

IMPACT OF SURGERY AND TELE-REHABILITATION ON FEEDING AND EARLY ECHOIC SKILLS DEVELOPMENT IN CHILDREN WITH CLEFT LIP/ PALATE DURING COVID-19 PANDEMIC

Priyadharshini R Ethiraj MSLP¹, Lal D V DCH, DNB, FSASS², Divya K R³, Surya Rao Rao Venkata Mahipathy MS, MCh (Plastic Surgery), FRCS (Glasg), FACS⁴, Praveen Ganesh N BDS, MDS, FCCS⁵

¹Assistant professor, Department of Speech-Language and Hearing Sciences, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha Medical College and Hospital, Thandalam, TN, India.

²Program co-ordinator, Saveetha Child Development Centre, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha Medical College and Hospital, Thandalam, TN, India.

³Senior Resident, Department of Plastic and Reconstructive surgery, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha Medical College and Hospital, Thandalam, TN, India.

⁴Professor & Head, Department of Plastic and Reconstructive surgery, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha Medical College and Hospital, Thandalam, TN, India.

⁵Dental Professor & Craniofacial Surgeon, Department of Plastic and Reconstructive surgery, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha Medical College and Hospital, Thandalam, TN, India.

Declaration of Conflicting Interests: None of the authors have any conflicting interests directly or indirectly.

Funding (Financial Disclosure): No external funding was involved in the study. However, help was offered by Smile Train India for the conduct of CL/P surgeries.

Ethical Approval /Patient consent: Institutional ethical committee approval was sort before study (SMC/IEC/2020/05/004) and written informed consent was taken for surgery as well as for the study separately.

ABSTRACT

Objective: To examine the parental knowledge, practices as well as perspectives about the feeding and investigate the early speech development (Early Echoic Skills) in children with Cleft lip and/or Cleft palate (CL/P).

Methods: Forty eight children of 2-24 months of both sexes, with CL/P (Cleft lip, Cleft palate, Cleft lip and palate, Small notch on lip, Split Uvula) recruited by consecutive sampling were analysed with a validated questionnaire on KAP of feeding practices and Early echoic skills and by clinical examination pre and post-surgery using direct and follow-up tele-interview.

Results: Overwhelming majority (93%) reported increased confidence and skills in feeding and 58.3% reported improvement in speech and language development through tele-rehabilitation. Nasal regurgitation was the major difficulty (68.7%) in pre-operative assessment, while in post –op assessment it was only 27%. Large variability in opinion on prevalence and treatment of feeding and effectiveness of treatment strategies designed to reduce nasal air emission was evident.

Conclusions: The findings of this study indicate a high degree of variability in opinion of parents regarding feeding and early echoic skills of children with CL/P. Parental education was helpful in supporting the needs for better feeding of children affected by CL/P. A multidisciplinary approach with parental involvement along with good parental awareness programmes for feeding and in early speech and language development (early echoic skills) before and after surgery creates significant improvements beyond cosmetic appearance by helping in speech and language development.

KEYWORDS: Feeding; Swallowing; Tele-rehabilitation; Early echoic skills; Cleft lip; Cleft palate.

INTRODUCTION

Cleft lip and palate (CL/P) are the commonest congenital abnormalities seen in childhood. CL/P and craniofacial syndromes are often associated with special challenges in feeding, swallowing process and beyond in children. The majority of CL/P appears to be due to a combined effect of genetics and environmental factors. (1) Children with CL/P present with several problems including in feeding², the degree of which will vary with the type and severity of the CL/P (unilateral, bilateral, incomplete, and complete). (2) Severe feeding and swallowing dysfunctions have the potential to cause nutritional and respiratory problems and create significant stress for families and caretakers. Children, especially those with a cleft palate, have improper sucking mechanism. In spite of having adequate sucking movements, they fail to generate the negative pressure required for sucking effectively to extract feeds. Generally, feeding takes a longer time and the child may tire before completion of the feed. (3) The caretaker and infant will be subjected to stressful feeding due to decreased volume of intake by the infant, leading to prolonged feeding time. The possibility of treating children with CL/P and growth deficits makes it essential to assess the growth of these children early. Orofacial cleft being a highly prevalent congenital anomaly needing multiple interventions makes it essential for a multidisciplinary approach in management. After birth,

feeding becomes a dilemma due to lack of intraoral negative pressure and improper nipple seal due to anatomical defects, compromising effective sucking and swallowing. Ineffective sucking predisposes to devastating effects on growth, choking, and dehydration. The dearth of data focusing on feeding problems and approaches to be developed to compensate for such difficulty in infants with CL/P is glaring. Breastfeeding, Sangu / Paladai (a traditional south Indian feeding utensil), and spoon are feeding utensils parents and caregivers use. Sangu/ Paladai feeding is the first choice for parents with CL/P in South India. The infant with syndromic CL/P and associated neurological issues will likely have feeding and swallowing issues that will require ongoing input from the paediatric speech pathologist concerning treatment and management. Children with CL/P may also have delay in speech and sound production. These children have poorly understandable resonance, the resonance may take on a nasal twang, and the speech may be difficult to understand. Though all children may not have these problems; reconstructive surgery may be curative for some, but many still will have speech disorders. The characteristics of speech disorders are mainly a combination of different articulatory features affecting not only the intelligibility but- also interfering in the social competence and emotional development. In clinical practice, articulation disorders are mainly evaluated by subjective tools, of which the simplest method is auditory perception, mostly performed by a speech-language pathologist.

This study was intended to profile the Parental knowledge, practices, and perspectives about the feeding of children with CL/P when it is corrected through surgery followed by tele-rehabilitation. It also aimed at investigating the early speech development (early echoic skills) in children with CL/P when a speech deficit was intervened through tele-rehabilitation during COVID-19 pandemic.

METHODS

A total of 48 children from birth to two years of both sexes, with CLP (Cleft lip, Cleft palate, Cleft lip and palate) were included in the study. This study conducted at a tertiary care teaching hospital in South India by recruiting the study subjects by consecutive sampling from the inpatient department. The children included in the study were admitted at the Cleft centre for surgical treatment from October, 2020 to July 2021. The mean age at which Cleft lip surgeries were performed was 45 days, and cleft palate closure was done around a mean age of 9 months, after getting written informed consent from each parent and approval of institutional ethical committee (SMC/IEC/2020/05/004). Demographic information, the Knowledge, Attitude and Practices (KAP) of feeding and their speech and language development was assessed using questionnaires as well as by clinical assessments, both pre and post-surgery. The questionnaires developed had two sections-A & B consisting of open and close-ended questions. Section A comprised of questions related to demographic information of the children, like name, age, sex, mother tongue, languages known, type of the cleft, contact details. Additionally, questions were related to feeding practices, what kind of utensils were used during feeding, experiences of time spent together during feeding (duration), additionally feeding difficulties were mentioned. Section-B comprised of 5 questions related to speech and language development to assess early speech development (Early echoic skills) in children with CL/P. In Section B, an empty space available for parents to document their comments and opinions if required. Both pre-op and post-op questionnaires consisted of same questions.

In this study the pre- and post-operative assessments were done for feeding practices and speech development (early echoic skills). In the pre operative assessment period, if delay in speech and language were found, they were offered a detailed speech and language assessment through Language Assessment Tool (LAT). Patients who satisfied the inclusion criteria and who gave signed informed consent were included for the study. We recruited 48 children for the study and followed STROBE guidelines. Language assessment test is a common tool which is used to measure receptive and expressive language in children within 0-3 years of age. Six children dropped out post-surgery following restriction due to COVID-19 pandemic.

Criteria for selection

The inclusion criteria for the study were as follows: Non-syndromic children with CL/P with systemic or congenital abnormalities; children who were operated on for CL/P at the Saveetha Medical College and Hospital, Kancheepuram within age group of birth to 2 years.

Exclusion criteria

The exclusion criteria were: children more than 2 years of age; and incomplete repair of CL/P. Syndromic cleft infants were excluded from the study as they may have comorbidities including neurological affecting the early speech and language development.

Statistical Analysis

The data were recorded on a standard proforma and entered on a master sheet. The paired-t test was applied to identify any statistical significance in the feeding practice and speech development in children with CL/P. Statistical analyses were performed using SPSS 23 and graphs were prepared using PRISM V software. P values (<0.05) were found where ever applicable.

RESULTS

Total number of patients dropped off from the post-surgery (6) due to Covid -19 pandemic. After the post-operative assessment, parents were trained on both feeding and speech developmental skills and intervention is given through tele-rehabilitation (Total-4 hours 30 minutes; 30 minutes therapy, 15 minutes counselling x 6 sessions).

The present study was aimed in profiling parental knowledge, practices and perspectives about feeding and language development in young children with CL/P. A total of 48 children with CL/P participated in the study. The results of the study are mentioned below.

In this study 48 parents whose Children suffered from CL/P deformity and underwent corrective surgery. Of the 48 cleft patients, (n=25) 52% were males and (n=23) 47.9% were females, 20% had isolated cleft lip, 76% had isolated cleft palate and 4% had CL/P. (**Table 1**)

	Severity	Right n (%)	Severity	Left n (%)	Total n (%)
1. Nostril Arch	-	-	-	-	0%
2. Nostril Floor	1	1 (100%)	2	1 (100%)	100%
3. Prolabium	-	-	-	-	-
4. Upper lip	2	4 (8.3%)	2	4 (8.3%)	100%
	3	1 (2%)	3	5 (10.4%)	
5. Alveolus	2	1 (2%)	1	1 (2%)	100%
6. Pre-incisive trigone	1(2)	1 (2)	100%		
7. Hard palate	1 (3) (6.25%)	1 (3) (6.25%)	100%		
	2 (9) (18.75%)	2 (7) (14.5%)			
	3 (3) (6.25%)	4 (1) (2%)			
8. Soft palate	4 (6) (12.5%)	3 (5) (10.4%)	100%		
	3 (5) (10.4%)	4 (2) (4.1%)			
9. Velopharyngeal valve function	-	1(1)	100%		

Table 1. Count and percentage based on Friedman's modification of Y classification of CL/P

Practices related to feeding

1. Feeding characteristics of study cohort

The practice of feeding from birth to 24 months in children with CL/P was investigated. In pre-assessment, 100% of mothers with children having CL/P had a negative attitude towards feeding in general, while in post-assessment 93.7% still retained negative attitude towards breast feeding. Although 86.1% of babies were initiated with breastfeeding only 10% of mothers were successful. Among the children with CL/P, 68% were able to hold bottle and 52% able to hold spoon independently, where 32% were not able to hold bottle and 48% not able to hold spoon independently in both pre-and post-assessments. Though children with CL/P from birth to 24 months did not experience an increase in infection as evidenced by no ear pain/ discharge in 77%, there was statistically significant difference in nasal regurgitation in pre and post assessments ($p < 0.03$).

Analysis of completion of entire meal and utensils used for feeding as well as duration of feeding was found to be statistically significant ($p < 0.001$). In the present study, 32% infants were not fed with bottles, 68% (n=17) had fed with conventional feeding bottles and 88% used Paladai / Sangu (a traditional south Indian feeding utensil) for feeding. The most common feeding method practiced, among all the subjects (n = 48), was Paladai followed by spoon (52%), bottle feeding (68%) which was found to be statistically significant at < 0.001 . About 48% of the parents reported that feeding method was practiced every 2 hours. Regarding the position of the child while feeding, (n=24) 50% of the parents followed the lie on top position, followed by (n=12 each) 25% each of individual sitting and semi-sitting position. Children among all the subjects, 64.5% (n=48) of the parents reported difficulties in feeding child with cleft (**Table 2**). Nasal regurgitation was the major difficulty reported by about 68.7% in pre-op assessment, while in post -op assessment only 27% difficulties were reported (n = 48) of the subjects and found to be statistically significant ($p < 0.001$). In a few cases vomiting on burping and inability to breastfeed the child were reported.

Nasal regurgitation		Vomitting or Burping		Unable to breast feed		Often cold	
Pre-op	Post-op	Pre-op	Post-op	Pre-op	Post-op	Pre-op	Post-op
68.7%	27%	31.2%	27%	6.25%	6.25%	25%	20.8%

Table 2. Feeding difficulties experienced by the children with CLP

2. Feeding challenges and speech evaluation

The isolated cleft lip infants showed no feeding difficulties in 76%. However, the remaining 24% presented minor issues such as lack of generating proper lip seal and milk regurgitation. Other problems (64%) reported by the parents were aspiration, coughing, choking, reduction in weight gain and inadequate growth.

The speech sound abilities of children with CL/P at 14-18 months was assessed. This study also recorded a few challenges like utterance of first word after palatal surgery was performed. More than one third of the parents (32.3%) reported that despite their efforts in engaging a child they failed to consistently get the child's attention. A few other participants (17.6%) reported difficulties in making children to imitate the sounds, actions, words and phrases. Because of the limitations of age group (<2 years) and anatomical variations with CL/P present in all, none of the children could attain the above skills, though they could imitate sounds and actions. The early echoic skills compared with type of cleft, imitation of simple and reduplicated syllables followed by three syllable combination, was found to be statistically significant at <0.001. (Table 3)

Can your child imitate/ make simple and reduplicated syllables		Two syllable combination- explain what all can the child express		3-syllable combination		Prosody: spoken phrases	
Pre	Post	pre	post	pre	post	pre	post
35.4%	79.1%	64.5%	76%	18.75%	60%	14.5%	60%

Table 3. Representation of early echoic skills in children with CLP Pre and post Tele-rehabilitation

Out of the 48 children, 93% (n=45) parents had reported increased confidence and skills in feeding their children and 58.3% (n=28) reported improvement in speech and language development through tele-rehabilitation (Total- 4 hours 30 minutes; 30 minutes therapy, 15 minutes counselling x 6 sessions). The comorbidities we found in the small group of CL/P patients are Hyperactivity, Mixed Receptive and Expressive Language disorder, Stuttering, Global Developmental Delay.

DISCUSSION

Healthy well-nourished children are the dream of every parent. CL/P as facial deformity impose a shock for every parent. Feeding is a huge challenge for parents, caregiver and health care team before surgical correction is done in CL/P. In this preoperative period for adequate weight gain, it is important to have adequate feeding intervention as they lack the ability to latch to the nipple, suck, and maintain proper seal. The knowledge, attitude, perspectives and awareness about the feeding methods for children with CL/P are very important for their parents. Even though the mothers are encouraged and motivated to breastfeed the child, most parents follow the method best adapted to their child. The most common feeding difficulty encountered by a CL/P child is the inability to suck milk due to non-achievement of effective negative pressure. (4-6)

In the present study, the difficulties posed while feeding a child with cleft, from mother's perspective were, nasal regurgitation as the major difficulty (68.7%) while 31.2% reported vomiting on burping. While assessing the feeding methods practiced by the mothers and the duration and position of feeding, it was found that at most of the hospitals/centres, the mothers were advised to express breast milk under sterile conditions and feed the child through Sangu / Paladai at a frequency of every 2 hours in an upright and in semi-sitting position. The preferred positions for feeding were 60% upright and 68% semi-sitting while feeding, were found to be in line with other studies⁴. The second most commonly used method include 52% spoon feeding, whereas few mothers practiced feeding with dropper and a 60.4% Sangu and glass as well. A similar study⁵ reported that spoon feeding (57.4%) and it's finding concord with feeding dropper, bottle with a Y-cut syringe, and a glass as well. Breastfeeding was also encouraged at a few centres.

Feeding difficulties and challenges had been reported by previous studies Almost all parents who participated in this study (97%) are keen to learn ways to fight back these challenges. Similar studies⁶ have reported lack of proper instructions to face feeding difficulties (40%) and few (55%) were given brief information on feeding interventions. In the present study, out of the 48 children, 93% parents had reported that they can feed their children

and 58.3% reported improvement in speech and language development through tele-rehabilitation. In a meta-analysis on speech therapy through tele-practice, lower percentage (17%) of studies addressed speech therapy through tele-practice and 83% of the scientific articles reported tele-practice in clinical setting. (7) Even after surgery and counselling only 6.25% (n=3) of parents could breastfeed their infants, leaving a large percent 93.7% (n=45) without the benefits of direct breastfeeding, probably the result of initial recommendation for withholding direct breast feeding and also due to nasal regurgitation following attempted breast feeding by the parent. Nineteen of those children had CL/P, 22 had cleft palate (CP) and 14 had isolated cleft lip (CL). Isolated cleft palate and CL/P reported lowest success rate in breastfeeding than cleft lip. Though infants with isolated cleft lip showed no feeding difficulty, the remaining 15.3% presented with minor issues such as lack of generating proper lip seal, milk regurgitation, postpartum fatigue which is in congruence with other studies. (8) Several studies reported the benefits of using syringe in compassion to spoon for increasing the volume of intake and speeding the normalisation of feeding time as in normal children. (9) Feeding bottle as an intervention was effective in providing infants with proper quantity needed for better weight gain. It has been reported that 45 of the 48 mothers with CL/P infants tried to breastfeed, but none of them was successful. It was observed that infants with CL/P from birth to 24 months were at risk for common manifestations of infection, such as middle ear infection, fever and diarrhoea, when compared to normal infants. The usage of bottles or spoon which could possibly result in contamination could be the reason for recurrent infections in CL/P children. Normal breast-fed infants were less susceptible to common infections. A previous study¹⁰ has reported that breast-feeding provides protection against upper respiratory infections, particularly otitis media. (10)

One of the significant and fundamental results of the study was its influence on parental perception about the impact of early echoic skills on speech and language development in children with CL/P from birth to 24 months. It is well known that the CP condition which affects the physical structure of the mouth and face has potential consequences for normal speech and language development. Although the children with CL/P were still behind their non-cleft peers for number of different sounds produced, they appeared to be making steady improvements. (11)

In the current study, based on parental reports, the impact of early echoic skills on speech and language development was significant in pre and post operative assessment. Before attending the tele-rehabilitation, nearly 35.4% of the children doesn't imitate the simple syllables and after rehabilitation, 79.1% were able to imitate the simple and reduplicated syllables. The results of the present research support the hypothesis that exposure to early echoic skills is positively related to young children's language abilities. During tele-rehabilitation, parents were trained about home training programme (i.e. questioning their children). Parents questioning their children provides the occasion of structure response, to use language and to improve their language abilities. In the current study, before attending the tele-rehabilitation, parents reported that two and three syllable combination were not present for the appropriate developmental age. After attending tele-rehabilitation, (76%) two and (60%) three syllable combination developed age appropriately. It may be noted that present study reproduces comparable results to previous studies done with face-to-face rehabilitation. This proves that tele-rehabilitation, post-surgery if done with properly selected patients and methods can produce comparable results to face-to face therapy and helps in reducing cost of therapy and save time and effort.

Of those in the cleft group, 8% had a diagnosis of ADHD which is in line with the observation that children with CL/P are at increased risk for hyperactivity or inattention needing multi-centric studies to determine this aspect. 8.3% of children were diagnosed with delay speech and language skills.

Parents overall satisfaction

The parents who participated in the study gained extensive knowledge about feeding and early speech development through tele-rehabilitation. Parents expressed desire to receive more professional support, and public education to create positive environment for feeding and early speech development.

CONCLUSION

The paucity of knowledge and awareness among parents about the feeding practices in CL/P is glaring. Parental education is helpful in supporting the needs for better feeding of children affected by CL/P. Feeding and swallowing difficulties can be corrected to a considerable extent by early interventions in non-syndromic CL/P population. The availability of evidence to support the use of specific therapy techniques, such as assisted feeding methods, is emerging for children less than 2 years with CL/P. The results of this preliminary study indicate the differences between bisyllabic and multisyllabic productions in children with CL/P less than 2 years. Therefore, a multidisciplinary team for corrective approach and management is the best suite in such cases and the team may include speech language therapists, social workers, nurses, caretaker and different specialities. Tele-rehabilitation if done with proper selection of children and methods can produce same results, saving time and money, especially at times of pandemic/resource constraint settings.

Recommendations: The multidisciplinary team should provide awareness programmes and training as well as participation to parents of children with CL/P for feeding and in early speech and language development (early echoic skills) before and after surgery and ensure continuous follow up and periodic assessments for supports

needed.

CONFLICT OF INTEREST:

Authors declare that there is no conflict of interest.

REFERENCES

1. Lee, S T. (1999). New treatment and research strategies for the improvement of care of cleft lip and palate patients in the new millennium. *Annals of the Academy of Medicine, Singapore*, 28(5), 760-767
2. Miller, C. Feeding Issues and Interventions in Infants and Children with Clefts and Craniofacial Syndromes. *Seminars in Speech and Language*. 2011;32(2):115-126.doi:10.1055/s-0031-1277714
3. Chase, L, Starr, D, Tvedte, C, Wagner, B. Comprehensive nursing care for cleft patients. In: Bardach, J, Morris, HL eds. *Multidisciplinary management of cleft lip and palate*. Philadelphia: Saunders, 1990
4. Goyal, A, Kaur, M, Jena, A. Nature of feeding practices among children with cleft lip and palate. *Journal of Indian Society of Pedodontics and Preventive Dentistry*. 2012;30(1):47. doi:10.4103/0970-4388.95581
5. Young, JL, O'Riordan, M, Goldstein, JA, Robin, NH. What information do parents of newborns with cleft lip, palate, or both want to know? *The Cleft Palate Craniofacial Journal*, 2001;38, 55–58. <https://doi.org/10.1597/1545-1569-2001-038-0055>
6. Visser, EE, Krüger, EE, Kritzing, AM. Feeding difficulties in infants with unrepaired cleft lip and palate and HIV-exposure. *African Health Sciences*. 2018;18 (4):1098. doi:10.4314/ahs.v18i4.31
7. Attia, H. Cleft Lip and Palate Nutritional Assessment and Feeding challenges. *Egyptian Dental Journal*. 2019;65(4):3357-3364. doi:10.21608/edj.2019.74769
8. Danner, SC. Breastfeeding the infant with a cleft defect. *NAACOG's clinical issues in perinatal and women's health nursing*, 1992; 3(4), 634-639
9. Jones, CE, Chapman, KL, & Hardin-Jones, MA. Speech development of children with cleft palate before and after palatal surgery. *The Cleft palate-craniofacial journal*, 2003; 40(1), 19-31
10. Klatt, RE. (2002). Hyperactivity and inattention in children with isolated cleft lip and palate or isolated cleft palate [Master's thesis, University of Cincinnati]. OhioLINK Electronic Theses and Dissertations Center. http://rave.ohiolink.edu/etdc/view?acc_num=ucin1027946980
11. Kalita, S, Girish, KS, Mahadeva Prasanna, SR, Dandapat, S. Objective assessment of cleft lip and palate speech intelligibility using articulation and hypernasality measures. *The Journal of the Acoustical Society of America*. 2019;146(2):1164-1175. doi:10.1121/1.5121310