fusion process. However it has been shown that neutralizing antibodies to TGF-β1 and TGF-β2 appear to have no effect on this fusion process. On the other hand, antibodies to TGF- β3 block palatal shelf fusion. On completion of stage III, the epithelia on the nasal aspect of the palate are pseudostratified ciliated columnar cells while those on the oral aspect of the palate are stratified squamous epithelial cells.[7]

Factors relating poor growth of maxilla

The growth of maxilla of cleft patient is influenced by several factors which are quite independent of surgical treatment. One is the variable primary deficiency of tissue adjacent to the cleft another is the growth potential of maxilla, which is deficient in some syndromic cleft. Studies of adults with unoperated clefts show that most have the potential for normal facial growth. At birth there is no maxillary antrum, no tuberosity & the pterygoid plates are extremely small.[8] Facial growth occurs essentially by two related processes; Displacement and Remodelling. Displacement involves movement of whole bones or bony complexes away from one another at their junction. These junctions may be movable joints or suture or synchondrosis. Osteogenic activity results in remodelling of a bone by deposition or resorptive process.[9]

Abnormal facial growth in cleft lip & cleft palate

The causes of mid-face retrusion & poor facial growth in repaired cleft lip & palate subjects are poorly understood & the cause is often, debatable. Lip surgery, palatal surgery, early surgery to the hard palate & repeated surgery, the competence of the surgeon, as well as the timing & nature of surgery have all been separately implicated. At other extreme, some authorities still claim that any growth disturbance is purely intrinsic & that surgery does no harm. Maxillary retrusion, evident in a proportion of operated cleft lip & palate subjects in teenage years, is a well recognized phenomenon. Such retrusion does not happen in all cases or to the same extent in every case.[10]

Adult un-operated cleft lip & palate studies

Subject with un-operated cleft lip & palate have the potential for normal facial growth. Indeed because the lip is not intact, maxillary growth is unrestrained and exuberant. Lip surgery in infancy, without palatal surgery, results in near normal facial growth with some retroclination of the upper incisor teeth. It is clear that when palatal surgery is undertaken the downward & forward movement of

the maxilla & it's subsequent propulsion from the cranial base is compromised & this is why many such patients demonstrate concave profiles.[11] This disruption to the trans-location of the maxilla from the cranial base is thought to be caused by band of scar tissue created around the circum-maxillary suture at palate repair. There is evidence that minimal invasive surgery produces fewer disturbances to facial growth than historical technique which are now obsolescent. If there is greater deficiency of tissue the resulting narrowing of cleft will produce an abnormal medial displacement of the lateral element of maxilla & "alveolar collapse" without the operation interfering with growth in any way. As a result there is no surgical regime that can guarantee to prevent some degree of collapse, but it's effects can be overcome by orthodontic treatment.[12]

CASE STUDY

A 10 year old male patient reported to the Department of Pedodontics & Preventive Dentistry with a chief complaint of pain & swelling on right side of face of one day duration. As the patient has some other associated problems due to inappropriate follow up after surgical intervention. As patient is poor it is much more important to assist him for social reasons. Peri-orbital cellulitis is on the right side of face is of one day duration patient does not give any further history of present complaint. Surgery for cleft lip was done at the age of one year & for palate at the age of 4 year. Family history revealed that no other member of family has ever been affected with cleft lip & palate in known generation. Clinical findings reveal normal height & weight with normal growth and facial asymmetry (Figure 1-3). His lower eye lid was raised partly due to infra-orbital swelling present on right side of face. Other findings included; concave facial profile, broad nasal bridge, compressed left Nares, scar on left side of philtrum of upper lip, Class III malocclusion, right side maxillary sinusitis, scar on left side palate, oro-nasal fistula with cellulitis. Orthopantomogram (OPG) and CT scan investigations done for more accurate structural assessments (Figure 4-6).

Fig 1; Frontal view of patient after surgical reconstruction.

Fig 2; Intraoral view of patient after surgical reconstruction.

Fig 3: Occlusal view of patient showing dentition status.

Fig 4; OPG confirming developing dentition with anomalies.

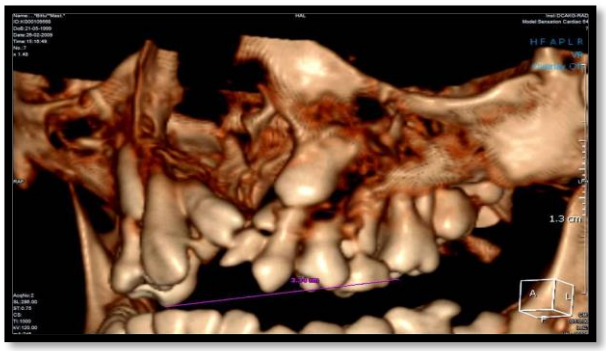

Fig 5; CT scan (Fronto-lateral view)

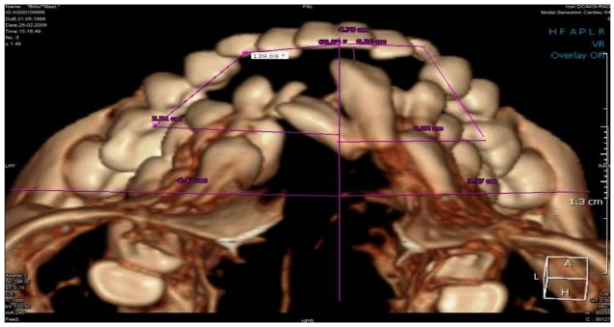

Fig 6; CT scan (Occlusal view).

DISCUSSION

Management of patient with cleft lip & cleft palate is best provided by an interdisciplinary team of specialists. It is responsibility of each team to be sensitive to linguistic, cultural, ethnic, psychosocial, economic & physical factor that affects the dynamic relationship between the team, parents & family. It is responsibility of team to monitor both short-term & long-term outcomes. Evaluation of treatment outcomes must take into account the satisfaction & psychological well-being of the patient, as well as on growth, function, & appearance. As members of the interdisciplinary team of physician, dentist, speech pathologist & other allied health professionals, pediatric dentist should provide dental services in close cooperation with their orthodontist, oral surgeon & prosthodontic colleagues. Regarding the choice of a treatment protocol, there are two basic areas of disagreement;[13]

(1) The timing of surgery &

(2) The operative technique.

Timing of surgery

Surgery in the neonate is more risky than in older babies; in particular, the cardiovascular & respiratory systems are immature & cleft can be associated with other congenital abnormalities, especially cardiac ones. It is therefore essential, if neonatal surgery is to be considered, that it is carried out in a centre with expert pediatric anesthesia & intensive care facilities and that a thorough preoperative assessment is performed by a Pediatrician. Majority of surgeons prefer to wait until the child is older- usually between 3-6 months before operating as, with reduced risk of anesthesia, they feel that they can spend more time & get a better result when the lip & nose are more developed. Many surgeon obtained soft tissue closure of the alveolar & anterior palatal cleft at the same time as they close the lip. There is no doubt that this area is easier to close at this stage when the cleft is wide open & access may be very difficult after lip closure. Never the less some leave this area untouched at primary operation as they believe such surgery interferes with growth; they close it at the time of alveolar bone grafting in later childhood. It has become clear that the child has a better chance of speaking normally if the cleft is closed early. Most surgeons, therefore, aim to close the whole palate by 18 month & many do so at six months. Unfortunately, surgery to bony cleft of alveolus & palate has been shown to interfere with maxillary growth & this made some people believe that no surgery should be performed on bony structure until maxillary growth is almost completed.[11]

The history of cleft lip repair is of a steady improvement in the cosmetic result, as surgeons have gradually come to appreciate the true nature of the defect. A repaired cleft lip may look excellent at rest, it will not move symmetrically unless the

muscles within it are properly realigned; the deformity will become apparent when the child smiles. Presurgical orthopedics should produce a similar effect in a more controlled but time-consuming way, and moreover can expand a collapsed lateral segment, which an adhesion cannot do. When the time comes for the definitive operation the available tissue does not have to be stretched across a wide gap, and the surgeon can concentrate on using it to the best advantage in constructing the features of the normal lip and nose.

Hallock was the first surgeon to advise a reliable method of closing cleft of hard palate & this operation is still widely practiced, with modifications.[11] The mucous membrane of hard palate is densely adherent to underlying periosteum & in this procedure the two are stripped off the bone as a single strong & thicker layer. The edges of the cleft are incised & the soft tissue of the two halves of the palate i.e. mucoperiosteum anteriorly & soft palate posteriorly are slide towards each other & stitched together in the midline. The mucoperiosteum is the elevated from the bone as a bipedicled flap, attached anteriorly & posteriorly & dissection is carried out through the posterior ends of the release incisions to relax the soft palate. If hard palate closure is to be delayed, the soft palate can be closed using an lntravelar veloplasty. The muscles can be covered by using flaps from the posteromedial hard palate or vomer, but this clearly involves losing some of the theoretical advantage of the procedure. The Furlow double reversing Z-plasty operation can also be adapted for use with delayed hard palate closure. After the soft palate has healed a large hard palate defect is left in front of it. The tension of the muscle suspend in the soft palate draws the halves of the maxilla together and narrows this fistula to such an extent that, when the time comes to close it, it may be possible to do so without making any lateral relaxing incisions, but simply by undermining the mucoperiosteum from the free edges of the palate. It is perhaps this conservative dissection, rather than the age at which it is done, which allows good maxillary development.

Delaire Regime

Delaire & precious close lip & soft palate at seven month, state that the vomerine mucosa must not be touched when repairing the hard palate.[14] They believe that the scarring produced by vomer flaps inhibits vertical growth of the maxilla & advocate closing the hard palate with a single layer of oral mucoperiosteum when this can be done with minimal elevation-at any age from 18 month to 6 or 7 year. Delaire also holds strong opinion on how the lip & nose should be repaired. The aim of the orthodontic treatment for children with complete cleft were limited to tooth alignment and arch expansion, followed by permanent artificial retention in the form of fixed bridgework or other prosthesis.[15-16] This was often linked with artificial restoration of the lateral incisor as the alveolar bony defect made orthodontic space closure impossible. Lack of supporting bone could also lead to so much loss of attachment of teeth adjacent to the cleft that they were lost after some years.[17] In patients with bilateral cleft, the mobility of the premaxilla made the retention of bridge-work difficult. Incisor alignment can be provided at this stage if requested for aesthetic regions. It is also possible that a correction of anterior crossbite at this stage will maximize the forward development of the maxillary dentoalveolar process. The optimum time of bone grafting will be decided individually.[18-19] In patients with a well-formed lateral incisor that is not too displaced from the arch, bone grafting is usually done quite early, around seven to eight years. Since the majority of patients have a missing ectopic or deformed lateral incisor, bone grafting may be postponed until 10 to 11 years. Small palatal defect do not interfere with speech, drinking or eating do not require obturator.[20-22] The closure of residual fistulae with tongue flaps. Palatal fistulae may be closed by obturator, constructed as an integral part of conventional denture. Speech bulps which extend into the velopharynx may be required where an unrepaired cleft of the hard & soft palate or velopharyngeal dysfuction.

CONCLUSION

This study was carried out to determine the facial growth & development in a patient with cleft lip & palate after surgical intervention. Healing process, growth and other parameter should be taken into account which actually determines future growth. As the patient has uncontrolled soft tissue growth after surgery, the impact of on speech, deglutition

should also be considered as one of the parameter which requires timely intervention with involvement of speech therapist & probably ENT surgeon. Multidisciplinary approach should be used in overall management of the patient in a holistic manner.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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