

# Early Prosthetic Rehabilitation in Newborns with Orofacial Cleft Using a Feeding Appliance: A Case Report and Review of Literature

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### ABSTRACT

A multidisciplinary approach is required for the successful management and rehabilitation of the infants born with orofacial cleft as there is difficulty in normal feeding, speech, hearing, and they also acquire recurrent nasopharyngeal and middle ear infections. Feeding is a major concern after birth in these infants and should be immediately addressed. Feeding appliance is a favorable feeding option in these infants as it creates a separation between oral and nasal cavities and thus helps in creating a negative intraoral pressure during suckling. This article presents a case report of infant with bilateral complete cleft lip and palate for whom feeding appliance was used as an immediate prosthetic rehabilitation method to resolve the feeding difficulties, along with reviews on the use of feeding appliance in infants born with orofacial cleft.

**KEYWORDS:** *Cleft lip and palate, feeding appliance, feeding difficulties*

## INTRODUCTION

Cleft lip and palate are the most common congenital anomaly of the craniofacial region affecting middle third of the face, with an incidence of 0.28–0.374 per thousand live births.<sup>[1,2]</sup> Cleft lip is an opening in the upper lip between the mouth and nose. It can be partial (ranging from slight notch in the lip), or complete (extending up to the nose). Cleft palate is characterized by the presence of abnormal oronasal communication. It can range from just an opening at the back of the soft palate to nearly complete separation of the hard and the soft palate. Cleft can be unilateral or bilateral, can occur in isolation or in combination, and can also involve other regions of orofacial complex. Among all the cleft patients, combined cleft lip and palate are the most common presentation (50%), followed by isolated cleft palate (30%), and then the isolated cleft lip (20%).<sup>[3]</sup> Cleft lip and combined cleft lip and palate are twice as common in males, whereas isolated cleft palate is twice more common in females.<sup>[4]</sup>

Cleft lip and palate may be syndromic or nonsyndromic in origin. Syndromic type is associated with other craniofacial malformations including Down's syndrome, Treacher Collins syndrome, Pierre Robin syndrome, Apert syndrome, and Waardenburg's syndrome.<sup>[5,6]</sup>

Isolated or nonsyndromic type cleft is multifactorial and heterogeneous in origin, and its origin is attributed to the genetic or environmental factors.<sup>[7]</sup> Family history of cleft in a first-degree relative increases the risk by 20%. Maternal periconceptional smoking and alcohol consumption,<sup>[8]</sup> obesity, higher maternal age, radiation exposure, maternal diabetes, and medications such as anticonvulsants may contribute to its etiology. In contrast, folic acid has been reported to have a protective effect.<sup>[6,9-11]</sup>

Abnormal oronasal communication and difficulty in making a lip seal around the nipple make it difficult for the newborn to create a negative intraoral pressure required for suckling.<sup>[12-14]</sup> Ineffective suckling results in profound weight loss and malnutrition.<sup>[13,15,16]</sup> Aspiration or choking may occur while feeding infants with cleft lip and palate as there is a lack of negative intraoral pressure necessary for triggering the swallowing reflexes. Feeding in cleft infants is also complicated by nasal regurgitation of oral fluid, burping due to excessive air intake during deglutition, gagging, and prolonged feeding time.<sup>[17,18]</sup>

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Cleft lip and palate is not merely a surgical problem its management involves multidisciplinary approach starting from obturator placement, surgical repair of the defect, orthodontic treatment to speech therapy. Surgical repair of the cleft lip and palate may completely close the lip defect and oronasal communication and is associated with better feeding, adequate velopharyngeal competence, and good speech and hearing development. Infants with cleft lip and palate are low birth weight, and malnourished therefore may not be able to withstand the surgical stress immediately after birth. The timing for the corrective surgical procedure for cleft lip and palate repair varies with limitation to the age, size, and weight of the neonate. This presurgical period is used to meet and maintain the adequate nutritional demand, promotes weight gain, and prepares the child to combat infection and helps to build strength to meet the stress of major corrective surgical procedure.<sup>[19-22]</sup>

Various feeding methods have been used in the past to resolve the feeding difficulties. Feeding appliance is a favorable feeding option in infants with cleft lip and palate as it creates a separation between oral and nasal cavities and provides a rigid surface to appose the breast or nipple during suckling.<sup>[23]</sup> This article presents a case report of feeding appliance placement in infant with cleft lip and palate and also review in detail about the use of feeding appliance in orofacial clefts.

## CASE REPORT

A 2-day-old female child was referred from the Department of Pediatrics to the Department of Dentistry in Dr. R.P. Medical College, Tanda, Kangra, with the chief complaint of difficulty in feeding due to palatal cleft. On examination, the child was found to be moderately built, with birth weight of 2.3 kg, having bilateral complete cleft of lip and palate (Veau's type IV) with no other facial malformation [Figures 1 and 2]. As



**Figure 1:** Extraoral view of infant with bilateral complete cleft lip and palate

immediate concern at this time was feeding and nutrition, decision to fabricate a feeding appliance was made. Parents were explained about the procedure, and the written and signed informed consent was obtained from them.

Customized perforated acrylic tray of adequate size that covers the entire palate along with the cleft was fabricated [Figure 3]. With nasogastric tube, *in situ* impression was made using polyvinyl siloxane putty material [Figure 4], with the neonate held in prone position to prevent aspiration of the impression material. A finger motion was used to clear the unset material posterior to the tray. The child kept crying during impression making which has an advantage of ensuring that the airway was patent. The impression was poured in type III dental stone [Figure 5].

Feeding appliance was made using a heat cure acrylic resin [Figure 6]. Appliance was trimmed, borders were rounded and polished to avoid trauma to the surrounding tissues. A hole was drilled in the obturator for tying a floss thread. Floss thread acts as a safety measure to prevent accidental swallowing and to facilitate easy removal of the appliance after feeding.

Appliance was placed in infant's mouth taking care not to irritate the surrounding soft tissue or activate gag. Fit of the appliance was checked, thereafter suckling response was observed by placing a finger in child's mouth. The child was able to suck and create a negative pressure onto the finger. Mother was instructed to breastfeed the child. Mother was instructed to manually express the breast milk into the neonate's mouth initially to compensate for the reduced suckling forces.

Initially, child was irritable and slow in suckling the milk. After few days, irritability decreased and mother was



**Figure 2:** Intraoral view of infant with bilateral complete cleft lip and palate



**Figure 3:** Prefabricated custom tray



**Figure 4:** Impression made using polyvinyl siloxane putty material



**Figure 5:** Impression poured in dental stone



**Figure 6:** Feeding appliance made of heat-cure acrylic resin with floss thread tied

able to feed the infant comfortably with the appliance in place. The infant was kept on regular follow-up to assess the success of feeding appliance by regular weight monitoring and for the periodic adjustments in the obturator.

Parents were instructed about the placement, removal, and cleaning of the feeding appliance. Appliance was advised to be worn only during feeding and should be kept submerged into the water when not in use. After feeding, thorough cleaning of infant's oral cavity and cleft with soft cloth or cotton soaked in warm water were advised.

#### Precautions to be taken while impression making

A mouth mirror should be used to depress the tongue and maintain the airway patency all the time. After impression making, wet cotton should be used to clean infant's mouth to remove any left remnants of impression material. Due to undercut engagement, difficulty in impression removal and fragmentation of the impression material may occur,

leading to aspiration and causing airway obstruction. In case of inadvertent aspiration, infant should be noted for the signs of airway obstruction. Back blows, chest thrust, and finger sweeps maneuvers should be used to relieve the airway obstruction. Blind finger sweeps should never be performed as it poses the risk of further pushing the fragments into the airways.

#### DISCUSSION

Children with cleft lip and palate are subjected to deficient facial growth, abnormal dentition, articulation defects, otologic problems, and velopharyngeal incompetence, therefore affects in normal functioning of speech, feeding, hearing, mastication, deglutition, and impairing the cognitive, linguistic, and emotional development in these children.<sup>[5,6,24]</sup> Among all, feeding is the worst problem present at birth and should be addressed immediately as it leads to ineffective intraoral suction,<sup>[20]</sup> nasal regurgitation of oral fluid, and low nutrition intake. Young *et al.* in their survey found that

feeding issues were a topic that 95% of parents with cleft lip and palate infant deemed “critical.”<sup>[3]</sup> Pandya and Boorman and Wilcox *et al.* showed in their studies the association of feeding difficulties in infants with cleft palate with failure to thrive and death in developing countries.<sup>[25,26]</sup> Furthermore, many authors reported slow weight gain in infants with cleft palate.<sup>[27,28]</sup>

A normal healthy infant holds the nipple in the mouth between the gum pads; lip seal is formed around the nipple, and expansion of the oral cavity is accomplished either through contraction of the tongue or by movement of the mandible to generate negative intraoral pressure. Following this, nipple is pushed against the palate and rhythmic tongue strokes are used to withdraw milk into the mouth. Failure to generate a negative intraoral pressure and form a lip seal around the nipple due to the oronasal communication and lip defect results in unsuccessful breastfeeding in infants with cleft lip and palate.<sup>[12]</sup> In infants with mild defects, feeding may be possible with some modification in mother and child position so as to obturate the cleft defect with mother’s breast. Children with isolated cleft lip have less interference during feeding than those with cleft palate.<sup>[13]</sup> A study conducted by Cubitt *et al.* in Uganda, reported greater malnourishment in cleft lip and palate group than in cleft lip before the surgery.<sup>[29]</sup>

Different approaches have been used in the past to resolve the feeding difficulties in cleft lip and palate infants that include, use of feeding tubes (orogastric and nasogastric), specially designed feeding equipments (compressible bottles and nipples), and the feeding appliance/palatal obturator. Feeding tubes are effective but should be used only for a limited period as they impair the development of normal suckling and swallowing reflexes of the infant. Specially designed bottles and nipples are available to facilitate easy feeding with reduced efforts which includes Mead Johnson nurser and Haberman Feeder.<sup>[17,30]</sup> These are soft, squeezable with enlarged opening thus allows feeder to deliver the milk directly into the infant mouth and also allows the infant to extract the milk from the bottle with reduced efforts. This is also called as assisted feeding.<sup>[31]</sup> Use of bottles and nipples was not sufficient for large clefts, and caution should be used with their use as rapid flow of milk into the mouth may impair infant’s ability to synchronize sucking, swallowing, and breathing. If not cleaned properly, these feeding bottles and nipples also increases the chances of infection in the infant.

It is in infants best interest to find a feeding technique as close to normal as possible, so it facilitates the normal physical and mental development of the newborn, maximizes oral stimulation and thus helps in normal oral

motor development,<sup>[32]</sup> and that can be used for large defects. Feeding appliance serves all the purpose and therefore is a good feeding option in infants with cleft lip and palate. Feeding appliance is passive prosthetic devices designed to provide a normal contour to the cleft alveolus and hard palate.<sup>[33]</sup> It restores the separation between oral and the nasal cavities and provides a surface to appose the nipple during sucking.<sup>[34,35]</sup> Feeding appliance has been used in cleft patients since ages and it greatly assists in direct breast feeding. The concept of early treatment of cleft palate patient with feeding appliance was pioneered by McNeil.<sup>[36]</sup>

Advantages of using a feeding appliance includes following.<sup>[19,21,23]</sup>

1. It helps to maintain the adequate nutrition by covering the cleft palate and providing a rigid platform toward which the infant can press the breast and extract milk.<sup>[37,38]</sup>
2. It assists in normal suckling and thus leads to the development of normal oromotor and swallowing reflexes
3. Reduces the feeding difficulties such as nasal regurgitation, choking, and shortens the feeding time
4. It positions the tongue in correct posture preventing it to enter into the defect, thus helps in the growth of maxilla and the maxillary shelves toward each other.<sup>[39]</sup>
5. Reduces the passage of milk into the nasopharynx and thus reduces the incidence of nasopharyngeal infections and otitis media.<sup>[40-42]</sup>
6. It also helps in presurgical nasopalveolar molding.<sup>[43]</sup>
7. After cheiloplasty provides cross arch stability and prevents maxillary arch collapse.<sup>[43]</sup>

Fabrication of feeding appliance in a newborn poses unique set of challenges including the size constraints of the oral cavity, variation in size, location, and severity of the cleft, infant’s inability to cooperate and follow commands. Selection of accurate impression tray size and the position to be adopted for impression making also is a critical step. A number of positions have been adopted including prone,<sup>[44]</sup> face down,<sup>[45]</sup> upright,<sup>[45]</sup> and even upside<sup>[46]</sup> down to prevent the accidental aspiration of the impression material. Customized perforated acrylic tray or commercially available prefabricated trays are used. Some authors have also advocated the use of wax and ice cream stick to carry the impression material.<sup>[47]</sup>

Various materials that have been used for making impression of cleft defects that include low fusing impression compound, alginate, and elastomeric impression material. Alginate has low tear strength and tears when extrudes deep into the cleft undercuts, therefore, is not suitable for making impression in cleft infants. Low fusing impression compound has good tear

strength. However, it is a thermoplastic material and if overheated can lead to scalding or burning of the oral tissues, also it has a volatile content leaching out that can be harmful to the infant. Elastomeric impression materials are considered superior for making cleft impressions due to good elastic behavior, high tear strength, accurate reproduction of surface detail, and a long dimensional stability enabling multiple cast pouring using the same impression. The best results are obtained with putty wash additional silicon impression.<sup>[48]</sup>

Feeding appliance can be made using acrylic resin (self-cured or heat cured), visible light cure acrylic, silicones, vacuum-formed polyethylene. Vacuum-formed obturator is light in weight, soft with smooth surfaces, and do not require any retentive wires, but they are expensive and their plastic content can irritate the palatal tissue. Infants feeding obturator is traditionally fabricated with acrylic resin. Heat-cure acrylic resin is preferred over self-cure acrylic resins as leaching out of the monomer from heat cure is less compared to self-cure acrylic resin that may cause allergies, more stability in the shape and size, and more finished surface of the heat-cured appliance. Heat-cure appliances have additional advantage of being more cost effective, easily available, simple to fabricate, good strength, and are easy to clean and maintain oral hygiene.<sup>[49]</sup>

Jones *et al.*<sup>[21]</sup> compared the feeding difficulties before and after the use of obturator in cleft lip and palate infants. After 8 months of wearing obturator reduced choking, nasal discharge, and bottle feed duration and improved parental confidence were reported. Goldberg *et al.*<sup>[19]</sup> reported improved weight gain with the infant having cleft of the soft palate with the use of feeding obturator.

No single intervention can be prescribed with confidence to improve the feeding outcomes for infants with orofacial cleft. The most appropriate feeding technique for the cleft infants would be any technique that delivers adequate milk into the mouth and allows the infant time to swallow, helps in normal physical development, and weight gain. A number of authors advocated the use of supplemental feeding for infants with cleft palate who were breastfeeding.<sup>[50,51]</sup> Feeding technique combination has shown to positively influence growth, and weight gain in cleft child therefore should be used along with the feeding appliance.

There is limited but promising evidence to support the use of palatal obturator and lactation education. Balluf and Udin<sup>[52]</sup> reported improved weight gain and shorter time to surgery in a case series of 57 bottle-fed infants with cleft lip and palate after early parental lactation

education and prescription of a feeding obturator. Similarly, in a study by Turner *et al.*,<sup>[53]</sup> five infants were studied to examine the effect of lactation education and the use of palatal obturator/feeding appliance. Time taken to breast or bottle feed the infant, volume of milk intake, and growth in infant at 4 weeks of age with cleft lip and/or palate compared to birth was found to be improved with the combined use of palatal obturator and lactation education.

Lactation education includes correct positioning of the infant during feeding, effect of cleft on feeding, bottle use, and advice on feeding duration and breast feeding techniques and observation of infants feeding cues. Upright<sup>[34-35]</sup> or semi-erect<sup>[54]</sup> feeding posture was recommended to increase the effectiveness of breastfeeding and minimizes the risk of nasal regurgitation and otitis. Small, frequent feeding episodes should be practiced as lengthy period may cause excess calorie expenditure with failure to establish well-defined period of satiety and hunger in the infant. Sidoti and Shprintzen suggested a 20 min feed duration for infant with cleft palate, with no more than 30 min as the upper limit.<sup>[55]</sup>

## CONCLUSION

Infants with cleft lip and palate require special attention and a team approach for correct management. Feeding difficulties should be immediately addressed and managed in these infants, as it not only delays the development of the newborn but also creates parental anxiety. Use of the feeding appliance aids in better feeding, speech, promotes adequate nutrition and weight gain, reduces nasopharyngeal and middle ear infection until the surgical correction of the defect is carried out and therefore ensures normal physical, mental, and psychological well-being of the infant and the parents.

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## Conflicts of interest

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