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Assessment of Prevalence Pattern of Orofacial Clefting and Etiological Risk Factors in Children with Cleft Lip and/ or Palate.

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ABSTRACT

Background: Aim: To assess prevalence pattern of cleft lip and palate and to assess the etiological risk factors like history of consanguinity, familial tendency and socio – economic status in children with cleft lip and/or palate. **Methods**: Case records (N=821) of patients with cleft lip and palate were accessed to collect the data. The following information was collected: Age, Gender, Education, Occupation and Income of the parent, History of consanguinity, Familial tendency and Chief complaint/s. The SPSS software was used for the statistical analysis to do the chi-square test and significance level adopted was 5% ($P < 0.05$). **Results**: Orofacial clefting was more common in males and laterality of cleft lip occurred more on the left side. The familial tendency was present only in 3% cases. The history of consanguinity was present in 35.7% cases. The socio – economic deprivation was prominent in cleft cases and values depicted statistical significance. **Conclusion**: Higher risk of orofacial clefting was seen in consanguineous relations and in families with socio – economic deprivation.

Keywords: cleft lip, cleft palate, consanguinity, India.

INTRODUCTION

Craniofacial anomalies encompass a significant component of morbid human birth defects. Craniofacial anomalies like Cleft Lip Palate (CLP), Cleft Lip alone (CL) and Cleft Palate alone (CP) affect about 1.09 for every 1000 live births in India.[1] It is estimated that there are about 10 lakh cases of untreated oral clefts in India.[2] Globally, every two minutes a cleft child is born.[3] In 2008, the World Health Organisation (WHO) has documented that non-communicable diseases, including birth defects cause significant infant mortality and childhood morbidity and they have also involved cleft lip and palate in their Global Burden of Disease (GBD) initiative.[4]

Cleft lip and palate may give rise to many complications in affected individuals including feeding problems and recurrent infections like otitis media, cholesteatoma,[5] maxillary sinusitis and bronchopneumonia.[6,7] The parents of such children usually more concerned about the child's appearance due to craniofacial anomaly rather than the complications that may arise from the clefts in those patients. Children born with cleft lip and palate and no other known malformations appear to have an increased risk of mortality not only in the first year of life but throughout childhood and adulthood.[7]

Consanguinity is the property of being from the same kinship as another person. It has been an age old ritual in South India and in many parts of the world to have consanguineous relationships.[8] Offspring of consanguineous unions are at increased risk of 1.7–2.8% above the background risk for birth defects for first cousin unions (third-degree relatives).[9] There was substantial correlation of children with clefts being born to parents who shared a consanguineous relationship.[1]

In addition to the psychological, societal and functional sequelae, cleft lip and palate imposes a financial burden for families and health care systems.[10] Higashi H et al conducted a study which showed that 59% burden of cleft lip and palate is amenable by surgery if surgical programmes cover the entire population with access to quality care.[11]

MATERIALS AND METHODS

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The present study included a retrospective analysis of patient records for a 24-month period between January 2011 and December 2012, treated in the Craniofacial unit at SDM College of Dental Sciences and Hospital, Sattur, Dharwad, India. Total of 821 patient records who were diagnosed with cleft lip and/or palate were accessed. The following information was collected from the patient records: Chief complaint, Age, Gender, (Education, Occupation and Income) of the parent. History of consanguinity, Familial tendency were recorded under medical history.

The inclusion criteria for the patients who were diagnosed with cleft lip and/or palate has been mentioned below:

Patients between the age ranges from birth to 12 years

Patients who were not operated for the clefts/not on any treatment/medication

Patients not associated with any syndromes

Cleft patients not suffering from any chronic systemic illness like immune compromising conditions, bleeding/clotting disorders, leukemia, juvenile diabetes etc.

The data of cleft type was documented according to the Kernahan and Stark's classification.[12] For convenience of comparison and statistical analysis the subjects were grouped into four groups namely Neonate, Infancy, Toddler and Childhood.[13] The cleft types were also further classified into three groups which are Cleft lip and Alveolus, Cleft Palate, Cleft lip and Palate.

The data was collected and added into Microsoft Excel sheets. In the process, all the patient records were coded and patient's identity was concealed. The waiver of consent was taken from the Institutional Ethics Committee before conducting the research.

The socioeconomic status was compared with the national averages using the revised Kuppuswamy's scale.[14] The SPSS software was used for the statistical analysis to do the chi-square test and significance level adopted was 5% ($P < 0.05$).

RESULTS

Out of the total 821 cases, 30% children were presented with cleft lip, 40% were cleft palate and 29% were cleft lip and palate.

The prevalence of the cleft lip and/or palate was seen to be higher in males (1.34:1) but there were no statistically significant values to correlate the pattern [Table I]. Our study showed that the left side was more involved (59.2%) when compared to the right side (40.8%) in the cleft lip cases with/without palate. Out of 821 cases 40% belonged to the Middle class and 55.3% belonged to the Upper Lower class [Table II] and the parameter is statistically significant (p=0.001). Familial prevalence was seen in 3% of the total cases (17 males and 8 females) and the values were statistically not significant [Table III]. The history of consanguinity was seen to be a risk factor in 35.7% of the total cases [Table IV]. Out of the 821 cases, 97% of the parents' chief complaint was about the cleft deformity (appearance).

Table 1: Prevalence pattern of Cleft groups

Gender		Cleft type			Total
		Cleft Lip and Alveolus	Cleft Palate	Cleft Lip, Alveolus and Palate	
FEMALE	Count	93	156	101	350
	% of Total	11.3 %	19.0 %	12.3%	42.6%
MALE	Count	153	177	141	471
	% of Total	18.6 %	21.6 %	17.2%	57.4%
Total	Count	246	333	242	821
	% of Total	30.0 %	40.6 %	29.5%	100.0 %

Pearson Chi-Square	Value	Degree of Freedom	P-value
	4.842	2	.089

Table 2: Comparison of Socio – Economic Status in different Cleft groups

Cleft type		Socio - Economic Status					
		Upper	Upper Middle	Middle	Upper Lower	Lower	Total
Cleft Lip and Alveolus	Count	0	6	102	132	6	246
	% of Total	0.0%	0.7 %	12.4 %	16.1 %	0.7 %	30.0%
Cleft Palate	Count	2	12	147	172	0	333
	% of Total	0.2%	1.5 %	17.9 %	21.0 %	0.0 %	40.6%
Cleft Lip, Alveolus and Palate	Count	0	13	79	150	0	242
	% of Total	0.0%	1.6 %	9.6%	18.3 %	0.0 %	29.5%
Total	Count	2	31	328	454	6	821
	% of Total	0.2%	3.8 %	40.0 %	55.3 %	0.7 %	100 %

Pearson Chi – Square	Value	Degree of Freedom	P-value
	27.495	8	.001

Table 3: Comparison of Familial Tendency in cleft groups

Cleft type		Familial Tendency		Total
		ABSENT	PRESENT	
Cleft Lip and Alveolus	Count	240	6	246
	% of Total	29.2%	0.7%	30.0%
Cleft Palate	Count	320	13	333
	% of Total	39.0%	1.6%	40.6%
Cleft Lip, Alveolus and Palate	Count	236	6	242
	% of Total	28.7%	0.7%	29.5%
Total	Count	796	25	821
	% of Total	97.0%	3.0%	100.0%
Pearson Chi – Square	Value	Degree of Freedom	P-value	
	1.400	2	.497	

Table 4: Comparison of History of Consanguinity in cleft groups

Cleft Type		History Of Consanguinity		Total
		Absent	Present	
Cleft Lip and Alveolus	Count	158	88	246
	% of Total	19.2%	10.7%	30.0%
Cleft Palate	Count	217	116	333
	% of Total	26.4%	14.1%	40.6%
Cleft Lip, Alveolus and Palate	Count	153	89	242
	% of Total	18.6%	10.8%	29.5%
Total	Count	528	293	821
	% of Total	64.3%	35.7%	100.0%
Pearson Chi – Square	Value	Degree of Freedom	P-value	
	.231	2	.891	

DISCUSSION & CONCLUSION

The prevalence of the cleft lip and/or palate was seen to be higher in males (1.34:1). This finding was comparable to study conducted by Mathews J L which showed increased prevalence of clefts in males (1.75:1).[15] Familial prevalence was seen in 3% of the total cases (17 males and 8 females) and the values were statistically not significant. Fraser FC conducted a study which revealed the frequency of clefts among the relatives to be 3.2%,[16] which is well within the observed range. Blanco R calculated the genetic risk score for the occurrence of clefts and found that Genetic Risk Scores showed no significant results when comparing cases and controls.[17] Our study showed that the left side of the face was more involved (59.2%) when compared to the right side (40.8%) in the cleft lip cases with/without palate. This finding was concomitant with the values of study conducted by Kesande et al which showed 60% involvement of the cleft lip with/without palate on the left side.[18] Our study showed that around 30.5% of the patients had a history of nasal regurgitation, out of which only 3% of the parents were aware of the nasal regurgitation. Indicative of lack of awareness of parents towards nasal regurgitation and its possible collateral complications of orofacial clefts. Nutritional intake is related to socioeconomic status and the increased occurrence of clefts among the offspring of less educated women highlights the importance of maternal nutritional status on reproductive outcome.[19] Our study showed that majority of the cases belonged to the middle and the upper lower class. Low socio-economic status was seen to be a risk factor for the occurrence of clefts.[20] Risk factors which could mediate the impact of socioeconomic status on the occurrence of congenital anomalies include nutritional factors, lifestyle, environmental and work-related exposures, access to and use of health amenities, parity and maternal age, and ethnic origin.[21] The data of socio-economic status should help in fund planning of the programmes conducted by organizations which provide free cleft surgeries.

The role of consanguineous relationship in clefts is inconclusive but it is reported to play a major role in the occurrence of congenital malformations.[22] The history of consanguinity was seen to be a risk factor in 35.7% of the total cases. Ravichandran et al,[23] reported the history of consanguinity to be a risk factor in 56.8% of families. The history of consanguineous relationships was affecting males (20.7%) more than females (15%). Bennett R et al,[24] reported the consanguineous unions to be at increased risk for birth defects in their offspring and attributed it to autosomal recessive mutations inherited from a common ancestor. The awareness of complications arising from consanguineous relationships should be conveyed through mass media.

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