European Journal of Molecular & Clinical Medicine (EJMCM)
ISSN: 2515-8260 Volume 09, Issue 01, 2022

Original research article

A Retrospective Study On Cleft Lip(Palate) And Associated Anomalies In Childrens

[1] Dr Sadhna Yadav,[2] Dr Zalak Modh,[3] Dr Manish D Patel

[1]Assistant Professor, Department of Otolaryngorhinology, Banas Medical College and Research Institute Palanpur, Gujarat

[2] Senior Resident, Department of Otolaryngorhinology, Banas Medical College and Research Institute Palanpur, Gujarat

[3]Assistant Professor, Department of Otolaryngorhinology, Banas Medical College and Research Institute Palanpur, Gujarat

Corresponding Author: Dr Manish D Patel

Abstract

Introduction and objective: All types of cleft-associated anomalies do not occur with equal frequency. It has not been well-known if specific types of anomalies are commonly related with clefts, or which organ is most commonly affected. Cleft lip and palate (CL and P) could be associated with many other structural abnormalities of the adjacent vital structures of the face.

Methods: Three hundred seventy consecutive syndromic and non-syndromic children with CL and P patients in Department of otolaryngorhinology ,Banas Medical college and research institute ,Banas Kantha, District and adjoining area of Gujrat and Rajasthan India, from Dec 2018- Dec 2020 were studied. Data were analysed using Statistical Package for Social Sciences (SPSS) version 16.

Results: A total of 370 cleft lip and palate patients were managed. More than fifty-five percent were male and 44.3% were female. 9.7% patients had ocular anomalies. Out of 36 CL and P patients, 41.7% patients had bilateral cleft lip and palate, 22.2% patients had unilateral cleft lip and palate.

Conclusion: The present study of Cleft lip and palate are the most common facial deformity and clefting may involve lip only, lip and palate and palate only

Keywords*:* Cleft lip and palate, Congenital anomaly

Introduction

Isolated cleft palate (CPO) is the least common form of oral clefting (approximately 33% of all oral clefts), affecting 1 to 25 per 10,000 newborns worldwide. Because of the rarity of CPO and its distinct embryologic origins and recurrence risks from cleft lip with or without cleft palate (CL/P), most studies either exclude CPO or conflate these cases with CL/P due to hypothesized common genetic and epidemiologic risks, although no studies to date have had sufficient power to evaluate these hypotheses. Thus, our knowledge about whether the risk factors for CPO do indeed differ from those of CL/P remains incomplete. Although much remains to be confirmed in human studies, there is a wealth of information from animal models

on the molecular biological pathways necessary for complete closure of the primary and secondary palate during embryogenesis.Clefts of the lip and/or palate CL/P are the most common congenital malformation of the head and neck (1). Prevalence rate for live births with cleft palate, cleft lip, or both was 1.39 per thousand live births (2). Although the incidence varies among different ethnic groups, highest amounts have been reported among Asians (3, 4), and the least amounts have been found amount Afro-Caribbean populations (5). Majority of CL/P Patients suffer from feeding difficulties in infancy and speech, hearing and dental problems as they grow older, and life-long social and psychological problems due to the facial deformity. The cause of cleft lip and palate is complex. Genetic and environmental risk factors have been identified as triggers for syndromic CL/P; however, the aetiology of the more common non-syndromic CL/P remains largely unknown (6). geographical area, Gender, population, dietary habit, use of drugs, tobacco use, drinking alcohol, low contaminated water sources and birth weight have all been theorized as factors cumulative the incidence rate of CL/P in new-borns (7-11). Cleft lip and palate (CL and P) could be associated with many other structural abnormalities of the adjacent vital structures of the face like the ears, eyes, nose, teeth and brain. CL and P are intrinsically known for functional difficulties disturbing the feeding, breathing hearing, speech, vision Moreover, their negative impact on cosmetic. From the neural crest cells, nearly all soft tissue components and skeletal of the craniofacial area are exceptionally derived (12). Because eyes originate as an extension of the forebrain, malformations involving ocular structures invariably accompany those of the face and brain and vice versa. Instabilities in normal relocation of eye fields from the lateral to frontal areas of the embryo's face in the fifth to eighth weeks have been proposed as a probable cause of facial clefting and ocular hypotelorism and hypertelorism (13). Ophthalmologists have a exclusive role in the initial recognition of many ocular anomalies associated with disorder. In this hospital-based study, an effort was made to identify the ocular anomalies seen in patients with CL and P in north Indian population, so that early treatment can be provided to these patients for the same.

Common problems associated with cleft lip and palate

The cleft lip and palate causes many problems:

1. **Speech problem**

Patients with a cleft palate have speech problems which result from velopharyngeal dysfunction. Inability of soft palate to move upward to provide a contact with nasal cavity results in a passing of air through the nose instead of oral cavity. This condition is known as hypernasality speech.This case can be treated with a surgery to provide the velopharyngeal closure. Pharyngeal flap and sphincter pharyngoplasty are considered as the reliable surgeries for correcting the velopharyngeal deficiency in patients with a cleft lip and palate.

2. **Hearing problem and ear infection**

Otitis media is a condition where fluid is accumulated in the middle ear and results in ear infection. This is due to the abnormal action of Eustachian tube opening by two muscles which are tensor veli palatine and levator veli palatine. This leads to the lack of ventilation to the middle ear cavity and accumulation of fluid inside the middle ear. This condition is presented in the child with cleft palate in the first six months of life.

3. **Dental problems**

Dental problems involve abnormalities in the size and shape of the teeth, For example, the permanent lateral incisor shows abnormalities in size and shape in the side of cleft, abnormalities in the position of teeth, delay of eruption of permanent teeth and delay of formation of permanent teeth.

4. Feeding and nutritional problems

Feeding problems in babies with cleft lip and palate occur because babies are incapable of sucking either their mother's nipple or from a bottle. Therefore, this affects the weight and growth of the baby because the amount of milk or food is not enough for growth. There are a variety of methods that enable the baby to feed and gain a normal weight such as the use of disposable syringe, spoon and cup and prosthetic obturator device.

5. Cosmetic problems

Patients with cleft lip have cosmetic problems and also cause problems for production of labial sounds. Babies with cleft lip face difficulty when they try to make a contact between upper and lower lips.

Materials and Materials

This retrospective study was carried out at Department of otolaryngorhinology. Banas Medical college and research institute ,Banas Kantha, District and adjoining area of Gujrat and Rajasthan India, from Dec 2018- Dec 2020 were studied. Informed written as well as verbal consent was obtained from each patient and a parent or guardian. Using an alpha level of 0.05 and the survey sample size determination table created by Bartlett et al. (14), we determined that the minimum sample size required for this study was 370 participants. 370 consecutive syndromic and non-syndromic children with cleft lip and palate patients (206 boys and 164 girls). These variables sub-grouped into demographic data such as age, gender, birth weight, age of mother, cleft type, family history of cleft.

Statistical analysis

Data were presented in number and percentage. The Statistical Package for Social Sciences (SPSS), Version 16.0 (SPSS Inc. Chicago, USA) was used to analyse the data. Chi-square test were performed to determine the significance of the findings. Statistical significance was set at $P<0.05$.

Results

Total 370 children, 206 (55.7%) were male and 164 (44.3%) were female, out of which 104 cases (28.1%) had cleft lip only, distributed as following: 82 cases (22.2%) with unilateral cleft lip and 22 cases (5.9%) with bilateral cleft lip. One hundred twelve cases (30.3%) had cleft lip and palate, 90 cases (24.3%) of which were unilateral and 22 cases (5.9%) were bilateral. The highest number of cleft belonged to cleft palate comprising 154 cases (41.6%) of total patients (Table 1). There was significance difference found in between gender with the type of cleft with commoner involvement in males as compared to females.

Table 1: Association of the gender with the incidence of type of cleft

Cleft Type	Male n(%)	Female n(%)	Total Number	Chi Square, P value
Unilateral cleft lip	52(63.4)	30(36.6)	82	16.14, 0.003
Bilateral cleft lip	16(72.7)	6(27.3)	22	
Unilateral cleft lip and palate	54(60.0)	36(40.0)	90	
Bilateral cleft lip and palate	16(72.7)	6(27.3)	22	
Cleft Palate	68(44.2)	86(55.8)	154	
Total	**206**	**164**	**370**	

Figure 1: Percent distribution of associated risk factors

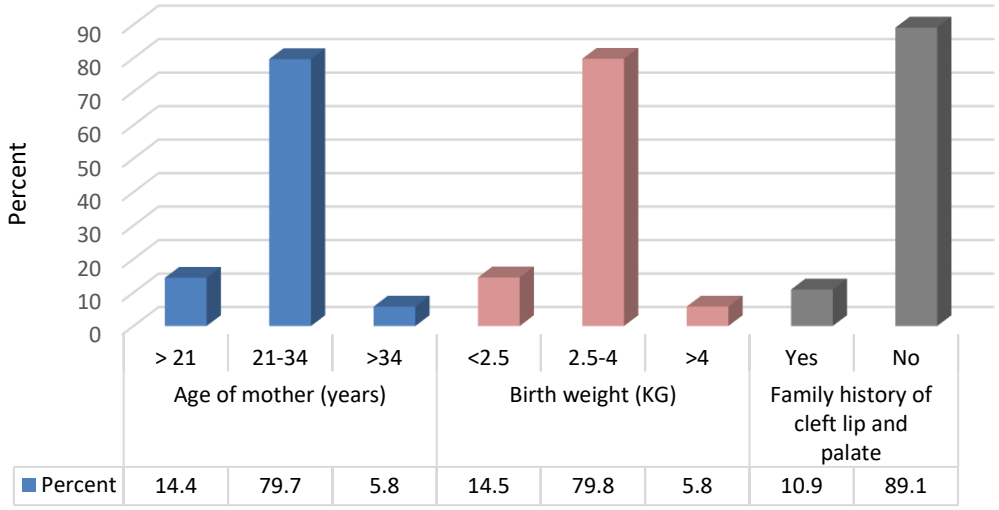

Figure 1 depicts that the percent distribution of associated risk factors. In which majority of mothers belongs to age group 21-34 years. Very few percent of mothers were above 34 years age. Most of children had weight at the time of birth were in between 2.5 kg to 4 .0 kg. Approximately eleven percent of the children had family history of cleft lip and palate, also seen among the associated factors for cleft lip and palate.

Discussion

Cleft lip & cleft palate is one of the commonest congenital facial anomaly in Indian population. Different types of cleft- associated anomalies occur with variable frequency. Loretz, Westmorel and, Richards (15) found that anomalies of the bones and joints are the most commonly found anomalies in conjunction with cleft lip and/or palate. According to Ivy (16) and McKeown and Record (17),anomalies of the nervous system occurred most frequently, while Kraus, Kitamura, and Ooe (18)reported that brachdactyly and syndactyly were the anomalies most frequently seen in cleft lip and/or palate fetuses. Other anomalies frequently accompanying cleft palate are: ocular anomalies; supernumerary fingers, toes, and teeth; malformed ears; clubbed hands or feet; tongue abnormalities; macroglossia and mandibular micrognathia(19).Some recent studies reported that females are bornwith anomalies in addition to cleft palate defects more frequently as compared to males. According to Lutz and Moor (20) 62% of the cleft associated defects were in females, while Ivy(16) found that slightly more than 50% of the associated anomalies were observed in cleft palate males. In our present study, cleft lip and palate was higher in male patients as compared to female patients. About 28% patients had cleft lip only, 22.2% with unilateral cleft lip and 5.9% with bilateral cleft lip. One third patients had cleft lip and palate. Moreover, Adesina et al (21) found that although the incidence of cleft anomaly was highest as isolated unilateral cleft lip, other associated anomalies (28%) was highest in patients with isolated cleft palate. Similar studies reported earlier by Stoll (22) and Natsume (23). Josef et al. however reported a higher incidence of associated anomalies among patients with combined cleft lip and palate (24). The findings of the current study also correspond with the findings of the study conducted by Figueiredo et al. (25) in relation to family history of cleft anomalies However, the present study showed a higher incidence of CL and P in babies born of mothers younger than 21 years old. Relatively consistent with the present study, Acuna-Gonzalez et al.(26) also found that the highest risk for CL and P was associated with variables related to family history background and family history of CL and P. The reported prevalence of associated anomalies varies widely across the literature; generally, a prevalence rate between 3% and 63% has been reported which is a reflection of varying data

collection (27). Among the ocular association various types of eyelid defects found were; coloboma, eury blepharon, ectropion, Ptosis and symblepharon. Developmentofeyelid and palatine process occur almost simultaneously in the intrauterine period and consequently a defect of the palate could lead to anocular defect and resultant coloboma of the lid (28).Ptosis of the eyelid which is a rare finding found in 2 patients in the present study. Ocular movement defects or squint is not uncommon in CL and P. In present study approximately 15% of patients had non-paralytic squint and this was attributed to the irregular shape and position of the orbital cavity and abnormal insertion of extra ocular muscles or both (29). Five of our patients included in the study had congenital dacryo cystitis. Because of entrapment of epidermal cells, four patients were found with limbal dermoids. Microphthalmos is a rare congenital condition where the size of the eyeball is smaller than normal. It is frequently associated with ocular anomalies which include epibulbar dermoid, anophthalmia and colobomas of upper eye lid. Other abnormal facial findings include hypoplasia, lateral cleft lip, preauriculartags/pits, deformity of middle ear and ossicles, deformity of pinna and external auditory meatusca using conductive hearing loss and hypoplasia of temporal bone. Blepharocheinodontic syndrome isa rare autosomal dominant condition of congenital facial clefting. Previous studies have found association between cleft lip and palate and eye lidretraction, euryblepharon, lagophthalmos. Thus, the wide variation in reports on prevalence of cleft deformity and associated ocular anomalies is found in previous literature which is in concurrence with our study. d (22). A larger sample based study of longer duration is to be planned in future to cover some other challenges other investigators have encountered are variation in the time of presentation of these cases after birth, level of knowledge of the investigators themselves and available technology, as well as variability in the clinical expressions of these associated anomalies (31).

Classification

Several authors classified the cleft lip and palate. Veau (1931) classified the clefts into four main groups:

- Clefts of soft palate.
- Clefts of hard palate.
- Unilateral clefts of the lip, alveolus and palate.
- Bilateral clefts of the lip, alveolus and palate

According to Koch *et al*.[32], (1995), Kernahan (1971) suggested a new classification of cleft lip and palate and it gives the shape of Y letter and includes:

- 1 and 4 represent the right and left side of the nasal floor, respectively.
- 2 and 5 represent the right and left side of the lip, respectively.
- 3 and 6 represent the right and left side of the paired alveolar segment, respectively.
- 7 represent the primary palate.
- 8 and 9 represent the secondary palate .

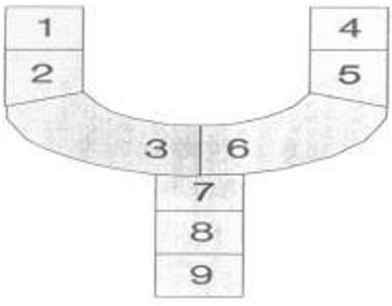

Figure 2: Kernahan classification of cleft lip and palate. (Koch *et al*., 1995).

Conclusion

The present study of Cleft lip and palate are the most common facial deformity and clefting may involve lip only, lip and palate and palate only. Environmental (such as smoking, alcohol, poor nutrition) and genetic factors (such as familial factors and chromosomes) are the main reasons of clefting in infants. Treatment of clefting involves a number of specialists who decide the best treatment plan depending on the site of defect and age of the infant

References

1. Srivastava D, Ghassemi A, Ghassemi M, Showkatbakhsh R, Jamilian A. Use of anterior maxillary distraction osteogenesis in two cleft lip and palate patients. Natl J Maxillofac Surg 2015;6:80-3.
2. Al Omari F, Al-Omari IK. Cleft lip and palate in Jordan: birth prevalence rate. Cleft palate croniaofac J. 2004;41:609- 12.
3. Jamilian A, Jamilian M, Darnahal A, Hamedi R, Mollaei M, Toopchi S. Hypodontia and supernumerary and impacted teeth in children with various types of clefts. Am J Orthod Dentofacial Orthop 2015;147:221-5.
4. Jamilian A, Nayeri F, Babayan A. Incidence of cleft lip and palate in Tehran. J Indian Soc Pedod Prev Dent 2007;25:174-6.
5. Wehby GL, Cassell CH. The impact of orofacial clefts on quality of life and healthcare use and costs. Oral Dis 2010;16:3-10.
6. Dixon MJ, Marazita ML, Beaty TH, Murray JC. Cleft lip and palate : understanding genetic and environmental influences. Nat Rev Genet. 2011;12:167-78.
7. Golalipour MJ, Kaviany N, Qorbani M, Mobasheri E. Maternal risk factors for oral clefts: a case-control study. Iran J otorhinolaryngol. 2012;24:187-92.
8. Murray JC. Gene/environment causes of cleft lip and/or palate. Clin Genet. 2002;61:248-56.
9. Gundlach KK, Maus C. Epidemiological studies on the frequency of clefts in Europe and word-wide. J Cranio - maxillofac Surg 2006;34 Suppl 2:1-2.
10. Romitti PA, Herring AM, Dennis LK, Wong-Gibbons DL. Meta-analysis: pesticides and orofacial clefts. Cleft craniofac J. 2007;44:358-65.
11. Cech I, Patnaik A, Burau KD, Smolensky MH. Spatial distribution of orofacial cleft defect births in Harris County, Texas, and radium in the public water supplies: a persistentbassociation? Texas Med 2008;104:56-63.
12. Sulik KK. Caniofacial development. In: Turvey TA, Vig KWL (eds)Facial clefts and cranio synostosis and principles and management. WB Saunders Company, Philadelphia, 1996. pp3-27
13. Nema HV, Singh VP, Nema N .Congenital anomalies of eye and adnexa. In: Agarwal S, Agarwal A (eds) Text book of ophthalmology; retina and vitreous in systemic diseases, 1st edn. Jaypee Brothers, New Delhi, 2002. pp 2939–2964
14. Bartlett JE, Kotrlik JW. Higgins CC, Organizational research: determining appropriate sample size in survey research, Information Technology, Learning, and Performance. 2001;19(1): 43–50
15. Loretz, W., Westmoreland, WW., Richmarps, LF., A study of cleft lip and cleft palate births in California. Amer. J. Pub. Health, 1961; 51, 873-877.
16. Ivy, R. H., Congenital anomalies: as recorded on birth certificates in the division of vital statistics of the Pennsylvania department of health, for the period of 1956 to 1960, inclusive. Plastic reconstr. Surg., 32, 361-367, 1961
17. McKrown, T, and Rrcorp, RG. Malformations in a population observed for five years after birth. Ciba Foundation Symposium on Congenital Malformations. Edited by G. E. W. Wolstenholme and C. M. O'Conner. Boston: Little, Brown and Co., 1960; 2-21.

18. Kraus, BS, Kiramura H, and Oor T., Malformations associated with cleft lip and palate in human embryos and fetuses. Amer. J. obst. Gyn., 1968; 86: 321-324.
19. Dorrancs, GW, The Operative Story of Cleft Palate. Philadelphia: W. B. Saunders Company, 1983.
20. Lutz KR., and Moor FB. A study of factors in the occurrence of cleft palate. J. speech hearing Dis., 1955;20:271-276.
21. Adesina OA, Efunkoya AA, Olaitan AC, Adesina OO. Associated anomalies in cleft lip and palate: analysis of 811 consecutive patients. South Sudan Medical Journal. 2017; 10(3):60-63
22. Stoll C, Alembik Y, Dott B, Roth MP. Epidemiological and genetic study in 207 cases of oral clefts in Alsace, north-eastern France. J Med Genet. 1991; 28(5):325–329.
23. Natsume N, Niimi T, Furukawa H, Kawai T Ogi N, Suzuki Y, et al. Survey of congenital anomalies associated with cleft lip and/or palate in 701,181 Japanese people. Oral Surg Oral Med Oral Radiol Endod. 2001; 91 (2):157–61
24. Josef M, Ola L, Catharina H, Margareta I. Associated Malformations in Infants with Cleft Lip and Palate: A Population-based Study. Pediatrics 1997; 100:180.
25. Figueiredo JC, Ly S, Magee KS, Ihenacho U, Baurley JW, Sanchez-Lara PA, et al. Parental risk factors for oral clefts among Central Africans, Southeast Asians, and Central Americans. Birth defect Res A clin Mol Teratol 2015;103:863-79.
26. Acuna-Gonzalez G, Medina-Solis CE, Maupome G,Corona Mde L. Family history and socioeconomic risk factors for non-syndromic cleft lip and palate: a matched case-control study in a less developed country. Biomedica .2011;31:381-91.
27. Sekhon, PS, Ethunandan M, Markus, A.F. KrishnanG, Bhasker Rao C., Congenital Anomalies Associated With Cleft Lip and Palate—An Analysis of 1623 Consecutive Patients Cleft Palate–Craniofacial Journal, 2011; 48 (4).
28. Larsen WJ . Development of head and neck. Text book of human embryology, 2nd edn. Churchill Livingston, New York, 1997; pp 347–374
29. Von Noorden GK, Campos EC . Etiology of heterphoria and heterotropia In: Binocular vision and ocular motility theory and management of stabismus, 6th edn. Mosby, St. Louis, London, 2000;pp 134–152
30. Kanski JJ (ed) . Disorders of the orbit. In: Clinical ophthalmology,4th edn. Butterworth Heinemann, Oxford, 1999; pp 552– 584
31. Wyszynski DF, Sarkozi A, Czeizel AE. Oral clefts with associated anomalies issues. Cleft methodological Palate, Craniofac J. 2006; 43:1–6
32. Koch H, Grzonka M, Koch J. Cleft malformation of lip, alveolus, hard and soft palate, and nose (LAHSN)- a critical view of the terminology, the diagnosis and gradation as a basis for documentation and therapy. British Journal of Oral and Maxillofacial Surgery, 1995; 33: 51-58.