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Research Article

A Study on Anatomical Variations in Non-Syndromic Clefts of Lip, Alveolus and Palate Belonging to the Eastern Part of India

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Abstract: Non-syndromic clefts of lip, alveolus and palate (CLAP) are purely physical deformities that are correctable surgically leading to normal facial growth provided the surgery is done in proper time and with the proper knowledge of anatomy of the cleft. This study in Eastern India gives an overview of anatomical variations of CLAP on which the surgical repair of the same depends. 250pediatric cases of non-syndromic CLAP of both sexes were studied over one year duration. They were classified into 3 subgroups namely cleft lip (CL), cleft lip and palate (CLP) and cleft palate alone (CP). The data was tabulated, statistically analyzed; correlation between the different subgroups and with existing datawas drawn.CLP(55.60%) is found to be the commonest type of CLAP deformity in studied population followed by CP(25.60%) and CL(18.80%). 181(72.40%) cases have complete clefts. Overall probability of complete cleft is highest in CLP subgroup followed by CP subgroup i.e. CLP are found to be most severe type of clefts. 149(59%) cases had unilateral clefts. Probability of unilateral clefts is higher in CL and CLP subgroups as compared to CP subgroup. It supports the fact that the etiology of CLP is different from that of CP. The present study findings may help in selecting the proper surgical procedure for correction of clefts that in turn depends upon the subtype, the extent and the laterality of the defect.
Keywords: clefts of lip alveolus and palate, cleft lip, cleft lip and palate, isolated cleft palate, complete cleft, incomplete cleft, unilateral clefts, bilateral clefts.

INTRODUCTION

Birth defects in general and particularly orofacial clefting is relatively common and significant problem for the individual patients born with these defects in terms of death or disability and for their families and society in general in terms of burden of care and health inequality. Despite significant advancement in treatment in high-income countries, problems in access to care and evidence base for cleft care still exist. While in the developing countries the consequences are lack of access to care and lack of infrastructure to help with quantification of the problem and consequently the ability to address it [1]. The Indian sub-continent remains one of the most populous areas of the world with an estimated population of 1.1 billion in India alone with estimated 24.5 million births per year. The birth prevalence of clefts is somewhere between 27,000 and 33,000 clefts per year [2]. According to The Smile Train records, every year nearly 35,000 new cleft patients are born in India alone and around 1 million are present with unrepaired clefts [3].

A CLAP child faces problem with feeding and talking. Other associated problems are ear infections, hearing loss, abnormal facial growth as well as dental and cosmetic abnormalities. Individuals with CLAP have low self-esteem and face difficulties during social interaction.

Epidemiology differentiates between cleft malformations as part of syndromes and the more common non-syndromic forms not associated with other physical or developmental deformities. Non-syndromic orofacial clefting is a polygenic, multi-factorial dic sorder so both genetic and environmental factors contribute to its aetiology. The environmental factors that contribute and the genes that predispose to the condition remain obscure despite decades of research [2].

CLAP abnormalities have been classified into several distinct subgroups but the typical distribution is [4, 5] Cleft lip alone 15%, Cleft lip, alveolus and palate 45%, Isolated cleft palate 40%. CLAP is a purely physical deformity that can be completely corrected surgically. There is displacement, deformation and mal/underdevelopment of the muscles and the skeleton of the face. Proper knowledge of the anatomy of the bones involved, alignment of muscles along with their blood and nerve supply is invaluable for restoring the normal facial growth and palatal functioning.

Surgery is planned taking into consideration the age and the weight of the patient. In United States, most surgeons follow the "rule of tens" and repair lip when the baby is 10weeks old, 10 pounds by weight and has a haemoglobin of 10. Palate repair is performed anywhere from 6 months onwards [6]. Patients taken in this study were operated at around the age of 3-4 months for cleft lip whereas surgery for cleft palate cases was done before the start of babbling speech i.e. between the age of 6-18 months. Weight of the patient at the time of surgery was atleast 5kgs for cleft lip and 8kgs for cleft palate repair. The surgical procedure selected depends upon the subtype, the extent and the laterality of a cleft, which is the basis of the present work. Here the CLAP is studied taking into consideration the sex of the patient, the type of cleft, its extent, laterality and sidedness.

MATERIALS AND METHODS

It is an observational/ correlational study where data collected by clinical study is analyzed statistically and correlation between the 3 subgroups and with the existing data is drawn.250 cases of non-syndromic CLAP of pediatric age group and of both sexes attending the OPD of the Dept. of pediatric surgery, Institute of Child Health (funded for surgery by Smile Train Express) where most of the patients from different parts of the West Bengal as well as its neighboring states of Bihar, Jharkhand and Orissa come for the treatment were studied over a period of one year. After getting ethical clearance from the authorities and written informed consent from the parents, the patients underwent history taking, physical examination and investigations. Children below 12 years of age and both sexes residing in West Bengal and those coming for treatment from its neighboring states as stated above were included. Cases from families who had migrated from their original place of residence within 3 years before the birth of the affected child were excluded. Those presented with clefts as part of other congenital syndrome or deemed unfit for surgery for some reasons were excluded. History was taken from either of the parents regarding the age, sex of the patient, and the associated problems faced by him/her. Physical examination was done to know the type of the cleft; its site, extent and sidedness; general and systemic examinations as well as investigations were done to exclude associated abnormalities (syndromic variety).

Data collected have been summarized by counts and percentages (Table 1). Categorical variables have been compared between subgroups by Chi-square test or Fisher's exact test as appropriate; $p< 0.05$ has been considered statistically significant. Statistica version 6 [Tulsa, Oklahoma: Stat- Soft Inc., 2001] software was used for analysis.

RESULTS

Of the studied population of 250 cases, CLP subgroup is found to have the highest frequency (55.6%) followed by CP and CL subgroups. More number of males 127 (50.80%) are found to be affected with CLAP than females 123 (49.20%).But no statistically significant sex difference is found between the 3 subgroups.181(72.40%) cases have a complete cleft. The probability of complete cleft is highest in CLP cases followed by CP cases. In 149(59.6%) cases clefts are unilateral. The probability of unilateral clefts is higher in CLP and CL subgroups as compared to CP subgroup. In both the cases p value < 0.001. But no statistically significant relation is found between CLP and CL subgroups. Of 149 unilateral clefts, 89 are left-sided and 60 are right-sided. Statistical comparison of the distribution between the subgroups shows that the probability of left-sided clefts is comparable between the 3 subgroups.

Table 1: Distribution of the study population within the subgroups

	Total cases	Males	Females	Complete clefts	Incomplete clefts	Unilateral clefts	Bilateral clefts	Left Sided clefts	Right Sided clefts
CL	47 (18.80%)	22 (46.81%)	25 (53.19%)	17 (36.17%)	30 (63.83%)	42 (89.36%)	5 (10.64%)	26 (61.90%)	16 (38.10%)
CLP	139 (55.60%)	78 (56.12%)	61 (43.88%)	123 (88.49%)	16 (11.51%)	100 (71.94%)	39 (28.06%)	56 (56%)	44 (44%)
CP	64 (25.60%)	27 (42.19%)	37 (57.81%)	41 (64.06%)	23 (35.94%)	7 (10.94%)	57 (89.06%)	7 (100%)	0 (0%)

Table 2: Comparison between the present study and the available data

	Present study (250 cases)	**Andhra Pradesh [9]**	**Madhya Pradesh [18] (839 cases)**	**CMC&H Vellore [19] (750 cases)**	**Fogh Anderson study[15]**	**Southern Thailand [13] (153 cases)**	**Ireland [20] (616 cases)**
CL	18.8%	33%	_	28.8%	25%	23.5%	26%
CLP	55.6%	64%	_	56.9%	50%	55.6%	35%
CP	25.60%	2%	2%	14.3%	25%	20.9%	39%
Males	50.8%	_	_	55.87%	_	_	_
Females	49.2%	_	_	44.13%	_	_	_
Complete clefts*	72.40%	_	_	_	_	_	_
Unilateral clefts	59.6%	79%	83.3%	_	_	_	_
Left-sided clefts	59.7%	64%	67.3%	_	64%	_	_

*Supporting Data Not Available

DISCUSSION

Although national epidemiological data of cleft lip and palate is not available, many studies from different parts of India have shown a wide variation in its incidence, from 0.25-2.29 per 1000 live births. The magnitude of the problem in India is enormous considering its large population and high birth rate. Cleft lip and palate care gets further compromised because of poor socioeconomic status [3], low literacy, lack of awareness and non-availability of health care in this areas [7].

Incidence of oral-facial clefting shows ethnic variation. It is generally thought that populations of Asian or Native North American descent have the highest incidence, with Caucasian populations having intermediate incidence and African populations having the lowest incidence [8, 9]. The frequency of clefts varies from country to country; it is different even between the different states of our country (India) as is evident from the data available [9-15, 18-20]. (Table 2). A review of studies for incidence of cleft lip and palate shows that there is no particular trend in different parts of the world [26]. Most of the studies show highest frequency of CLP, as is the finding of our study. This diversity in the findings can be explained by the fact that there is wide variation in environmental conditions, which have a great impact on the causation of clefts of different types. India being a vast country with varied climate and socioeconomic conditions also show diverse results.

Most studies [9, 15-19] report a male predominance in the sex ratio in cleft lip and palate patients and a female predominance in patients with isolated cleft palate defects. But results of our study are not found to be statistically significant (Table 2). This may be due to the fact that only those cases are taken, which have come for the treatment in the OPD. Parents of female child with these defects have to face difficulties while looking for a suitable groom for her, which might be the cause of more girls being brought for the treatment, which is being provided free of cost.

Complete clefts predominate in our study (72.40%) probability of which is highest in CLP cases followed by CP cases. This corroborates with the results of some other studies that have provided the ratio but not the percentage [19, 20]. This also shows that the CLP are more severe type of clefts than the CP and strengthens the fact that there is a difference between the cleft aetiology between CLP and CP cases. Fogh-Andersen and Fraser have noted that the clefts involving the anterior structures (lip and primary palate) (CL/CLP) could be separated on both genetic and embryologic grounds from those involving only the secondary palate (CP) [21, 22]. Predominance of complete clefts in the study sample maybe due to the fact that these are the more severe deformities leading to functional difficulties hence forcing the parents to bring their ward for treatment. We have not found any association between the severity of the cleft i.e. complete or incomplete and the sex or laterality as is found in a study done in 4 different regions in UK [23].

We found a predominance of unilateral clefts (59.6%) which corroborates with the results of most studies that give a ratio between unilateral and bilateral cleft lips to be predominantly favoring unilateral clefts [9, 16, 18, 24, 25] (Table 2). No explanation for the same could be ascertained. But we have found that the probability of unilateral clefts is higher in CLP and CL subgroups as compared to CP subgroup, which again supports the fact that etiology for the causation of the clefts i.e. CL/CPL and CP is different.

It is also widely accepted that left-sided unilateral clefts are more common than right-sided unilateral cleft lips [9, 11, 15], which is supported by this study (Table 2). But the statistical comparison of the distribution between the subgroups shows that the probability of left-sided clefts is comparable between the 3 subgroups.

The biggest limitation of this study lies in the fact that the data collected here isthe hospital-based data where the Berkesonian bias may creep in. But still in case of cleft defects for which routine data from the community is sparsely available in India particularly in its Eastern part, depending upon hospital statistics is perhaps the only pragmatic approach at present especially if that hospital has a dedicated project running for cleft defects (Smile Train Express).

CONCLUSION

Non-syndromic clefts are multifactorial in origin with both genetics and environmental factors interplaying for their causation. A large number of studies all over the world and in India have failed to ascertain the definitive cause of the same, which is one of the main causes of failure of controlling their occurrence. With the passage of time new factors are being identified related to the causation. New epidemiological factors as well as mutations in genes are further leading to increase in the incidence of the same. Hence to tackle this problemthe solution lies not only in primary prevention but in early detection of cases and providing adequate treatment facilities (multidisciplinary) to those affected since CLAP is purely a physical deformity that can be completely corrected surgically leading to normal facial growth with good aesthetic and functional results. The affected child needs to be treated at the proper time with the proper knowledge of anatomy of the defect. For the provision of the treatment facility the motivation of the parents, caregivers, society, government and other authorities andmedical fraternity is highly recommended. The surgical procedure selected depends upon the subtype, the extent, the laterality and the sidedness of a cleft. The finding of the present study may facilitate in that process of surgical decision-making.

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REFERENCES

1. Mossey PA, Modell B; Epidemiology of oral clefts 2012: an international perspective. Front Oral Biol., 2012; 16: 1-18
2. Peter M, Julian L; Addressing the challenges of cleft lip and palate research in India. Indian J Plast Surg., 2009; 42(Suppl): S9–S18.
3. Aggarwal P, Banerjee B, Gupta P, Datta AK; Effect of socioeconomic status on clefts of lip, alveolus and palate in population belonging to the eastern part of India. National Journal of Medical Research, 2014; 4(3): 222-224
4. Williams NS, Bulstrode CJK, O'Connell PR; Bailey & Love's Short Practice of surgery. 25th edition, London: Edward Ernold (Publishers) Ltd., 2008: 661.
5. Karoon A; Classification of cleft lip and palate: An Indian perspective. Journal of Cleft Lip Palate and Craniofacial Anomalies, 2014; 1(2): 78-84.
6. Prater ME; Cleft lip, alveolus and palate, Dept. of Otolaryngology, UTMB, Grand Rounds, May 2000.
7. Bhateja A et al,; Evaluation of surgical protocol and treatment need of operated unilateral cleft lip and palate patients in Delhi. J Indian Soc Pedo Prev Dent., 2001; 19(1): 10-17.
8. Gorlin RJ, Cohen MM, Hennekam RC; Monographs on medical genetics syndromes of the head and neck. Minneapolis, USA: Oxford University Press, 2001.
9. Reddy SG1, Reddy RR, Bronkhorst EM, Prasad R, Ettema AM, Sailer HF *et al.*; Incidence of cleft Lip and palate in the state of Andhra Pradesh, South India. Indian Journal of Plastic Surgery, 2010; 43(2): 184-189
10. Drillien Cecil M, Ingram TTS, Wilkinson EM; The causes and natural history of Cleft lip and Palate. British Journal of Surgery,1966; 53(8): 744.
11. Iregbulem LM; The incidence of cleft lip and palate in Nigeria. Cleft Palate J., 1982; 19: 201-205.
12. Croen LA, Shaw GM, Wasserman CR, Tolarovα MM; Racial and ethnic variations in the prevalence of orofacial clefts in California 1983-1992. Am J Med Genet., 1998; 79: 42-47.
13. Jaruratanasirikul S; Cleft lip and/or palate: 10 years experience at a pediatric cleft center in Southern Thailand. Cleft Palate Craniofac J., 2008; 45(6): 597-602.
14. Sridhar K; A community-based survey of visible congenital anomalies in rural Tamil Nadu. Indian Journal of Plastic Surgery, 2009; 42(3): 184-191.
15. Fogh-Andersen P; Epidemiology and etiology of clefts. Birth Defects Orig Artic Ser., 1971; 7(7): 50-53.
16. Abyholm FE; Cleft lip and palate in Norway, I. Registration, incidence and early mortality of infants with cleft lip and palate. Scand J Plast Reconstr Surg., 1978; 12(1): 29-34.
17. Wu Y, Zeng M, Xu C, Liang J, Wang Y, Miao L *et al.*; Analyses of the prevalences or neural tube defects and cleft lip and palate in China from 1988 to 1991. Hua Xi Yi Ke Da Xue Xue Bao, 1995; 26(2): 215-219.

18. Bhattacharya S, Rai A, Shrivastava P; Cleft data from surgical camps on rails: A doorstep health care delivery. Journal of Cleft Lip Palate and Craniofacial Anomalies, 2014; 1(1): 38-42.
19. Theogaraj SD, Joseph LBM, Mani M; Statistical analysis of 750 cleft lip and palate patients. Indian Journal of Plastic Surgery, 2007; 40(1): 70-74.
20. Milerad, J. et al. (1997) Duarte, R., Leal, M.J. (1999). Associated malformations in infants with cleft lip and palate: a prospective, population-based study. Pediatrics, 1997; 100(2 Pt 1): 180-186.
21. Fogh-Andersen P; Inheritance of harelip and cleft palate. Copenhagen: Munksgaard, 1942.
22. Fraser FC; Thoughts on the etiology of clefts of the palate and lip. Acta Genet Stat Med., 1955; 5: 358–369.
23. Carroll K, Mossey PA; anatomical variations in clefts of the lip with or without cleft palate. Hindawi Publishing Corporation, Plastic Surgery International, 2012; 6 pages. Available from http://www.hindawi.com/journals/psi/2012/542078/
24. Womersley J, Stone DH; Epidemiology of facial clefts. Arch Dis Child., 1987; 62: 717-720.
25. Jensen BL, Kreiborg S, Dahl E, Fogh-Andersen P; Cleft lip and palate in Denmark, 1976-1981: Epidemiological, variability, and early somatic development. Cleft Palate J., 1988; 25: 258-269.
26. Global Strategies to Reduce the Health Care Burden of Craniofacial Anomalies. Report of WHO meetings on International Collaborative Research on Craniofacial Anomalies. Geneva, Switzerland, 2000.