ANALYTIC RETROSPECTIVE STUDY OF CLEFT LIP & PALATE REFERRED TO PLASTIC UNIT IN BASRAH TEACHING HOSPITAL

Zuhair F. Fathallah

ABSTRACT

The data of this study of cleft lip & Palate are related to a sample of 638 patients who were born with this deformity and were referred to the plastic unit between1994-2003. These patients were evaluated in respect to age at presentation, family history, and type of deformity, sex ratio, laterality and clinical severity. All patients were operated on then followed up for a period of 5 months to 2.5 years. The results of the analysis of the data show that there is steady increase in the number of referred cases each year. The bulk of the orofacial defect is the isolated cleft lip, which account for 44.4% of the cases with male predominance except in cleft palate. Left sided cleft with or with out palate was found to be as twice common as right side cleft lip. The unilateral cleft is three times as bilateral; while in cleft lip and palate it's twice as common. The increase in the number of referred cases may reflect the increase in the birth rate and also increase in the number of cases due to increase environmental pollution.

INTRODUCTION

Cleft lip & palate are among the most frequent congenital deformities seen.[1] Clefting results when developing facial structures fail to fuse between the fourth and eighth weeks of gestation. [2] This failure is stimulated by genetic & environmental factors. The genes that cause clefting may be passed from either parent. A parent with a cleft has 5% chance of passing the trait. [3] If clefting is associated with a recognized genetic syndrome in which the genes are dominant rather than recessive the chance of inheritance become 50%. [3] Clefting may also result from environmental disruptions in development which may be triggered by: drugs as phenytoin,[4] retinoids [4] and steroids. [5] tobacco smoking by either parent[2,6] alcohol consumption,[4] maternal viral infection as rubella,[3] diet as folic acid deficiency. [4] This malformation, because of its unknown aetiological factors, is considered as multifactorial.[7] Falconer[8] in 1963 hypothesized that each case is unique, representing a combined liability or summation of risk factors. Once the composite liability exceeds a threshold a cleft lip & / or palate will occur. Myth and superstition are reported to accompany the birth of a child with orofacial cleft. [9] Here in our local community the condition is associated with the superstition that the pregnant woman might have seen a rabbit or a hare & the condition is called by lay people and even by doctors as “Rabbit or Hare Lip”. The total number of the cases studied is 638 who were admitted to the plastic unit. All the cases were included in the analysis for estimation of the incidence of cleft lip (CL), isolated cleft palate (CP), and cleft lip & palate (CLP). Also estimation of sex ratio, laterality, clinical severity and age at presentation. The striking features in this study, is the steady increase in the incidence with the years, this finding is proved worldwide. [3] It raises the question of increase environmental pollution during the early 90s in the south of Iraq. In good number of cases, patients were either living in a polluted area near the early nineties gulf war battlefield, or areas of low socio-economic status, where there is lack of drinking water and sanitation.

MATERIALS & METHODS

This study was carried out in Plastic Surgery unit at Basrah Teaching Hospital over a period of ten years. It’s a retrospective; analytic and observational study of 638 cases with cleft lip with or without palate and isolated cleft palate referred to the plastic unit in Basrah Teaching Hospital, which is the only plastic center in south of Iraq. The study includes all the cases referred to the unit between January 1994 and December 2003. These children were born to parents who are normally resident in and around Basrah, Amarrah and Nassyria provinces. They were assessed, operated on by a single surgeon

Zuhair F. Fathallah, MBChB, MSc, Department of Surgery, College of Medicine, University of Basrah, Iraq.

and followed up for the period of 5 month to 2.5 years.

RESULTS

Incidence: Over the 10 years there was increase numbers of cases with CL & Palate refereed to the plastic unit for surgery. In 1994 there were 43 patients (24 males & 19 females) admitted for treatment, while in 2003 the number has increased to 79 (42 males & 37 females) (Table-1). During the years of the study, males to female's ratio remained more or less constant, ranging from 0.8-1.5 with the mean of 1.28. In this series two coloured (Negro) patients were found with cleft lip that make the racial incidence of coloured to Caucasian is 2:636. The sample include 5 pairs of twins, two girl twins were both affected by cleft palate and the other three only one of the twins was affected.

Table 1. Number of cases admitted / year.

Year	Total	Males	Females	M:F ratio
1994	43	24	19	1.3
1995	35	21	14	1.5
1996	37	21	16	1.3
1997	42	24	18	1.3
1998	50	27	23	1.2
1999	72	32	40	0.8
2000	80	48	32	1.5
2001	103	58	45	1.2
2002	97	61	36	1.7
2003	79	42	37	1.14
Total	638	358	284	1.28

Type of deformity: Analysis of the data show that CLP found in 173 of the cases (27.1%), CL found in 283 cases (44.35 %) and CP found in 182 cases (28.5 %). The data show that the most common lip deformity is the left unilateral cleft lip with or without cleft palate (222 cases) followed by cleft palate in 182 then the bilateral in 123 and the last is right unilateral cleft lip with or without cleft palate in 111 cases, (Table-2).

Table 2. Types & number of Cleft lip & Palate deformity.

Type of deformity	Total	Males	Females
Cleft Lip – Palate (n = 283)			
Right unilateral	75	44	31
Left unilateral	141	76	65
Bilateral	67	38	29
Cleft lip + Palate (n = 173)			
Right unilateral	36	24	12
Left unilateral	81	53	28
Bilateral	56	34	22
Isolated Cleft Palate			
	182	89	93

Age at presentation: Patients were divided into 2 groups; those who presented for the operation before the age of two years, and those who were first seen after the age of two years. The first group forms 552 patients (86.5%), those presented after the age of two forms 86 patients (13.4%). (Table-3).

Table 3. Age of Presentation

Age	Total		Males		Females	
	No.	*%*	*No.*	*%*	*No.*	*%*
< 2y.	552	86.5	316	88.3	236	84.2
>2y.	86	13.4	42	11.7	44	15.7

Family history: This was positive in 27.1% of the cases (173 out of 638), which gave a history of some sort of orofacial cleft in relatives of first and second degree. About 5% of parents are not sure about that. In one third of the cases (212 cases) the parents are first cousins.

Sex ratio: In general, the M: F ratio is 358: 280 or 1.28:1. The sex ratio for each group is deferent, CL alone has a sex ratio of 1.26:1, CLP ratio is 1.8:1 and last CP is 0.95:1. Isolated CP is more common in females while CL & CLP are more common in males. (Table-4).

Table 4. Sex ratio in Cleft lip & Palate

Type of deformity	Male	Female	Sex ratio
Cleft lip	158	125	1.26
Cleft lip + palate	111	62	1.8
Cleft palate	89	93	0.95

Laterality: Means the side that is more common in each group. In CL the L: R ratio is 1.9:1 & CLP the ratio is 2.3:1. From both figures it's clear that the left side is more common in both sexes than right. (Table-5).

Table 5. Laterality in Cleft lip & Palate.

Type of deformity	Left	Right	L:R Ratio
Cleft lip	141	75	1.9
Cleft lip + palate	81	36	2.3
Cleft Lip ± palate	222	111	2

Clinical severity: It is the ratio between unilateral cleft lip & bilateral cleft lip. In isolated CL the unilateral to bilateral ratio is 3.2:1 while CLP the unilateral to bilateral ratio is 2.1:1. This indicates that in isolated CL the unilateral cleft is more common up to 3 times than bilateral. (Table-6).

Table 6. Clinical severity in Cleft lip & Palate.

Type of deformity	Unilat.	Bilat.	Ratio
Cleft lip	216	67	3.2
Cleft lip + palate	117	56	2.1
Cleft Lip ± palate	333	123	2.7

DISCUSSION

Incidence: The incidence of cleft lip & palate was calculated from a sample of 638 cases born with this anomaly referred to plastic unit outpatient. The incidence of cleft lip & palate is usually stable in given populations. The value of CL and / or Palate worldwide vary from 0.95-1.6, CL vary from 1.21-0.27.[10] Racial difference is important in determining the incidence e.g. coloured have low incidence while Japanese have high incidence. In this series there were 2 coloured out of 638 cases, Das et al[11] found that the incidence in white is 1.36/, While it is 0.54 in colored people. (Table-1).

Type of Deformity: The CL incidence form 44.4%. CLP forms 27.1% and CP 28.5%, if we compare these data with the series from Saudi Arabia, [12] which is 38%, 37.4% and 22.4%, compare with the series from north Ireland [13] the figures are 16%, 26% and 53% respectively, and the figures from Scotland [14] it's 25%, 30% and 45%. These 4 sets of data show that in this local area it is CL that makes the bulk of the OFC, while in British Isles for example it's CP, which is the main defect. (Table-2).

Age of presentation: The figures from (Table-3), show that 86.5% of all patients were presented early for the operation while 13.4% were delayed after 2 years, those who came late were nearly equal in sexes, in males 42 patients (11.7% of male cases) while in females 44 patients (15.7% of female cases), which indicate that there is no sex discrimination by the parents but its only a matter of negligence from the parents or due to poverty. Rajabian [15] reported that 79% of the patients presented between 1-18 months, it is nearly the same as this series finding which is 86.5%.

Family history: It is not an easy task to get information from the family; they regard any history of OFC in the first or second-degree relatives as a social stigma. In this series the family history is positive in 27.3% of the cases. The figures reported by Kumar et al [12] from Saudi Arabia are positive in 26.8%. These two sets of figures are the same. About 5% of parents (32 cases) were unable to give any information either they don't know or they don't like. In about 33% of the parents they were first cousins.

Sex ratio: A significant high sex difference was found with males' predominance, in CL (1.26:1)

and CLP (1.8:1). The more sever forms had higher values. While in isolated CP (0.95:1) females show predominance. This corresponds with findings of other authors [10, 14] where M: F in Czechoslovakian data is 1.37 & 1.1in Scottish data while in Singapore[16] and in China[17] its 1.33. (Table-4).

Laterality: Cleft on the left side is significantly more common in CL & CLP than the right. There is a universal agreement on this finding[6, 13,16], but right is more common in females. [13] In this series left side cleft is more common than the right in both sexes. It was suggested as a possible explanation that major blood vessels supplying the right side of the head of the fetus leave the aortic arch closer to the heart and more in line with blood flow, than those going to the left side, [18] (Table-5).

Clinical severity: The severity of cleft lip & palate is the ratio of unilateral cases to bilateral cases. In this series the ratio was 3.2 in CL and 2.1 in CLP (Table-6). This ratio means that the chance of having bilateral deformity in CLP is more than with CL alone. When these figures are compared with Czechoslovakian data, [10] which is 10.9 in CL and 2.5 in CLP, then these two sets are equal in CLP but widely different in CL. In this series the chance of having cleft lip and bilateral is high. The Danish data [15] is the same as Czechoslovakian data.

In conclusion, an average of 63.8 new cases with cleft lip & palate are expected to be presented every year, and the number is continuing to rise may be due to increase in rate of births or increase in the environmental pollution, if that added to the increase in cigarette smoking [2,6] by the parents or increase alcohol consumption [4] and maternal nutritional deficiency and consumption of drugs the net result will be more and more cases. Because of the complicated nature of this deformity as lip, palate and alveolus with or without deafness, difficulty in speech, ear infection and teeth deformity, the babies with cleft lip and palate require a multidisciplinary medical approach by the collaboration of Plastic surgeon, Orthodontist, Speech therapist, Pediatrician, nursing, Otolaryngologist and social & psycological services, this means increase spending from the health authority and the parents. The effort therefore have to be directed towards prevention of this deformity by research work by both Pediatricians and Gynecologists to identify the causes and find the measures to avoid this deformity weather its congenital, maternal or environmental.

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