CASE REPORT

Assisted Feeding using Customized Feeding Appliance in Cleft lip and Palate Patients: A Report of Two Cases

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ABSTRACT

Cleft lip and palate (CLP) is one of the most serious congenital anomalies affecting children globally. The defect seriously impairs the health and living standard of an infant due to difficulties in feeding (nasal regurgitation), mastication, phonation, and esthetics. Maintaining a good palatal seal to generate adequate negative pressure required for the suckling of milk is highly essential in infants and could not be achieved in patients with cleft palate. Infants with cleft palate frequently experience nasal regurgitation and choking due to the inability of the palate to separate the nasal and oral cavities. Inability to receive adequate nutrition results in reduced weight and stunted growth in infants that hinder further course of surgical correction in CLP patients. Different types of feeding bottles with modified nipple shapes are currently available to facilitate feeding in CLP infants; however, such modified feeding bottles cannot generate the required palatal seal necessary to produce adequate negative pressure for the suckling of milk. Hence, a feeding prosthesis delivered in a single-visit appointment is always required to overcome not only the feeding difficulties in infants with CLP defects but also to avoid the inconvenience of multiple visits for the parents having CLP infants. Therefore, the present article depicts a report of two cases of assisted feeding using customized feeding prostheses delivered in a single-visit appointment to infants with CLP.

Key words: Nasal regurgitation, cleft lip and palate, cleft lip, cleft palate, feeding prosthesis

INTRODUCTION

Cleft lip and palate (CLP) is one of the most common congenital craniofacial defects. At the embryonic stage, a cleft lip occurs due to the failure in the fusion of the medial nasal process and maxillary processes, while a cleft palate occurs due to the failure in the process of elevation, migration, or fusion of the palatal shelves.[1] The global incidence of CLP is 1.4/1000 live births, while the global prevalence is about 0.45 in every 1000 live births.[2,3] Incidence of CLP in India has been stated to be 1 in every 781 live births.[4] The incidence of CLP is more in males compared to that in females, while the incidence of the isolated cleft palate is more in females.[5] The site predilection for clefting is more common on the left side.[4] The incidence of CLP is more evident among Asians followed by American Indians and has got less prominence among African Americans.[6]

The pathophysiology involved in the occurrence of CLP is highly complex, intriguing, and multifactorial in nature. Different genetic factors, environmental factors, and gene-environmental interactions are broadly found to be responsible for the occurrence of CLP.[7] Mutation of different genes such as the interferon regulatory factor-6 associated with Van der Woude syndrome,[8] muscle segment homeobox 1 (MSX1),[9] transforming

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growth factor-alpha (TGF-α),[10] T-box transcription factor-22 (TBX22),[11] bone morphogenetic protein 4 (BMP4),[12] paired box protein 7 (Pax7),[13] runt-related transcription factor 2[14] associated with cleft of the palate or submucosal clefting of the palate, methylenetetrahydrofolate reductase (MTHFR) gene,[15] wingless-type MMTV integration site family-member 9B (expressed in maxillary processes and medial and lateral nasal ectoderm),[16] and myosin heavy chain 9.[17] MSX1 gene plays a pivotal role in the regulation and stimulation of the homeobox protein which is essentially required in the dedifferentiation process of the cell; hence, mutation of this gene is strongly associated with CLP.[18] Similarly, the TGF-α gene plays a crucial role in the development of the palate and expresses itself in the medial edge epithelium of the palatal shelves.[18] Scientific studies have demonstrated a significant relationship between TGF-α and CLP.[10,19] Similarly, the TBX22 gene is known to encode a transcription factor responsible for the mesenchymal proliferation and elevation of palatal shelves before their fusion.[11] However, the Pax7 gene is a transcription factor shown to have a role in neural crest development by regulating the expression of neural crest markers. This gene is also expressed in the palatal shelves, Meckel's cartilage, and various nasal structures including the nasal epithelium.[13] Ludwig *et al.* have reported the significant role of Pax7 gene in the etiology of non-syndromic CLP defects.[20] The BMP4 gene plays a crucial role during the formation of cartilage, bone, and orofacial development.[18] Suzuki *et al.* have reported that mutation of the BMP4 gene in a child is highly associated with the occurrence of CLP.[12] MTHFR is the major enzyme involved in folic acid metabolism. Studies have reported several associations between the polymorphisms of the MTHFR gene and the risk of occurrence of non-syndromic CLP.[15,21]

Besides the genetic factors, environmental factors such as maternal smoking results in hypoxia and deficient detoxification mechanism and maternal alcohol consumption (reduced folate and retinoic acid synthesis) cause fetal alcohol syndrome, increased maternal age is associated with double and triple risk of occurrence of CLP above 35 and 39 years of age respectively, hyperthermia, stress, maternal obesity, occupational exposure (lead and aliphatic acids), ionizing radiation, nutritional deficiency (folic acid, B6, and B12), intake of excessive vitamin A supplementation resulting in extreme apoptotic activity, contracting different infections during pregnancy, exposure to chemicals, use of certain drugs like anti-abortifacient drugs, anti-metabolites like methotrexate which inhibits deoxyribonucleic acid synthesis through competitive folic acid antagonism, and anti-epileptic drugs like phenytoin, trimethadione, paramethadione, carbamazepine, valproic acid, and phenobarbital that reduce the rate of mesenchymal cell proliferation in facial prominences.[22-28] Furthermore, gene-environmental interactions such as the association of the rare TGF-α variant (TaqI C2 allele) and maternal smoking can increase the risk of cleft palate by 6–8 times.[29]

Children with CLP defects manifest a multitude of functional as well as esthetic problems which include feeding difficulties at birth such as difficulty in the suckling process, nasal regurgitation, hearing impairment due to the middle ear defect, speech impairment due to nasal escape, and deficient facial growth, thereby leading to numerous oral and esthetic problems at a later stage of the life.[30] Out of the several problems that children with CLP defects encounter, feeding is of utmost concern for any clinician as improper feeding hinders the normal growth pattern in a child. Furthermore, this can be a major concern for infants scheduled to undergo surgical correction for CLP. Hence, the present case report depicts a detailed account of the preliminary palliative care of fabricating a feeding plate to be used in infants born with CLP.

CASE REPORT 1

A 20-day-old female infant was brought to the outpatient department of the Department of Pedodontics and Preventive Dentistry with a chief complaint of facial deformity, difficulty in the suckling of milk, and nasal regurgitation on feeding. The family and medical history were non-contributory. The infant was the first child of her parents and weighed about 2 kg at birth. The infant's general health and weight were found to deteriorate due to improper feeding. On clinical examination, a left-side unilateral cleft of the upper lip, hard, and soft palate was observed (Veau's classification: Type III). To overcome the feeding problem, immediate palliative care by fabricating a feeding plate was undertaken and a further course of surgical correction was planned at its specific timeline.

Fabrication of the Feeding Plate

The first step involved in the fabrication process is the making of an impression for recording the

accurate details of the palatal surface. Elastomeric impression material (polyvinyl siloxane) was used for making the impression. A heavy body putty material was used, incorporating the base and catalyst in an equal proportion of 1:1 using a measuring scoop uniformly. Both the materials were added and kneaded well to obtain a moldable mass of uniform consistency. The moldable mass was then spread uniformly over the first of the clinician gloved using latex-free gloves roughly in accordance with the size of the infant's palate. The infant was held either upright or in a semi-supine position with a lowering down of the head at about 45 angulation to prevent aspiration of the impression material. The impression obtained was poured using diestone and the cast was retrieved after proper setting. For the fabrication process, a separating medium was first applied to the cast. Autopolymerizing clear acrylic resin was used following the sprinkle-on technique of polymerization. After polymerization, the feeding plate was removed from the cast and a bilaterally extending loop shape retentive wire component of approximately 6–8 mm was made from a 19-gauge stainless steel wire. The retentive wire component was, then, acrylized along the entire edge facing toward the cleft lip. After complete acrylization, the feeding appliance was trimmed, smoothened, finished, and minimally polished. The overextended borders were meticulously trimmed and made smooth to avoid irritation to the mucosa and to prevent any blockage of the airway passage [Figure 1]. All the necessary instructions about the feeding technique, cleaning, and maintenance of the feeding plate (to be stored in water, while not in use) were explained to the mother. Adequate maintenance of oral hygiene was to be maintained by wiping the developing gum pads with moistened cotton balls or commercially available baby oral wipers. A follow-up appointment after a week was scheduled to review the feeding ability of the infant with the help of the feeding appliance.

CASE REPORT 2

A 1-month-old male child was brought to the department of pedodontics and preventive dentistry with a chief complaint of an opening in the palate and difficulty in the suckling of milk. The infant was also diagnosed with a congenital cardiac anomaly since birth. The infant was the first child of his parents and weighed only 1.5 kg. The infant's general health and weight were found to deteriorate due to improper feeding. Family history was non-contributory. On clinical examination, only a cleft of the hard and soft palate was seen (Veau's classification: Type II). To overcome the feeding problem, immediate palliative care by fabricating a feeding plate was undertaken and a further course of surgical correction was planned at its specific timeline.

Fabrication of the Feeding Plate

A similar procedure as mentioned earlier was also followed for the fabrication of the feeding plate except for the cast which was poured using plaster of Paris and the acrylization of the feeding plate was done using self-cure acrylic resin [Figure 2]. All the other steps followed in the fabrication procedure were similar to those followed in the previous case.

DISCUSSION

CLP is a complex congenital defect caused due to an intricate interplay between a multitude of genetic, environmental, and gene-environmental interactions. Fogh-Andersen stated that genetic factors highly contribute to the occurrence of the non-syndromic CLP after observing an increased frequency of clefting in relatives of a patient with a cleft.[31] Similarly, Marazita *et al.*,[32] and Mitchell[33] reported that segregation analysis and twin studies, respectively, supported an underlying genetic etiology for the occurrence of the non-syndromic CLP. CLP has been shown to exhibit a high rate of familial history which significantly demonstrated that the risk of occurrence for CL has been estimated to be almost 32 times higher in children with a familial history compared to those without a familial history of CL.[34]

Prenatal screening is usually done by 2D and 3D ultrasonography (USG), magnetic resonance imaging, and karyotyping procedures. Chorionic villus sampling or amniocentesis is performed at 10–12 weeks and 15–18 weeks of the gestational period respectively to obtain fetal specimen tissue for karyotyping procedure, even metabolites from the embryonic palatal tissue, maternal amniotic fluid (choline level), and serum fluid can also be used to detect CLP defects at a prenatal stage.[35,36] USG is the most sought-after radiodiagnostic aid used for the prenatal detection of various congenital anomalies, where fetal facial morphology can be successfully evaluated by a transabdominal scan at about 16 weeks of the gestational period.[37] Jones and Klein *et al.*, (2006) have stated that almost 20–30% of fetuses with an orofacial cleft can be diagnosed prenatally during the routine USG of the expectant

Figure 1: (a-e) Depicting the pre-operative view of the CLP infant and fabrication of the feeding plate

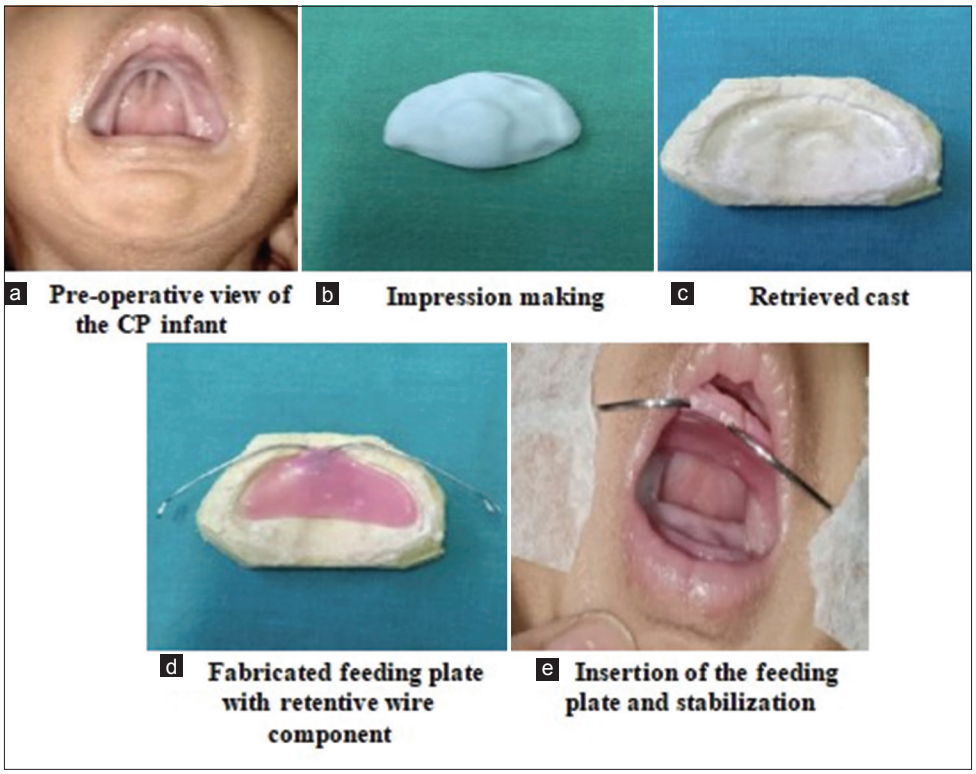

Figure 2: (a-e) Depicting the pre-operative view of the CP infant and fabrication of the feeding plate

mother.[38,39] Maarse *et al.*, have stated that 2D USG screening for CLP in a low-risk population has a relatively low detection rate since it is associated with few false-positive results, while 3D USG can achieve a reliable diagnosis for cleft lip and CLP defects. However, it is considered to be less reliable in detecting isolated cleft palate defects.[40] Limitations for obtaining a definitive diagnosis of clefting during a routine USG are mainly due to the lack of meticulous attention paid to the facial morphology in the absence of risk factors, position of the fetus, maternal obesity, multiple pregnancies, reduced amniotic fluid, and skills of the radio-diagnostician.[38]

Appropriate feeding is an important aspect to be taken care of in children with CLP. Feeding difficulties encountered in these children should be addressed immediately to provide preliminary palliative care to accelerate the growth process in CLP patients and to deem them fit to undergo surgical intervention at the appropriate timeline. An obturator or more commonly known as a feeding plate is delivered to an infant with CLP to attain the requisite suction pressure and to serve as a platform against which an infant can press any feeding object for suckling the fluid matter.[41] Jones *et al.* reported that the feeding plate prevents the tongue from intruding into the cleft, thereby facilitating the physiological process of suckling.[42] Adjunct to the feeding plate, there is also the availability of specialized feeding bottles with different designs such as the Mead Johnson bottle, Haberman feeder bottle, and feeding bottles with pigeon nipples to facilitate proper feeding.[43] Hence, proper fabrication of the feeding plate is highly recommended to facilitate adequate feeding, thereby necessitating the role of impression-making to be pivotal in the process of fabrication.

Impression-making is highly challenging in children with CLP defects, mostly due to certain limitations such as limited mouth opening, the small size of the oral cavity, and inadequate accessibility. Different kinds of impression materials such as heavy-body silicone impression material, polyvinyl siloxane, low-fusing impression compound, and alginate have been routinely employed for impression-making in infants with CLP defects.[44] In the present case report, an impression was made using elastomeric impression material (polyvinyl siloxane putty material) since it exhibits excellent properties of reproducing all the areas of interest with good surface details, resisting wear, and tear on the removal of the impression, thereby making the process atraumatic to the infant, its high viscosity reduces the risk of aspiration, and it also remains dimensionally stable to facilitate accurate recording of surface details on pouring multiple casts.[45]

Similarly, various materials are available for the fabrication of the feeding plate such as vacuum-adapted low-density polyethylene (ethylene-vinyl acetate), thermoplastic resin sheets, heat-cure acrylic resin, clear acrylic resin, and auto polymerizing self-cure acrylic resin.[44] An acrylic feeding plate provides a rigid platform to generate adequate pressure for suckling to facilitate appropriate feeding. Feeding plates made up of self-cure acrylic resin are not pliable in nature and their sharp margins can easily hurt the infant. However, these obturators are more cost-effective compared to those obturators made up of other materials.[45] In the present case report, the infants were delivered a feeding plate fabricated using clear and self-cure acrylic resin, respectively, to aid in the immediate delivery of the feeding appliance in a single visit and to avoid multiple visits. In the first case, clear acrylic resin was used for the fabrication of the feeding appliance, mostly due to its easy availability, adequate strength, and ability to maintain smooth margins on finishing and polishing to avoid impingement of the margins on the soft tissue. The fabricated plate was attached to an extended wire component by acrylization to facilitate easy and smooth insertion and removal of the prosthesis and also serves as a safety measure to prevent accidental aspiration of the appliance. Lodhi *et al.*, and Imthiyas *et al.*, fabricated a feeding plate using self-cure acrylic resin, due to the similar above-mentioned advantages.[45,46] Ali and Kamel and Hela *et al.*, also used clear acrylic resin for the fabrication of the feeding palate in patients with cleft palate.[47,48] In contrast to this, Agarwal *et al.*, used ethylene-vinyl acetate for fabrication due to its known advantages such as smooth surface, pliability, and less cumbersome due to the absence of the retentive wire component in comparison to that of a clear acrylic feeding appliance.[49] However, a few of its noted disadvantages are that it is highly expensive, compromises oral hygiene, and can cause irritation to the palate. Gupta *et al.* used thermoplastic resin sheets for the fabrication of the feeding plate since it also exhibited similar properties to that of the ethylene-vinyl acetate.[50] Considering all the advantages and disadvantages associated

with different materials used for the fabrication of the feeding plate, the authors of the present case report used clear and self-cure acrylic resin due to its advantageous properties of having adequate strength to provide a firm base for generating the necessary pressure to initiate suckling motion, cost-effectiveness, common availability, and ease of finishing and polishing to obtain smooth margins and contour of the appliance.

CLP patients entail an elaborative treatment regimen to improve their quality of life. Advanced treatment modalities ranging from cleft lip repair, primary and secondary palate surgery, bone grafting, and plastic surgery are carried out at specific timelines in accordance with the pattern of growth and development in a child. A multidisciplinary approach is needed for teamwork comprising a pedodontist, maxillofacial surgeon, pediatric surgeon, plastic surgeon, orthodontist, prosthodontist, otolaryngologist, pediatrician, nutritionist/dietitian, psychologist, audiologist, speech therapist, geneticist, nurse, and social worker.

CONCLUSION

The present case report gave a brief account of two cases of CLP requiring immediate palliative care by delivering a feeding plate to facilitate adequate feeding in infants. It emphasized the importance of preliminary palliative care which is essential to make an infant deem fit to undergo further surgical correction of CLP. The role of a pedodontist is highly pivotal not only in treating an infant with CLP but also in addressing the psychological aspect of the parents in bringing up children born with congenital defects. Hence, the role of a pedodontist is paramount in guiding an infant's overall growth and development from birth till adolescence.

DECLARATION OF THE PATIENT'S CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported to the journal. The patient understands that his/her name and initial will not be published and efforts will be made to conceal his/her identity, but anonymity cannot be guaranteed.

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