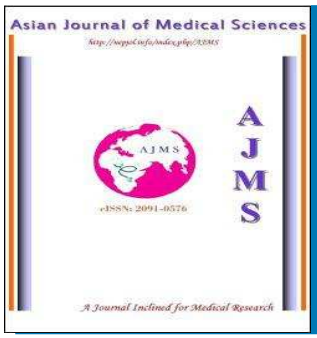

PILOT EPIDEMIOLOGICAL STUDY OF CLEFT LIP AND /OR PALATE IN KOTA KINABALU, SABAH

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"Male patients were more predominant in cases of left sided cleft lip with or without palate but there was preponderance towards female patients in isolated secondary cleft palate. Late presentation of patients reflected the poor understanding regarding the diagnosis and management of cleft among the patients and health care providers"

ABSTRACT

Objective: To identify number of cases and the type of cleft lip and/or palate managed in government tertiary center (Queen Elizabeth Hospital) in Kota Kinabalu; and to analyze the associative factors of cleft lip and/or palate.

Methods: A retrospective cross-sectional study carried out in Hospital Queen Elizabeth, Kota Kinabalu, Sabah from January 2011 to December 2012. Data from 162 new cases, which were referred for cleft lip and/or palate, were included in the study.

Result: Cleft lip and palate was the most common type. While cleft lip with or without palate had higher preponderance towards male patients, secondary palate however was more common among female patients. These results were statistically significant.

Conclusion: Further and larger scale study need to be carried out to identify the incidence of cleft lip and/or palate in Sabah, and its associated genetic and environmental risk factors. Early identification and intervention for cleft lip and palate need to be emphasized.

Keywords: cleft lip, cleft palate, epidemiology, risk factor of cleft

INTRODUCTION

Congenital cleft lip and palate are among the commonest congenital craniofacial anomalies referred to and managed by plastic surgeons[1]. To optimize treatment outcomes, patients born with cleft lip and palate require coordinated care from multiple specialties and support teams. Many epidemiologic studies for oral cleft anomalies have been carried out worldwide and international literature estimates the incidence of cleft lip and palate to be approximately 0.8 to 1.6 for every 1000 live births. Risk factors of cleft lip and/ or palate including genetic factors and environmental factors for example consumption of alcohol, anticonvulsant and smoking. The aim of this pilot study is to analyze the number of cleft cases in the years 2011 and 2012, as well as to be a stepping stone to establish the incidence of cleft lip and/or palate in Sabah for possible further research.

MATERIALS AND METHODS

This retrospective cross-sectional study was conducted at the Hospital Queen Elizabeth, Kota Kinabalu which happens to be the sole tertiary government center in Sabah that provides plastic and reconstructive surgery services and receives referrals from all 14 districts within the state, as well as Labuan (Federal Territory) and Lawas (northern Sarawak). A total of 170 new patients were referred to the plastic surgery clinic from 1st January 2011 to 31st December 2012. However, 8 patients were excluded in this study as 2 patients were post operative and 6 patients had defaulted initial appointments. Data of the remaining 162 patients were obtained from the clinic register and individual case notes, and subsequently tabulated and analyzed using IBM SPSS statistics Version 20 (Clinical Research Center, Hospital Queen Elizabeth).

RESULTS

A total of 170 new referrals of cleft lip and/ or palate from Sabah to Hospital Queen Elizabeth, Kota Kinabalu from 1st January Year 2011 to 31st December Year 2012. Exclusion criteria were cases which cleft deformities been repaired and incomplete data. Therefore, 162 patients were included in the study. There were 108 (66.7%) male patients and 54 (33.3%) female patients (Table 1).

Classification of cleft type

The most frequent cleft type was cleft lip and palate, accounting for 71% overall, followed by cleft palate 17.3% and cleft lip 11.7%. Male patients were more predominant in the results of the study, which accounts for 15 out of 19 cases of cleft lip; and 81 cases from total 115 cases of cleft lip and palate. There was slight preponderance of female patients represented in the case of isolated cleft palate which accounted 16 out of 28 cases (Table 2).

Cleft Laterality

In case of cleft lip, the majority cases were left sided (52.6%). Cleft lip and palate were also more often on left side compared to right which accounted for 38.3%. Overall left sided oro-facial cleft was more common compared to right sided cleft, followed by bilateral cleft (Table 3).

Gender differences in cleft laterality

Left-sided cleft was more common for cleft lip with or without palate, accounting for 54 patients (33.3%), with predominance in male patients ($p < 0.05$) (Table 5).

Frequency of Associated Syndromic and Nonsyndromic Congenital Abnormality

From 170 patients, only 2 (1.2%) patients were noted to be syndromic. One of the syndromic patients was found to have chromosome study of 46XY add (22)(p33) associated with congenital abnormality i.e. CTEV; and the other was a confirmed case of Trisomy 23.

7 (4.3%) patients were noted to have congenical abnormality. Out of 7 patients, 2 patients associated with 2 congenital abnormalities. 5 out of 7 patients noted to have congenital heart diseases.

Table 1: Distribution of Gender and Age Group of Patients

Variable	Frequency n	Percentage (%)
Gender		
Male	108	66.7
Female	54	33.3
Total	162	100
Age Group (months)		
1 to 6	126	77.8
6 to 12	18	11.1
> 12	18	11.1
Total	162	100

Table 2: Classification of Cleft Type

	n (%)	Gender		X^2 stat[a] (df)	p-value
		Male n (%)	Female n (%)		
Classification					
CL	19 (11.7%)	15 (9.2%)	4 (2.5%)		
CLP	115 (71.0%)	81 (50.0%)	34 (21.0%)	9.167 (2)	0.01
CP	28 (17.3%)	12 (7.4%)	16 (9.9%)		
Total	162 (100%)	108	54		

a Chi-square test for independence, p-value <0.05

CL *Cleft Lip* CLP *Cleft Lip Palate* CP *Cleft Palate*

Table 3: Classification of Cleft Laterality

	n (%)	Cleft Type		X^2 stat[a] (df)	p-value
		CL n (%)	CLP n (%)		
Cleft Laterality					
Right sided cleft	45	8 (42.1%)	37 (32.2%)		
Left sided cleft	54	10 (52.6%)	44 (38.3%)	5.002 (2)	0.082
Bilateral cleft	35	1 (5.3%)	34 (29.6%)		
Total		19 (100%)	115 (100%)		

a Chi-square test for independence, p-value <0.05

CL *Cleft Lip* CLP *Cleft Lip Palate*

Table 4: Gender Differences in Cleft Laterality

	n (%)	Cleft Laterality				X^2 stat[a] (df)	p-value
		Right sided cleft n (%)	Left sided cleft n (%)	Bilateral cleft n (%)	Secondary Cleft n (%)		
Gender							
Male	108 (100%)	33 (30.6%)	39 (36.1%)	24 (22.2%)	12 (11.1%)	8.850 (3)	0.031
Female	54 (100%)	12 (22.2%)	15 (27.8%)	11 (20.4%)	16 (29.6%)		
		45 (27.8%)	54 (33.3%)	35 (21.6%)	28 (17.3%)		

A Chi-square test for independence variables, p-value <0.05

Table 5: Frequency of Associated Syndromic and Nonsyndromic Congenital Abnormality

	No	Total patient (n)
Syndromic		
Syndrome with chromosome 46 XY add (22)(p33)	1	2
Down Syndrome	1	
Congenital Abnormality		
CTEV (Congenital Talipes Equino Varus)	1	
Haloprosencephaly	1	
VSD (Ventricular Septal Defect)	2	
PDA (Patent Ductus Ateriosus)	1	7
ASD (Atrial Septal Defect)	2	
Ano malformation	1	
Micropthalmia	1	
* 2 patients had 2 congenital abnormalities		

Table 6: Distribution Family history

		n	Frequency n	Percentage %
Family History				
No		124	124	76.5
Yes		38		
	1st degree relative		11	6.8
	2rd degree relative		17	10.5
	3rd degree relative		10	6.2

In 38 (23.5%) patients, there was family history of cleft deformity, these included 11(6.8%) patients with history of 1st degree relative, 17 (10.5%) patients with history of 2nd degree relative and 10 (6.2%) patients had 3rd degree relative. In group of patients with family history, male patients accounted for 23 (60.5%) patients whereas female patients accounted for 15 (39.5%)(Table 6).

DISCUSSION

Hospital Queen Elizabeth, Kota Kinabalu received a total of 170 patients with cleft lip and palate from January 2011 until December 2012. However only 162 patients fulfilled the inclusion criterias and were included in this study. The remaining 8 patients were excluded (6 defaulted clinic appointment pre-operatively and 2 requiring scar revision).

Despite capturing the number of patients mentioned, it does not reflect the true incidence of cleft lip and/or palate in Sabah. This is mainly due to the lack of integration of information obtained from district hospital that constructed a significant number of patients with cleft lip and/or palate managed at the district level by visiting Plastic Surgeons.

From this study, 88.9% (n=144) of the patients had a 1st visit at the age less than 12 months and were treated thereafter. On the other hand, there remaining 11.1% (n=18) of the patients presented to us at the age more than 12 months old. The oldest patient, in the latter group, was 29 years old and the youngest was 2 years.

4 of these late-presenters were left undiagnosed with secondary palate during neonatal period; whilst 11 patients missed early treatment due to logistic problem and parents' unawareness of the importance of treatment. These patients were referred again at later age during childhood when presented with speech difficulty in school. The remaining 3 patients were home-delivered and referred when such clefts were incidentally diagnosed. This finding is important to reflect the poor understanding of cleft lip and/or palate in terms of diagnosis and management among the health care providers, and in educating parents. Public education and campaign need to be organized for the public as well as health care providers.

It had been established in several epidemiologic reviews that cleft lip and palate was the most common type, [2-4]; whereas most of cleft lip with or without palate was more predominant in male patients and significantly occurred on left side. [2--8] Such findings also correspond to the result of this study. In the Sabah population, cleft lip and palate is of the commonest type. Left-sided cleft lip, and cleft lip and palate (total 33.3%) occurred more often and involving more male patients. According to Hirayama,[9] one of the reasons for greater incidence on left side cleft is that facial artery development is slower on the left side compared to right, however this has not been fully confirmed and proven. Nevertheless, there was a slight predominance in female patient for isolated cleft palate.[2-3,5] Gender and cleft laterality as discussed showed statistically significant with $p<0.05$.

Cleft lip and or palate are commonly associated with central nervous system malformation, club foot and cardiac abnormalities. The overall incidence associated with anomalies in all cleft is 29%; with highest association in isolated cleft palate.[10] Only 8 out of 162 patients (5%) were presented with syndromic or non-syndromic congenital abnormalities; all of which have cleft lip and palate. Even though the incidence of associated congenital abnormalities was lower compared to other studies,[3,11] more effort should be carried out to identify the possible associated familial risk factors for example consanguineous marriage and parental age.

Positive family history of cleft lip with or without palate increases the risk for cleft deformity in newborn. Mehboob et al. [3] and Feliciano[5] reported

17% and 11.4% of patients with family history of cleft lip and /or palate in their respective studies. In this study, 23.5% of patient noted to have positive family history of cleft lip and /or palate. Based on this result, a comprehensive genetic evaluation would have been ideal for future study.

CONCLUSION

In conclusion, this study revealed the number of cases for cleft lip and /or palate been managed in Hospital Queen Elizabeth. Late presentation of patients with cleft deformities reflected the poor understanding or lack of knowledge of cleft and /or palate among the health care provider and public.

This study revealed that cleft lip and palate was the commonest type of cleft, predominance occurred on left side and male patients. This pattern of distribution was supported by few international literatures. Family history was the only risk factor was recorded in this study.

Despite limitation of this study, clinical and developmental problem related to cleft lip and /or palate in Sabah had been highlighted. In future, a larger scale of study is advocated to document a national prevalence and to identify associated genetic and environmental risk factors. A comprehensive plan which involved multidisplinary teams is needed to provide better and complete care to patients with cleft lip and /or palate.

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