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A Preliminary Investigation into Orofacial Clefts in the Central and Western Regions of Saudi Arabia

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Epidemiology of non-syndromic orofacial cleft (NSOFC) in Medina, Saudi Arabia

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Abstract:

Aim

This study investigates the prevalence of non syndromic orofacial cleft (NSOFC) in Medina city, Saudi Arabia and their relationship to consanguinity.

Material and Methods

Infants born in the only cleft center in Medina city (Maternity and Children Hospital (MCH)) from 1st of January 2010 to the 31st of December 2011 were included in this study to calculate the prevalence. Referred NSOFC infants were included to investigate the effect of consanguinity. Data was collected through clinical examination, and parental interview.

Result

In two years, 78 NSOFC infants were born. Out of them, 50 (64%) were males. The most common phenotype seen was cleft lip (CL) (43% of NSOFC cases). Consanguineous parents represented 66.7% of NSOFC patients. NSOFC prevalence was 1.9/1000 live births. CL prevalence was 0.89/ 1000 live births, the prevalence of cleft lip and palate (CLP) was 0.68/ 1000 live births and the least prevalence was for cleft palate (CP) 0.36/1000 live births.

Conclusion

The prevalence of NSOFC in Medina is 1.9/1000 live births. The prevalence of CL was greater than CLP, and CP. The reason for this ratio needs further research.

Key words: cleft lip, cleft palate, prevalence, consanguinity, Medina

Introduction:

Non-syndromic orofacial cleft (NSOFC) is the most common orofacial congenital abnormality that is seen frequently around the world and has a birth prevalence rate ranging from 1/1000 to 2.69/1000 live birth amongst different parts of the world.[1, 2] Epidemiologic studies of NSOFC have been conducted worldwide, often resulting in varying prevalence rates. Differences in geographic and ethnic distributions may account for some but not all of the variations. Another determinant that can lead to the variation in rates is the system used to select the study group. Hospital-based records are frequently used in prevalence studies of NSCOFC, but the data are liable to ascertainment or selection bias and may lead to under reporting or over reporting of the prevalence of the cleft condition.[3-5] No study on the prevalence of NSOFC was carried out in Medina. Therefore, the aim of this study was to determine the epidemiology of non syndromic orofacial cleft (NSOFC) in Medina city, Saudi Arabia and to assess their relationship to consanguinity.

Material and Methods:

The Ministry of Health proposed the Maternity and Children Hospital (MCH) as the cleft center in Medina. It is also the largest hospital with labor facilities and the highest number of maternity patients in the region. Any patient born in this hospital from 1st of January 2010 to the 31st of December 2011 was included in this study. Also, infants referred to the hospital during the same period were included for the purpose of studying the characteristics of NSOFC and their relationship to consanguinity. Information on born and referred patients with NSOFC was obtained by the research coordinator who actively inquired about patients in neonatal intensive care (NICU) and clinics.

Data collection and parental interview was carried out through clinical examination by one person (the research coordinator). In addition, ascertainment of diagnosis was reassured by confirming NSOFC diagnosis through reviewing the medical records.

NSOFC phenotype was classified according to LASHAL classification which subdivided cleft lip (CL) according to side (right or left), and cleft palate (CP) to hard and soft. [6] In this study, CP was not further divided to soft and hard because this information was not always available because some referred patients were seen after the surgery which made it difficult to accurately record CP sub-phenotype.

In order to calculate the prevalence, the total number of NSOFC infants born in this hospital from 1st of January 2010 to 31st of December 2011 was compared to the total number of births in the same hospital in the same period of time. The total number of births was taken from the statistical records of the hospital. The additional referred NSOFC patients were not included in the estimation of birth prevalence.

Statistical analysis:

The data were analyzed using SPSS version 16. The descriptive epidemiology of NSOFC was presented, with statistics displayed in frequency and percentage. Chi square was used to test for significance in the relationship between consanguinity and severity of cleft. Significance level was set as $P<0.05$.

Results:

There were 78 patients seen in the MCH in Medina. Out of them, 50 (64%) were males (male to female ratio was 1.8:1), and 75 (96.2%) were Saudis. Parental residency included; 61 infants (78.2%) from Medina city, 15 infants (19.2%) from urban and rural areas of Medina region (6 infants (7.7%) from Yunbo four infants (5.1%) from Al-Hanakia, two infants (2.6%) from Mahd-Althahab and three infants (3.8 %) from AL-Khober, Al-Asheara and Abear-Almashi), and two infants (2.6 %) from Jeddah. Father mean age was 36.2 years with 8.4 SD

and mother mean age was 31.7±5.2. Cases with CL were seen more often in the MCH (43%) than cleft lip and palate (CLP) (33.7%) and CP (23.3%). CL was greater on the left side (21 cases, 26.9%) than on the right side (8 cases, 10.3%) The most common sub-phenotype seen for CL was incomplete left CL (14 cases, 18%) and the least was bilateral complete CL (one case, 1.3%). For CLP, the left side (11 cases, 14.1%) was also more common than the right side (6 cases, 7.7%). The most common sub-phenotype seen in CLP was left complete CLP (9 cases, 11.5%) (table 1).

In comparison of gender differences and cleft sub-phenotype, complete unilateral cleft lip with or without cleft palate (CL/P) occurred in both gender with similar proportion; males were 19 cases (38%) complete and 18 (36%) cases incomplete, while females were 10 cases complete (35.2) and incomplete cleft were 9 cases (32%). However, in comparing the gender in isolated CL cases, complete CL occurred more in Males (9 cases, 33.3%) than females (2 cases, 28.6%) and incomplete CL was more in females (5 cases, 71.4%) than males (18 cases, 66.7%).

Infants with consanguineous parents represented 52 (66.7%) of the total NSOFC cases. The most common NSOFC phenotype related to consanguinity was CP. But, there was no significant relationship (P=0.734) (table 2). The most common type of parental consanguinity was 1st cousin marriages; 30 cases (57.7%) but there was no significant differences (P=0.741) (table 3).

Birth prevalence:

During the period from 1st of January 2010 to 31st of December 2011 there were 28,134 infants born in the MCH. Out of them 54 infants were born with NSOFC. The prevalence of NSOFC was 1.9/1000 live births. The prevalence of CL was 0.89/ 1000 live births, the prevalence of CLP was 0.68/ 1000 live births and the least prevalence was for CP 0.36/1000

live births. The most common CL sub-phenotype was left incomplete CL (14.8%) However, the most common CL sub-phenotype in males was left incomplete CL (19.3%). In CLP, the most common sub-phenotype was complete left CLP (13%) and it was more common in females (21.7%). Cleft palate was more common in Females (26.1%) than males (12.9%) (see table 4).

Discussion

The purpose of this study was to report the epidemiology of NSCFC in Medina, Saudi Arabia. In this study, CL was found to be the most common NSOFC phenotype (43.5%) followed by CLP (33.3%) and then CP (23%). This was different from what was reported by previous literatures which found that CLP was more common than CL.[7, 8]

There were 50 males and 28 females with male to female ratio of 1.8:1. Gender differences were similar to previous reports, with CL and CLP found more often in males and CP found more often in females. [2, 9, 10] With regard to the reasons of gender-related differences in cleft patterns, Ross et al. suggested a relationship between female sex hormones and the palatine process. Accordingly, the timing of initial movement of bilateral palatine processes in the course of palate development occurs earlier in males and later in females, and this difference affects the degree of cleft .[11]

In terms of cleft laterality, CL occurred more common in the left side (61.8%) as well as CLP (46.2%). However, a comparison of gender differences by cleft type revealed that complete unilateral CL occurred more often in males (33.3%) than in females (28.6) while incomplete cleft occurred more in females (71.4%) compared to males (66.7%). This was different from previous reports.[8, 12]

Infants with consanguineous parents represented 52 (66.7%) of the total NSOFC case. Although this prevalence is high, it was lower than the prevalence of consanguinity in the

general population of Medina city as reported by El-Hazmi et al 1995 to be 73%. [13] However, this needs further research as consanguinity could be related to a specific NSOFC sub-phenotype rather than NSOFC in general. In addition this study found a higher prevalence of CP cases with consanguineous parents (72.2%) than other type of clefts but the different was not significant (P=0.73).

The prevalence of NSOFC in Medina was not discussed before. For the purpose of studying the prevalence, NSOFC cases born in MCH was compared to the total births in the same hospital. The prevalence of NSOFC was found to be 1.9/1000 births. This prevalence is considered to represents the prevalence of NSOFC in Medina because MCH is the main Maternity hospital and the only cleft center in Medina. The prevalence reported in this study is higher than the global prevalence which is 1.2.[2] This could be related to the high consanguinity marriages in Medina city (73%) that is considered one of the highest in the world.[13] However, it is still in the range of the global birth prevalence which ranged from 0.43-2.45/1000.[14] It is also in the range of the various prevalence reported by previous studies in Saudi Arabia and the Middle East which ranged from 0.3 to 2.19/1000 live births.[15-20] A systematic review carried out on the prevalence of NSOFC in Saudi Arabia and the Middle East discussed the reason behind the multiple reported NSOFC prevalence. They explained that having a hospital based studies in large cities with many primary centers and in a community like Saudi Arabia with multiple ethnic originality and with different and high consanguinity marriages could be the reason behind the several reports.[21]

Conclusion:

The prevalence of cleft lip and/or cleft palate is 1.9/1000 live births. The prevalence of CL is greater than CLP, and CP. CL was more frequent on the left side than on the right side. The prevalence of clefts was greater in males than females 1.8:1. Further research on the etiology of NSOFC and their relation to parental consanguinity is needed.

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Table 1: Distribution of NSOFC sub-phenotypes born or referred to MCH in 2010 and 2011 according to gender and sub-phenotype:

Phenotype N (%)	**Sub-Phenotype**	**Frequency of NSOFC in 2010 & 2011 (%)**		
		Male	**Female**	**Total**
CL 34 (43.5%)	Right incomplete CL	4 (8%)	1 (3.6%)	5 (6.4%)
	Right complete CL	2 (4%)	1 (3.6%)	3 (3.8%)
	Left incomplete CL	10 (20%)	4 (14.3%)	14 (18%)
	Left complete CL	7 (14%)	0	7 (9%)
	Bilateral incomplete CL	4 (8%)	0	4 (5.1%)
	Bilateral complete CL	0	1 (3.6%)	1 (1.3%)
CLP 26 (33.3%)	Right incomplete CLP	0	1 (3.6%)	1 (1.3%)
	Right complete CLP	2 (4%)	3 (10.7%)	5 (6.4%)
	Left incomplete CLP	1 (2%)	1 (3.6%)	2 (2.6%)
	Left complete CLP	4 (8%)	5 (17.9%)	9 (11.5%)
	Left incomplete bilateral CLP	1 (2%)	0	1 (1.3%)
	Bilateral incomplete CLP	2 (4%)	2 (7.1%)	4 (5.1%)
	Bilateral complete CLP	4 (8%)	0	4 (5.1%)
CP 18 (23%)	CP	9 (18%)	9 (32.1%)	18 (23%)
Total		50	28	78 (100%)

Table2: distribution of NSOFC according to consanguinity and cleft phenotype:

Cleft type	Consanguinity (%)		Total (100%)	P value
	Yes	No		
CL	22 (64.7%)	12 (35.3%)	34	0.734
CLP	17 (65.4%)	9 (34.6%)	26	
CP	13 (72.2%)	5 (27.8%)	18	
Total	52 (66.7%)	26 (33.3%)	78	

Table 3: distribution of NSOFC cases according to type of consanguinity

Cleft type	Type of consanguinity (%)			Total (100%)
	1st cousin	2nd cousin	Same tribe	
CL	13 (59.1%)	6 (27.3%)	3 (13.6%)	22
CLP	8(47.1%)	5 (29.4%)	4 (23.5%)	17
CP	9 (72.2%)	2 (27.8%)	2 (15.4%)	13
Total	30 (57.7%)	13 (25%)	9 (17.3%)	52

Table 4: NSOFC and distribution according to sub-phenotypes born in 2010 and 2011 in MCH:

Phenotype N (%)	Sub-phenotype	Male	Female	Frequency (%)
CL N=25 (46.3)	Right incomplete CL	3 (9.7)	1 (4.3)	4 (7.4)
	Right complete CL	2 (6.5)	1 (4.3)	3 (5.6)
	Left incomplete CL	5 (16)	3(13)	8 (14.8)
	Left complete CL	6 (19.3)	0	6 (11)
	Bilateral incomplete CL	3 (9.7)	0	3 (5.6)
	Bilateral complete CL	0	1 (4.3)	1 (1.9)
CLP N=19 (35.2)	Right complete CLP	2 (6.5)	3 (13)	5 (9.2)
	Left incomplete CLP	1 (3.2)	1 (4.3)	2 (3.7)
	Left complete CLP	2 (6.5)	5 (21.7)	7 (13)
	Bilateral incomplete CLP	0	2 (8.7)	2 (3.7)
	Bilateral complete CLP	3 (9.7)	0	3 (5.6)
CP N=10 (18.5)	CP	4 (12.9)	6 (26.1)	10 (18.5)
	Total	31	23	54