

40 Years Ago

Statistical analysis of 750 cleft lip and palate patients

San D. Theogaraj, LBM Joseph. Mani M

Christian Medical College and Hospital, Vellore, India

INTRODUCTION

The period under study is from January, 1959-June, 1967 and 750 consecutive patients have been analysed.

In an earlier analysis, we presented 492 cases in 1964, utilizing the classification of Davis and Ritchie.^[1] In this paper we have favoured the Harkins et al classification,^[2] since it is based on more sound embryological principles.

INCIDENCE [TABLE 1]

From 1959-1967, we have had 645,197 new out-patients and 750 of these have been patients with cleft lip and/or palate. This is an incidence of 1:860. Of course, this is by no means the absolute incidence in the general population, but only an indication of its relative frequency.

During this period we have had 13,821 live births in our hospital and of these there were 26 cases of cleft lip and/or palate; an incidence of 1:532 live births. Oldfield,^[3] found an incidence of 1:600 live births.

CLEFT TYPE [TABLE 2]

Prepalatal clefts, in the Harkins classification, are those that involve the region anterior to the naso-palatine foramen. These clefts occur between the 4th -7th week and are due to

lack of mesodermal penetration.^[4]

Palatal clefts are those that are posterior to the naso-palatine foramen. These become cleft due to failure of fusion of the two shelves and this takes place between the 7th - 12th week. The Davis and Ritchie^[1] classification lays stress on the alveolus and divides clefts into pre-alveolar, post-alveolar, etc. Since the dividing zone ought to be the foramen and not the alveolus, we have elected to use the Harkins classification, throughout.

In all of the studies including our own, clefts of the prepalate and palate (2 and 3) rank the highest. In our series and in that of Keys Smith, prepalatal clefts are next in frequency and isolated clefts of the palate the least. This is exactly reversed in the Caucasian series.

This difference may be an artificial one. Parents in the East are more likely to bring a child with a cleft lip to the hospital, than if it were to have an isolated cleft of the palate, because this would not be obvious to the casual observer and also would not serve to remind the parents of the defect everyday. When the child begins to talk, however, the defect becomes revealed and the children then tend to be brought for treatment. This statement is borne out by scrutinizing the analysis of the age at First Visit to Hospital [Table 3].

Most children with clefts of the prepalate with or without palate involvement are brought to hospital before their first year. Clefts of the palate alone, are brought in much later.

SEX DISTRIBUTION [TABLE 4]

The male: female ratio is 4: 3. A male preponderance for clefts has been observed in all major series of patients.

Clefts of the palate alone are more common in females F: M

Table 1: Incidence In C, M. C. H. (1959-1967)

New patients with C L and/or P	New O.P.D. patients
750	645,197
INCIDENCE IS 1: 860	
Live births with C L and/or P	Total no. of live births
26	13,821
INCIDENCE IS 1; 532	

This article was published 40 years ago in 1986 in the Indian Journal of Plastic Surgery, in volume 1, issue 1, page 11-21.

Table 2: Cleft types

Type of (Denmark)	Fogh-Andersen 1942 (Leeds)	Oldfield 1959 1960	Rank and Thomson 1961 (Tasmania)	Fraser and Calnan 1963 (Oxford)	Woolf et al. 1962 (Utah)	Keys Smith 1967 (Singapore)	C. M. C. Hospital Cleft (Vellore)
1. Prepalate only	138 (19.6%)	233 (22.4%)	50 (22.6%)	93 (20.4%)	128 (23.1%)	90 (25.1%)	216 (28.8%)
2 and 3. Prepalate and palate	360 (51.2%)	450 (43.2%)	97 (43.9%)	152 (33.3%)	290 (52.5%)	224 (62.4%)	428 (56.9%)
4. Palate only	205 (29.2%)	358 (34.4%)	74 (33.5%)	211 (46.3%)	135 (24.4%)	45 (12.5%)	106 (14.3%)

Table 3: Age at first visit to hospital (1959-1967)

Type of Cleft	0-30 Days	1 mth. -1 Yr.	1-2 Yrs.	2-3 Yrs.	3-4 Yrs.	4-5 Yrs.	5-9 Yrs.	9-14 Yrs.	Above 14 Yrs.	Total
Prepalate only	1	85	21	15	9	8	28	16	33	216
Prepalate and palate (complete)	12	132	67	37	26	14	50	35	35	408
Prepalate and palate (incomplete)	0	3	6	1	2	0	3	1	4	20
Palate only	3	11	13	17	12	12	18	8	12	106
Total	16	231	107	70	49	34	99	60	84	750

(6: 4). The ratio is exactly reversed in clefts of the prepalate with or without palate involvement, M: F 6: 4.

FAMILY HISTORY [TABLE 5]

The overall incidence of clefts in families is 12.1%. Oldfield found a 12.5% family history in a study of 1,041 patients.

OTHER CONGENITAL ANOMALIES

They were found in 60 cases. There were 63 anomalies in all. Isolated clefts of the palate had a higher incidence than the other varieties. Drillien et al.,^[8] came to a similar conclusion in their Edinburgh study.

CLEFTS OF THE PREPALATE [TABLE 6]

Left sided clefts occurred in half of all prepalatal clefts. Right sided and bilateral clefts constituted 25% each. Complete clefts by far outnumbered the incomplete. Fogh

Table 4 (a): Sex incidence

Males	Females'	Total
419	331	750
Cases	Cases	Cases

Table 4 (b): Sex Distribution

Type of Cleft	Macmahon and Mckeown 1953 ^[5]		Fogh Andersen 1942 ^[6]		Rank and Thomson 1960 ^[7]		C. M. C. H. Series 1967	
	M	F	M	F	M	F	M	F
1 Prepalate only	60.6%		65.2%		64.0%		54.6%	
2 and 3. Prepalate and palate	59.0%		71.4%		76.3%		60.0%	
4. Palate only		28.8%		64.9%		56.8%		61.3%

Andersen^[6] had 64% left sided clefts in his series, Rank and Thomson^[7] 68% and Fraser and Calnan^[9] 51.6%. The preponderance of left sided clefts of the pre palate seems therefore to be a universal finding.

CLEFTS OF THE PALATE [TABLE 7]

In order to compare our series with that of others, we have simultaneously classified cleft palates, according to the Veau (1931) classification.

The figures in our series and in that of Keys Smith's,^[10] are almost identical. More severe clefts, tend to be brought to hospital more often, in our part of the world.

Table 5: Family history and other congenital anomalies

Type of cleft	Family history	Other congenital anomalies	Total
Prepalate only	20 (8.3%)	11 (5.1%)	216
Prepalate and palate (Complete)	53 (13.0%)	33 (8.1%)	408
Prepalate and palate (Incomplete)	2 (8.7%)	1 (4.3%)	20
Palate only	16 (15.0%)	15 (14.1%)	106
Total	91 (12.1%)	60 (8.0%)	750

DISTRIBUTION STATEWISE [TABLE 8]

As is to be expected, the majority of patients that attend our hospital, come from our own state. The rest are mainly from the neighbouring regions. More detailed analysis of the 250 cases (1965-1967).

CONSANGUINITY [TABLE 9]

One hundred and thirty five out of 250 cases had parents that were very closely related to each other. The prevalent custom in our part of the country, however, is towards an uncle-niece or 1st cousin marriage. It is difficult, therefore, to assess how much weight ought to be given this finding.

AGE AT CONCEPTION [TABLE 10]

Majority of the mothers conceived between 15-30 years of age (86.0%). Loretz *et al.*^[11] in a study from California, found 13;0 of mothers of affected children between 30-40 years of age, as compared with less than 10%[®] of mothers of all infants. We also have 13% of mothers in the same age group, but have no figures as yet for the general population.

BIRTH RANK [TABLE 11]

Malpas^[12] and Murphy,^[13] were of the opinion that maternal age over 40 years and later parities predisposed to the birth of children with clefts. Oldfield,^[3] Knox^[14] and others have not found this to be so. Our findings are similar to the latter group of workers. More than half of our patients fall within the 1st two birth ranks.

ADVERSE ENVIRONMENTAL FACTORS [TABLE 12]

Diseases and other noxious influences during the 1st trimester were found in only 6% of cases. In a retrospective study, one is hard put to attach too much significance to this, especially when numbers are so few and a comparable history from mothers of normal infants is lacking. Detailed questioning, for all possible teratogenic influences operating in the 1st trimester, was undertaken in the last 250 cases, but the response from the informants was far from satisfactory.

Frank middle ear disease has been detected in only 14 cases [Table 13]. There was sensory neural loss in one case. This gives a percentage of 9% of 172 cases, with hearing loss. Peer

Table 6: Clefts of the prepalate

Left		Right		Bilateral	Total
Incomplete	Complete	Incomplete	Complete		
58 (9.0%)	267 (41.5%)	41 (6.4%)	114 (17.7%)	164 (25.4%)	644 (100%)
50.5%		24.1%		25.4%	100%

Table 7: Clefts of the Palate

Veau Types	Veau 1931	Oldfield 1949 ^[3]	Keys Smith 1962 ^[10]	C. M. C. H. series 1967
Types I and II	516 (51.6%)	190 (46.0%)	60 (22.3%)	126 (23.6%)
Types III and IV	484 (48.4%)	222 (54.0%)	209 (77.7%)	408 (76.4%)
Total	1000 (100.0%)	412 (100.0%)	269 (100.0%)	534 (100.0%)

Table 9: Consanguinity

Relationship	Number of cases
Uncle-Niece	60
First cousins	42
Other	33
Total	135
Not related	115
Total	250

Table 8: Statewise Distribution

Type of cleft	Tamilnad	Kerala	Andhra Pradesh	Mysore	Others
Prepalate only	149	19	32	11	5
Prepalate and palate (complete)	224	61	64	37	22
Prepalate and palate (incomplete)	15	1	4	0	0
Palate only	65	22	12	4	3
Total	453	103	112	52	30

Table 10: Age of mother

Age at Conception	Number of cases
Below 14 years	2 (1.1%)
15-20 years	51 (29.6%)
20-30 years	97 (56.4%)
30-40 years	22 (12.9%)
Over 40 years	0
Total	172
Not known	78
Total	250

Table 11: Birth rank

Type of Cleft	Parity											Total
	1	2	3	4	5	6	7	8	9	10	Not known	
Prepalate only	22	14	11	13	9	3	-	1	1	1	3	78
Prepalate and Palate (Complete)	35	40	14	14	5	5	3	2	1	-	7	126
Prepalate and palate (Incomplete)	2	1	3	2	1	-	-	-	-	-	1	10
Palate only	11	6	6	5	2	2	1	-	2	-	1	36
Total	70	61	34	34	17	10	4	3	4	1	12	250

Table 12: Adverse environmental factors

Type of cleft	Disease			Total	Percentage	
Prepalate only	Hyperemesis		Malaria?	2	2.5% of 78 cases	
Prepalate and palate (complete)	2	Viral infect 5	1	Asthma 2	10	8.0% of 126 cases
Prepalate and palate (incomplete)		N	I	L		
Palate only		N	I	L		

Table 13: Incidence of deafness

Type of cleft	Otitis Media	Sensory neural loss	Otosclerosis
Prepalate only	1		
Prepalate and palate	4	1	
Palate only	1		1
Total	5	1	1

et al^[15] detected 60% and Masters *et al.*^[16] 50% of children with cleft palates, to be afflicted with significant hearing loss. Routine E.N.T. examinations have been conducted in all our cases, but audiograms have been done only when felt indicated and hence, our low figures. Recently, Stool and Randall^[17] reported on middle ear pathology in 94% of cases with clefts, when routine myringotomies were performed.

Since the physiology of the Eustachian tubes is interfered with when a cleft is unrepaired, the onset of middle ear disease is not surprising. The longer after 18 months a cleft of the palate is unrepaired, the higher the incidence. All of our affected cases had repairs after 18 months and the more severe the cleft, the greater the number of cases with middle ear diseases. Masters^[16] was of a like opinion.

SUMMARY

1. 750 cleft lip and palate cases have been analysed.
2. Clefts which involve the prepalate and palate proper, are seen most commonly.
3. M:F ratio is 4:3.
4. There is a positive family history in 12.1%.
5. Left sided clefts of the prepalate are found in 50% of cases.

6. There was a history of consanguinity in over 50% of 250 cases.
7. Maternal age and birth rank are not of much significance.
8. Middle ear disease was found in only 9% of cases and the reason for this is offered.

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Source of Support: Nil, **Conflict of Interest:** None declared.