

Epidemiology of Cleft Lip and Palate in Singapore-A 10-Year Hospital-based Study

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Abstract

During the study period from January 1985 to December 1994, there were 1105 new cleft cases seen in the Department of Plastic Surgery, Singapore General Hospital. These included newborn as well as unoperated children and adult cases. During the same period, the total number of recorded births in Singapore was 474,542. Out of the 1105 new cleft cases seen, 984 were Singaporeans. The incidence of this hospital-based study of cleft population in Singapore was 2.07 per 1000 livebirths. Chinese had the highest incidence of 1.64 per 1000 as compared to Malay, Indian and other races. The most common type of cleft deformity was complete cleft lip and palate. The left side was found to be more affected than the right side in all types of cleft deformity. There was no significant difference in sex distribution; the male to female ratio was 1.1:1. However, females had a higher incidence of cleft palate than males. Associated congenital deformities occurred in 1.5% of the total cleft population.

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Key words: Associated abnormalities, Cleft types, Incidence patterns, Surgical repair

Introduction

With the control of most infectious diseases and malnutrition, congenital malformations are now one of the most important causes of morbidity and mortality among infants in Singapore. Children born with cleft deformities require specialised multidisciplinary care from infancy till adulthood. The basis of preventive measures within the family and the population at large should be based on the study of incidence and epidemiological patterns.

The aim of our study was to evaluate in detail all the cleft lip and palate cases seen and operated in the Department of Plastic Surgery, Singapore General Hospital (SGH) for the 10-year period from 1985 to 1994. A previous case-controlled study carried out at Kangar Kerbau Maternity Hospital had reported that cleft lip and palate was the fourth commonest birth defect in Singapore.^{1,2}

Materials and Methods

A retrospective study was carried out covering a 10-year period from January 1985 to December 1994. All new cases of unoperated cleft lip $\pm$ palate were included in the survey. The total number of cleft cases was 1105. Out of this, the number born in Singapore was 984. The

remaining 121 non-Singaporean comprised Indonesian, Malaysian, Chinese and Indian from other countries and a few Caucasian patients. Birth and population data were obtained from the computerised records of the Ministry of Health for the years 1985 to 1994. All the case records were retrieved from the hospital medical record data file.

A standard epidemiological survey form was prepared for entry of all relevant information obtained from the casenotes. This included the patient's biodata, the cleft classification, laterality of cleft, degree of cleft, presence of Simonart's band and association with other abnormalities. The operative history, method of operation, date of operation and name of surgeons were noted. Apart from this, drug history during pregnancy and consanguinity history of previous pregnancy were also studied. Stillbirths with cleft deformities were not included in this study.

Results

A total of 474,542 livebirths were recorded in Singapore during the 10-year study period (1985 to 1994). There were 984 Singapore patients with primary cleft deformity seen in a similar period, giving a prevalence of 2.07 per 1000 livebirths. There were 1105 new cases

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who attended the plastic surgery outpatient department of SGH and these included both newborn infants with clefts as well as children and adult with unrepaired cleft lip $\pm$ palate.

Cleft Types

These were 546 cases of complete cleft lip and palate giving an incidence rate of 1.16 per 1000 livebirths, 208 cases of cleft lip giving an incidence rate 0.43 per 1000 livebirths and 230 cases of cleft palate giving an incidence 0.48 per 1000 livebirths (Table I).

TABLE I: INCIDENCE OF BIRTH DEFECTS

	No. of cases	Rate per 1000 livebirths
Cleft palate	230	0.48
Cleft lip	208	0.43
Cleft lip and palate	546	1.16
UCLP/BCLP	984	2.07

UCLP: unilateral cleft lip and palate; BCLP: bilateral cleft lip and palate

Race and Gender Incidence

Singapore has a multiracial population comprising 77.8% Chinese, 14.1% Malays, 6.9% Indians and 1.2% others. Chinese were found to have the highest incidence of cleft. There were 778 new cleft cases, giving an incidence of 1.64 per 1000 livebirths.

The total number of cleft cases in the Malay population was 141, giving an incidence rate of 0.29 per 1000 livebirths. Amongst the Indian population, there were 49 cases of cleft deformities, giving an incidence rate of 0.10 per 1000 livebirths. There were 16 cases of cleft affected children in other races, giving an incidence of 0.04 per 1000 livebirths (Table II). Males were found to be slightly more affected than females, giving a sex ratio of 511 males to 473 females or 1.1:1.

Laterality of Clefts

Out of a total of 375 complete unilateral cleft lip and

TABLE II: RACE DISTRIBUTION

Race	No. of cases	Incidence per 1000 livebirths
Chinese	778	1.64
Malay	141	0.29
Indian	49	0.10
Others	16	0.04
Total	984	2.07

palate (UCLP) cases, the left side was affected in 275 cases and the right side in 100 cases. Complete bilateral complete cleft lip and palate (BCLP) cases were seen in 171 cases (17.4%). In all these subgroups, males were more affected than females and Chinese was the most affected of all the races.

In 208 cases of cleft lip (CL), males were also more affected than females. There were 118 left-sided clefts compared with 67 right-sided clefts and 23 bilateral clefts. An interesting reversal of sex ratio was noted in the isolated left-sided cleft lip being more common in females than in males, the ratio being 1.2:1.

For the posterior cleft palate, females were more often affected than males. Distribution was again predominant in the Chinese population (Table III). There were 18 cases of Simonart's Band present in cleft lip, giving an incidence of 2.4%.

Association with Other Congenital Anomalies or Syndromes/Rare Clefts

There were 15 cases in which other associated congenital syndromes were present such as Pierre Robin syndrome (5), APERT syndrome (2), Goldenhar syndrome (4), congenital heart disease (3) and spina bifida (1), giving a percentage of 1.5%. There were 8 cases of microform cleft lips, 2 cases of bilateral angular cleft and 2 cases of median cleft with a total number of 12 unusual cleft cases.

TABLE III: SUMMARY OF CLEFT DISTRIBUTION PATTERN AMONG THE DIFFERENT RACES AND SEXES

Cleft pattern	Male	Female	Ratio	Total	Chinese	Malay	Indian	Others
Cleft lip and palate (n = 546 cases)								
Right UCLP	60	40	1.5:1	100	83	12	3	2
Left UCLP	139	136	1.0:1	275	231	19	17	8
Cleft lip & palate	114	57	2.0: 1	171	132	30	9	0
Cleft lip only (n = 208 cases)								
Right cleft lip	38	29	1.3:1	67	54	8	5	0
Left cleft lip	52	66	1.0:1.2	118	89	19	6	4
Bilateral cleft lip	15	8	1.8:1	23	18	5	0	0
Cleft palate (n = 230 cases)	93	137	1.0:1.4	230	171	48	9	2
Total	511	473		984	778	141	49	16

UCLP: unilateral cleft lip and palate

Operative Procedures

All cleft cases were operated by staged repair depending upon the type and severity of the cleft deformity. Usually cleft lip was repaired at 3 months of age, cleft palate at 9 months of age, the tethered nasal tip deformity in BCLP cases was repaired at 4 to 5 years before going to school, alveolar bone graft with lip revision was carried out around 9 years before eruption of the canines, LeFort osteotomy and/or sagittal setback of mandible was carried at about 17 to 18 years for class III malocclusion. Procedures like corrective rhinoplasty, correction of whistle tip deformity, lip scar revision, Abbe's flap for deficient upper lip in bilateral cleft cases, pharyngoplasty for velopharyngeal incompetence (VPI), and finer adjustment operations for the lip were carried out at the appropriate time but before the final outcome assessment at 21 years of age.

The repair of some cleft lip and palate cases was sometimes delayed due to the child's general condition and unfitness for general anaesthesia. There may be associated congenital problems like congenital heart disease or failure to thrive that necessitated delay of surgical repair.

There were 15 cases of cleft lip repairs which were delayed because of associated abnormalities. Out of these, 5 cases were repaired around 5 years of age and the remaining 10 cases were operated before 10 years. For palate repairs, 19 cases were delayed for operation until 4 to 5 years of age.

Complete cleft lip and palate cases usually requires at least 3 procedures comprising lip repair, palate repair and alveolar bone graft. Out of the total of 984 Singapore patients, 172 required more than 3 procedures: 83 cleft-affected children had 4 procedures before final assessment, 38 had 5 procedures, and 51 had more than 5 procedures. There were 38 cases of unrepaired clefts of the secondary palate presenting in adulthood, the oldest of whom was 55 years of age. In 10 adult unrepaired cleft lip cases, there were 5 cases between 10 and 20 years, and 5 cases between 20 and 30 years.

Technique Of Repair

As far as the technique of surgery was concerned, for the lip, most of surgeons used the modified Millard's technique (734 cases). Nineteen cases were done using Skoog's method and 1 case by Randall's modification of Tennyson's technique.

For the palate repair, most of the cases (202 out of 230) were repaired by Veau-Wardil Kilner technique and 28 cases by Von Langenbeck technique. There were 7 surgeons who had carried out the different procedures (Tables IV and V).

Discussion

Because of the high degree of health awareness and a

centralised health care delivery system, most of the cases in Singapore were referred soon after birth. This study actually represents the total incidence of cleft per total livebirth population in Singapore, which reveals the high incidence of cleft lip and palate in Singapore and ranks it amongst the highest in the world.

The overall prevalence of cleft deformity seen in the Department Plastic Surgery, Singapore General Hospital was 2.07 per 1000 livebirths. Other reported studies³⁻¹⁹ from 17 different countries (Table VI) including Western as well as Eastern countries had shown incidence rates ranging from 0.69 per 1000 livebirths to 2.19 per 1000 livebirths. Out of these, the highest incidence rates (above 1.89/1000 livebirths) were found in five other countries besides Singapore^{6,8,11,15,19} (Table VII). A previous study of cleft deformity in Singapore (1977-1981) by Fong et al¹² had shown the high incidence rate of 2.0 per 1000 livebirths but a case-control study from Kadang Kerbau Maternity Hospital, Singapore (1986 to 1988)^{1,2} had reported the rate as 1.72 per 1000 livebirths.

Our 10-year study (1985 to 1994) as compared to that of a previous 5-year study¹² showed an increasing rate in Singapore. The highest rate of incidence (2.19 per 1000 livebirths) was found in the study from Saudi Arabia¹⁹ and the next highest incidence rate of 2.06 per 1000

TABLE IV: SURGICAL REPAIR OF LIP AND PALATE

Lip — cleft lip alone or cleft lip and palate	Randall's	Skoog's	Millard's
	1	19	734
Palate (Isolated)	Von Langenbeck		Veau-Wardil Kilner
	28		202

TABLE V: SURGEONS VERSUS SURGICAL PROCEDURES DONE

Surgical procedure	Senior consultant surgeon	Other surgeons
Cleft lip repair	381	221
Cleft palate repair	142	323
Left ABG	66	114
Right ABG	75	55
Nose repair-unspecified	34	31
Cartilage graft	7	18
Silastic implant	7	8
Tip release	31	10
Scar revision	73	70
Abbe's flap	6	18
Palatopharyngoplasty	11	29
Fistula closure	13	34
LeFort osteotomy/Bimax		42
Total	846	973
Total no. of operations	1819	

ABG: alveolar bone graft

TABLE VI: INCIDENCE

Author	Name of country	Year of study	Incidence per 1000 livebirths
Fogh-Andersen ³	Denmark	1942	1.31
Knox & Braithwaite ⁴	Northumberland & Durham, UK	1949-1960	1.42
Woolf et al ⁵	Utah, USA	1951-1961	1.51
Lowry et al ⁶	British Columbia, Canada	1952-1986	1.98
Owens et al ⁷	Liverpool, UK	1960-1982	1.40
Welch & Hunter ⁸	Canada	1964-1977	2.00
Womersley & Stone ⁹	Glasgow, Scotland	1974-1985	0.69
Tolarova ¹⁰	Czechoslovakia	1975-1982	1.63
Jensen et al ¹¹	Denmark	1976-1981	1.89
Fong et al ¹²	Singapore	1977-1981	2.00
Stoll et al ¹³	Northern-eastern France	1979-1987	1.75
Gregg et al ¹⁴	Northern Ireland	1980-1990	1.28
Natsume et al ¹⁵	Japan	1982	2.06
Morrison et al ¹⁶	Western Cape, South Africa	1983	1.00
Boo & Arshad ¹⁷	Malaysia	1986-1987	1.24
Srivastava & Bang ¹⁸	Kuwait	1990	1.48
Borkar et al ¹⁹	Al-Gassim, Saudi Arabia	1992	2.19
Singapore General Hospital	Singapore	1985-1994	2.07

livebirths was from Japan.¹⁵ Another study from Canada (1964 to 1977)⁸ had shown the incidence rate of 2.0 per 1000 livebirths whereas Denmark (1976 to 1981)¹¹ and British Columbia (1952 to 1986)⁶ had incidences of 1.89 per 1000 livebirths and 1.98 per 1000 livebirths, respectively.

When studying the pattern of cleft distribution and sex incidence amongst these six countries with high prevalence rates, we found that the combined type of cleft lip and palate deformity (UCLP and BCLP) was the commonest clinical variant. Males were more often affected than females in combined cleft lip and palate as well as in isolated cleft lip deformity whereas posterior cleft

palate was commonest in females in all these countries. Therefore the pattern of cleft distribution and sex incidence was found to be the same in all the countries with high incidence rates. A comparison with a previous study from SGH (1977 to 1981)¹² showed that there were no major changes between sex and race distribution apart from a slight increase in incidence rate (Table VIII).

As far as the laterality of cleft was concerned, the left side was found to be commonest than the right side in both types-the complete cleft lip and palate and cleft lip. The complete cleft lip and palate deformity was twice as common as the cleft lip or cleft palate deformity.

According to a previous study in Singapore,⁷ increased incidence of facial clefts was found to be associated with early menarche, and the intake of traditional Chinese herbal medicine by mothers of cleft affected children. Therefore prospective studies will be necessary to elucidate the role of environmental risk factors during pregnancy such as smoking, intake of herbal medicine, trauma, viral infections, drug intake and also the maternal age of onset of menarche. This may help us to have a better understanding of the causative factors so that

TABLE VIII: COMPARISON OF THE RESULTS IN SGH-RACIAL AND SEX INCIDENCE IN CLEFTS

	5-year study, 1977 to 1981 (n = 450)		1 O-year study, 1985 to 1994 (n = 984)	
	No.	%	No.	%
Race				
Chinese	369	76.9	778	79.0
Malay	61	14.6	141	14.3
Indian	15	6.4	49	7.9
Others	5	2.19	16	1.6
Sex				
Male	228	50.6	511	51.9
Female	222	49.4	473	48.1

TABLE VII: COMPARISON AMONG THE COUNTRIES WITH HIGHEST INCIDENCE RATE

Country	Cleft lip		Cleft lip and palate		Cleft palate		Total no.		Incidence per 1000 livebirths
	M	F	M	F	M	F	M	F	
Denmark, 1978-1981	149	78	185	80	77	109	411	267	1.89
British Columbia, 1952-1986	581		1014		978		2573		1.98
Japan, 1982	31		39		13		83		2.06
Saudi Arabia, 1989-1992	56		62		19		137		2.19
	M	F	M	F	M	F	M	F	
Singapore General Hospital, Singapore, 1985-1994	105	103	313	233	93	137	511	473	2.07

preventive measures can be taken. The results from this study provide basic information for the planning of health service requirements for this group of cleft patients and future resource allocation should take epidemiological data firmly into account.

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