

PROPORTIONS OF ORAL CLEFTS IN PATIENTS VISITING THE CLEFT CENTER OF CHILDREN'S HOSPITAL AND INSTITUTE OF CHILD HEALTH LAHORE: A FIVE YEAR STUDY (2000 - 2005)

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ABSTRACT

The objective of the study was to identify proportions of oral clefts in patients visiting Children's Hospital / Institute of Child Health, Lahore during five year period from 24-01-2000 to 24-01-2005.

This observational descriptive study included 128 subjects comprising 72 males and 56 females. Age of these patients ranged from one day old to 13 years. The method used for this study was Kernahan modified double 'Y' with numeric coding system. A Total of 24 combinations were used to describe all types of clefts. The data was analyzed in SPSS 10 statistical package.

Out of the whole sample size an increased number of male subjects (72) were examined as compared to female subjects (56). The most common finding of this study was complete bilateral clefts affecting 54 patients (42.2%). The next common type was complete unilateral cleft of the lip and palate, occurring in 27 patients (21%). Among these, clefts involving left side were more common and were seen in 19 patients (14.8%). An unusual finding of this study was complete unilateral and partial clefts of the opposite side.

Increased number of complete bilateral cleft of the lip and palate was because of referrals to the CH & ICH from various surrounding and remote localities for the tertiary level treatment of this particular deformity. The unusual type of the cleft was identified with Kernahan's modified double stem of the symbolic 'Y'

Key Words: *Proportions, incidence, Kernahan's double Bilateral Clefts, complete unilateral cleft and partial opposite cleft.*

INTRODUCTION

Cleft of the lip and palate are the most common congenital facial deformities. These clefts may vary from minor notching of lip or bifid uvula to complete unilateral or bilateral cleft of the lip and palate. Incidence of the cleft lip and palate deformities in various countries were tabulated by Fogh Anderson¹⁻². Bellis &


Wohlgemuth³ conducted a study over twenty years period in the south east of Scotland (Edinburgh cleft units catchments areas) on 502 patients (291 Males, 211 Females). The incidence of clefts was reported as 1.4 per 1000 live births. Blanco—D avila⁴ conducted a 10 years study in the north east of Mexico on 206 patients and concluded that the incidence of cleft lip and palate in the cited hospital was 1.1 per thousand births. As per

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text book report from Singh G⁵ the incidence of cleft lip and palate varies widely among different races, being least in the Blacks (0.4) and highest in the Mongoloids (Afghans 4.9). The incidence described for Caucasians is 1, Japanese 1.7, American Indians 3.6 and Indians 1.7 per 1000 births.

Clefts of the lip and palate are reported to be more common in males than females^{1,2,3,4,5}. Among the etiological factors, about 40% cases of cleft lip and palate are hereditary in nature according to Fogh and Anderson. Main mode of transmission maybe either by single mutant gene producing a large effect, or by a number of genes (polygenic inheritance), each producing a small effect. Multifactorial inheritance theory implies interaction of contributory risk genes with one another and the environment resulting in a defect in the developing fetus. The theory explains transmission of isolated cleft of the palate. Amongst the environmental factors are drugs like corticosteroids, thalidomide, radiations, stress and infections like rubella virus etc. Embryologically, failure of adequate mesodermal migration or proliferation can result in complete or incomplete, unilateral or bilateral cleft of the lip and primary palate. Clefts of the secondary palate occur due to lack of elevation of the palatal shelves at the critical time or failure to contact or inadequate epithelial breakdown due to deficient mesoderm.

Classification of oral clefts may be embryological, based on incisive foramen separating the junction between primary and secondary palate or may be morphological based on location of the cleft relative to the alveolar process. A number of classifications have been proposed. The first recognized classification was designed by Davis and Richie⁶. This classification was morphological in origin and it divided all clefts into three groups. Veau classified clefts into four broad groups on morphological basis. Fogh Anderson devised embryological classification of these clefts, which has a major group for harelip and divided clefts into three groups. Kernahan and Stark⁸ classified clefts on embryological basis and divided Clefts into three groups. Later on, Kernahan⁹ devised a symbolic classification to overcome confusion regarding descriptive items or numbers used in classifications. Cleft lip and palate deformity was represented as a  The incisive foramen can be represented symbolically at the junction of the limb by a small circle. Each right and left limb of the Y was divided into three portions representing the lip, alveolus and the area between alveolus and incisive foramen respectively. The stem of the Y was also divided into three portions. In the striped Y system,

complete cleft was represented by shading black, partial cleft with dots and secondary palate clefts with vertical lines.

Kernahan's Y was recently modified into double Y that describes all clefts of the primary and secondary palates comprising a total of 24 combinations¹⁰. According to this classification, clefts anterior to incisive foramen are classified as complete, incomplete, right—left clefts involving nose, lip alveolus and primary palate and Clefts posterior to the incisive foramen are classified as complete, incomplete, right-left clefts of the secondary palate (Fig.1). This classification allows easy diagrammatic visualization and record keeping in a computer program. Any special patient and research programs are classified under special code for easy recall.

It was an observational descriptive study, and the objective was to identify various types of oral clefts in patients visiting the children hospital/Institute of child health from 24-1-2000 to 24-01-2005.

MATERIAL AND METHODS

The subjects included in this study comprised 128 patients (72 Males & 56 Females). Age range varied from 1 day old to 13 years of age.

The method used for this study was Kernahan's Modified Double 'Y' that allowed easy diagrammatic visualization and record keeping in computer program (Figure 1). A total of 24 combinations were used to describe all cleft types. Clinical examination of the patients was done along the chair side to see location of the clefts. On identification, these clefts were related to the numeric codes of the Kernahan's symbolic double Y to get ultimate classification.

This data was analyzed in SPSS 10 statistical package to calculate frequencies and percentages of cleft type and sex cross tabulation. The analysis also included the frequencies and percentages of the areas to which the subjects belonged.

RESULTS

In this study, an increased number of male patients were observed; 72 males compared to 56 females (Table 1). Out of the whole sample size, 54 patients (Males 35, Females 19) were classified as complete bilateral cleft of the lip and palate with the percentage of 42.2% (Table 2). Complete unilateral cleft of the left side was found in 19 patients (Males 12, Females 7) 14.8%. Cleft involving complete unilateral right side was identified in 8 patients (Males 7, Female 1) 6.3%.

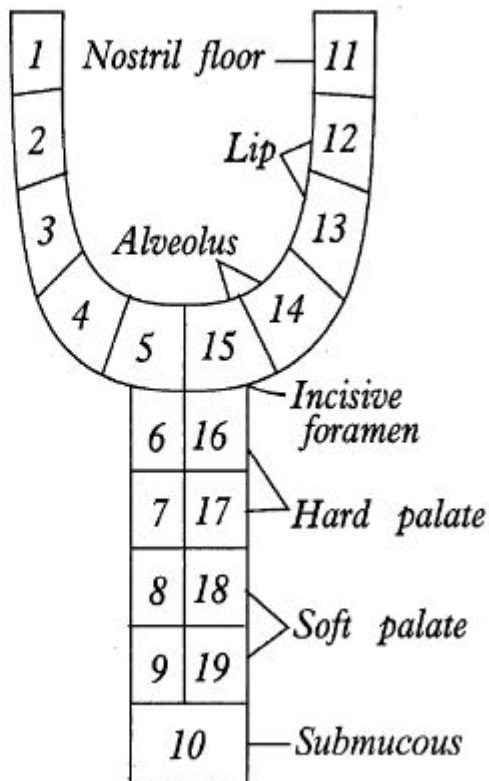


Fig 1: Kernahan's MOdified Double 'Y'

TABLE 1: KERNAHAN'S MODIFIED DOUBLE 'Y' CLASSIFICATION SEX CROSS TABULATION Count

		Sex		Total
		Male	Female	
Kernahan's modified double "Y" classification	bifid uvula 10	1		1
	median soft palate 8, 9, 18, 19, 10	3	7	10
	bilateral comp cleft lip & palate 1-10-11-19	35	19	54
	L complete unilateral 11, 12, 13, 14, 15, 16, 17, 18, 19, 10	12	7	19
	R complete unilateral 1, 2, 3, 4, 5, 6, 7, 8, 9, 10	7	1	8
	R cleft nose, lip, alveolus, primary palate 1, 2, 3, 4, 5, 6	2		2
	R cleft nose, lip, alveolus 1, 2, 3, 4		1	1
	L cleft nose, lip, alveolus 11, 12, 13, 14, 15 anterior to incisive foramen	1		1
	1, 2, 3, 4, 5, 15, 14, 13, 12, 11	1	2	3
	L alveolus 13, 14		1	1
	sec & soft palate 6, 7, 8, 9, 10, 16, 17, 18, 19	4	8	12
	L comp+R sec & soft palate 11-19, 7, 8, 9, 10	3	7	10
	R comp+L sec & soft palate 1-10, 17, 18, 19	5	1	6
	Total	72	58	128

TABLE 2: KERNAHAN'S MODIFIED DOUBLE 'Y' CLASSIFICATION

		Frequ- ency	Percent	Valid percent	Cumulative percent
Valid	bifid uvula 10	1	.8	.8	.8
	median soft palate 8, 9, 18, 19, 10	10	7.8	7.8	8.6
	bilateral comp cleft lip & palate 1-10-11-19	54	42.2	42.2	50.8
	L complete unilateral 11, 12, 13, 14, 15, 16, 17, 18, 19, 10	19	14.8	14.8	65.6
	R complete unilateral 1, 2, 3, 4, 5, 6, 7, 8, 9, 10	8	6.3	6.3	71.9
	R cleft nose, lip, alveolus, primary palate 1, 2, 3, 4, 5, 6	2	1.6	1.6	73.4
	R cleft nose, lip, alveolus 1, 2, 3, 4	1	.8	.8	74.2
	L cleft nose, lip, alveolus 11, 12, 13, 14, 15 anterior to incisive foramen	3	2.3	2.3	77.3
	1, 2, 3, 4, 5, 15, 14, 13, 12, 11				
	L alveolus 13, 14	1	.8	.8	78.1
	sec & soft palate 6, 7, 8, 9, 10, 16, 17, 18, 19	12	9.4	9.4	87.5
	L comp+R sec & soft palate 11-19, 7, 8, 9, 10	10	7.8	7.8	95.3
	R comp+L sec & soft palate 1-10, 17, 18, 19	6	4.7	4.7	100.0
	Total	128	100.0	100.0	

TABLE 3: TABULAR REPRESENTATION OF THE LOCALITIES OF CLEFT PATIENTS

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Lahore	67	52.3	53.6	53.6
	Sahiwal	7	5.5	5.6	59.2
	Gujranwala	10	7.8	8.0	67.2
	Jhang	3	2.3	2.4	69.6
	Muzaffarabad	1	.8	.8	70.4
	Sialkot	4	3.1	3.2	73.6
	Bahawalnagar	2	1.6	1.6	75.2
	Sheikhupura	5	3.9	4.0	79.2
	Kotli	1	.8	.8	80.0
	Hafizabad	2	1.6	1.6	81.6
	Patoki	2	1.6	1.6	83.2
	Rawalpindi	1	.8	.8	84.0
	Multan	2	1.6	1.6	85.6
	Kasoor	6	4.7	4.8	90.4
	Bahawalpur	1	.8	.8	91.2
	Narowal	3	2.3	2.4	93.6
	Tobateksingh	1	.8	.8	94.4
	Pakpattan	2	1.6	1.6	96.0
	Rahimyar Khan	1	.8	.8	96.8
	Gujrat	3	2.3	2.4	99.2
	Sargodha	1	.8	.8	100.0
Missing	missing	3	2.3		
Total		128	100.0		

Complete unilateral clefts on the whole were 21% of all the clefts and amongst these, clefts involving left side were seen more common being 70% of all complete unilateral clefts compared to 29% Complete unilateral cleft of the right side.

In 6 patients (Males 5, Female 1) 4.7% complete unilateral clefts of the right side and secondary and soft palate of the left side were observed.

In the above mentioned four categories of the clefts, male dominance was found statistically significant.

Clefts of the secondary and soft palate were identified in 12 patients (Males 4, Females 8), 9.4 %. Ten patients (Males 3, Females 7) 7.8% were classified as left complete and right secondary and soft palate. Clefts anterior to incisive foramen were seen in 3 patients (Male 1, Females 2) 2.3%. Clefts of the Median soft palate were seen in 10 patients (Male 3 Female 7) 7.8%. In these four categories of the clefts female gender dominated and was found statistically significant.

Clefts occurring exclusively in female patients were seen in three categories and those in male patients were found in two categories. Two female patients 1.6% were identified as right side cleft of the nose, lip, alveolus, primary and secondary palate. Cleft of the right side nose, lip, alveolus was seen in one female neonate 0.8% and similarly cleft of left alveolus was identified in another female newborn 0.8%. One male baby was exclusively seen with bifid uvula 0.8% and another with left side cleft of the nose, lip and alveolus 0.8%.

DISCUSSION

In this study, an increased number of female patients were observed; 72 males compared to 56 females. This finding was supported by description of almost all investigators 1, 2¹, 3^{4,5} Complete bilateral cleft of the lip and palate were seen in 54 patients i.e., 42% (35 males, 19 females), being the largest proportion of the study. This finding was contradictory to the report of Bellis and Wohlgemuth³, Singh G⁵, Hagberg Larson, Milerad¹¹ and Baek, Moon, Yang". The reason for an increased number of these clefts is probably due to the fact that children's hospital gets referral from other hospitals for this exclusive treatment, using custom made anterior acrylic ring orthopedic plate". Bellis and Wohlgemuth have reported 11% distribution of complete bilateral cleft of the lip and palate comprising 75% male and 25% female patients. Singh G has quoted 20%

frequency of bilateral clefts. Hagberg Larson and Milerad had also reported 0.3/1000 frequency of bilateral clefts compared to 0.6-0.7/1000 births of unilateral clefts. Baek, Moon, Yang reported an over all distribution of unilateral and bilateral clefts as 76 and 24 %. Greater occurrence of complete bilateral cleft of the lip and palate in males is however supported by almost all researches.

The next largest type examined was complete unilateral cleft which on the whole were calculated to be 21% of all the clefts. Amongst these, clefts involving left side were more common being 14.8% compared to 6.3% complete clefts of the right side. These frequencies were supported by the finding of Bellis and Wohlgemuth who reported 13% occurrence of complete unilateral cleft of the left side and 6% of the right side. Baek, Moon, Yang on the other hand reported maximum proportion of unilateral cleft that is 76% compared to 24% complete bilateral clefts. Blanco-Davila⁴ has also reported greater occurrence of unilateral clefts of the left side. As per text book report from Singh G, proportion of unilateral complete clefts is 80% of all the clefts and those involving left side are more common (70%), where as bilateral clefts affect 20% of the total cleft patients. This finding of bilateral clefts frequency goes against the current study because of the above mentioned reason. Increased number of unilateral complete clefts of the left side, however, is supported by most of the reference studies. The third larger proportion examined in this study was clefts involving secondary and soft palate. This category comprised 12 patients (4 Males, 8 Females) making a percentage of 9.4%. Baek, Moon and Yang had shown 9.6% occurrence of the cleft palate in 12 year studies. Their finding supports the current study. Bellis and Wohlgemuth on the other hand, reported the largest proportion of 45% involving secondary palate. Cleft of the Median soft palate were seen in 10 (7.8%) neonates (3 Males, 7 Females) born with Pierre Robin's sequence. These infants were referred from the Neonatology Department of the CH & ICH. The least common types of clefts were identified in four categories of this study, each presenting one patient (0.8%). These included a male neonate with bifid uvula, a female patient with right and a male patient with left side cleft of the nose, lip and alveolus. The last of these four types appeared in a female as cleft of the left alveolus. Baek, Moon and Yang however, have reported 19.2% proportion of cleft lip and alveolus.

An unusual finding of this study was complete unilateral cleft along with opposite secondary and soft

palate cleft. This type has not been reported in previous studies in such a detail. It has been possible probably because of Kernahan's modified double stem of the symbolic 'Y' that provided detailed diagrammatic representation and record keeping of each part and either side of the cleft area. The median line of the double stem of the Kernahan's "Y" represents the mid Sagittal plane. This parameter has enabled to identify this uncommon type of the cleft.

Sandy et al¹⁴ as well as Hammond and Stassen¹⁵ emphasized that statistics on the prevalence of these clefts and their clinical outcome are essential for the progress of research, clinical audit and planning of clinical services and specialist training. Moreover, it is important that such statistics are based on valid and accurate data. In order to find incidence of oral clefts, accurate data of congenital anomalies must be maintained by the government agencies and that data should range from individual unit's record to the national register. Registration of the cleft patients should be made compulsory rather than on voluntary basis, involving government, semi-government and private sector. Such an authentic and valid data collection is being implemented in certain developed countries.

In Pakistan, however, no such system is in place and calculating incidence of the clefts in Pakistani population is not possible as such. On that basis, this study aimed at identifying the proportions of clefts in patients visiting children's hospital during five year period. The patients referred to the Cleft center, however, belong to different places (Table 3). Twenty one cities were recorded in the data. One patient reported from Kotli, Sindh, while all others were referred from both adjacent and remote areas of the Punjab. Majority of the sample (67 patients, 52.3%) belonged to the area of the cited hospital i.e., Lahore. The next larger number analyzed was 10 patients (7.8%) from Gujranwala.

CONCLUSIONS

This study concluded:

- Maximum number of patients presented with complete bilateral cleft of the lip and palate.
- The next common finding was complete unilateral cleft of the lip and palate and among these patients; clefts involving left side were more common.

- Clefts involving secondary and soft palate were the third largest in proportion.
- An unusual finding of this study was complete unilateral and partial cleft of the opposite side involving secondary and soft palate.

Further research based on modified double "Y" classification system is suggested to delineate information regarding the cleft deformities.

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