

Prevalence of nonsyndromic oral clefts in a reference hospital in the state of Minas Gerais, Brazil, between 2000-2005

Prevalência de fissuras orais não-sindrômicas em um hospital de referência no estado de Minas Gerais, Brasil, entre 2000 e 2005

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Abstract: The aim of the present study was to analyze the prevalence of nonsyndromic oral clefts in children receiving treatment at the Center for the Rehabilitation of Craniofacial Anomalies, José do Rosário Vellano University, Alfenas, MG, Brazil. All the data for the epidemiological study was retrieved from the files of 126 pediatric patients with oral clefts without any additional malformation, who came to the center for treatment between 2000 and 2005. A predominance of clefts was observed in Caucasians, and the ratio of male to female was 1.3. Males were 2.57 times more affected by cleft lip and palate (CLP) than females. CLP with a prevalence of 39.68% and isolated cleft lip (CL) with a prevalence of 38.09% were the most common anomalies, followed by isolated cleft palate (CP; 22.23%). Complete and unilateral CLP (26.19%) presented the highest prevalence, followed by incomplete and unilateral CL (23.81%). The present study presents the experience of a reference hospital in the state of Minas Gerais; however, the real prevalence of oral clefts in Brazil is still unknown. Our findings differ from those of a few previous Brazilian reports because they suggest similar prevalences of CLP and CL, and a higher prevalence of CLP in Caucasian males.

Descriptors: Cleft palate; Cleft lip; Epidemiology.

Resumo: O objetivo do presente estudo foi analisar a prevalência de fissuras orais não-sindrômicas em crianças que receberam tratamento no Centro de Reabilitação de Anomalias Craniofaciais da Universidade José do Rosário Vellano, Alfenas, MG, Brasil. Todos os dados epidemiológicos deste estudo foram obtidos dos arquivos de 126 pacientes pediátricos com fissuras orais sem outra malformação adicional que compareceram ao Centro para tratamento entre 2000 e 2005. O predomínio das fissuras foi observado em caucasianos e a relação entre homens e mulheres foi de 1,3. Os homens foram 2,57 vezes mais atingidos por fissuras lábio-palatais (FLP) do que as mulheres. FLP com prevalência de 39,68% e fissuras labial isolada (FL) com prevalência de 38,09% foram as anomalias mais comuns, seguidas por fissuras palatinas isoladas (FP, 22,23%). As FLP completas e unilaterais (26,19%) foram as de maior prevalência, seguidas por FL incompletas e unilaterais (23,81%). O presente estudo mostrou a experiência de um Hospital de Referência em Minas Gerais; contudo, a verdadeira prevalência de fendas orais ainda é desconhecida. Nossos achados divergem dos de alguns poucos estudos anteriores por demonstrar prevalências similares de FLP e FL, e um acometimento maior de FLP em caucasianos do sexo masculino.

Descritores: Fenda palatina; Fenda labial; Epidemiologia.

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Introduction

Clefts of the lip and/or palate account for 65% of all head and neck anomalies, and can be found as an isolated finding or in association with other features, as part of syndromes.⁴ The etiology and pathogenesis of oral clefts, particularly of nonsyndromic origin, remain poorly understood. This is in part a reflection of the complexity and diversity of the molecular mechanisms involved during embryogenesis, with both genetic and environmental factors playing an influential role.⁸

Epidemiologic studies of isolated clefts, such as cleft lip (CL) and cleft palate (CP), or the combination of both (cleft lip-palate, CLP) have been conducted worldwide, often resulting in varying prevalence rates.⁷ Differences in geographic and ethnic distributions may explain some of the prevalence variations but not all. The incidence in European Caucasian populations ranges from 1 to 2.21 per 1,000 livebirths,⁶ among individuals with the same ethnical origin. In Brazil, there is a paucity of studies on oral clefts in children. The few published studies came from the Hospital for the Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru, SP, Brazil. Studies from this center revealed a prevalence of oral clefts of 0.19 per 1,000 livebirths,⁵ and a predominance of complete CLP in females.^{1,3} The Center for the Rehabilitation of Craniofacial Anomalies, University of Alfenas (José do Rosário Vellano University), Alfenas, MG, Brazil was established in 1992 and has attended more than 1,500 patients presenting craniofacial malformations, especially oral clefts. The purpose of this study was to analyze the prevalence of oral clefts in patients that received treatment in this hospital from 2000 to 2005.

Material and Methods

The study sample consisted of 126 children that were referred to the Center for the Rehabilitation of Craniofacial Anomalies, University of Alfenas (José do Rosário Vellano University), Alfenas, MG, Brazil, between the years 2000 and 2005. Clinical examination included classification of the cleft type according to the classification previously proposed by Spina *et al.*⁹ (1972). Clefts were classified in CL,

CP or CLP, and sub-classified in complete or incomplete and in unilateral or bilateral. Patients with previous history of surgical procedure for correction of the malformation were excluded from the study in order to avoid misclassification. Patients with recognizable syndromes or rare clefts were also excluded. The study protocol was approved by the Ethics in Research Committee, School of Dentistry, José do Rosário Vellano University. All patients's parents or guardians were informed about the study's purpose before they consented to participate. The chi-square test was performed considering 0.05 as statistically significant, and all types of variables (gender, race and types of clefts) were analyzed using their sub-classification.

Results

Table 1 describes the clinical features, according to gender and race, of the patients affected by nonsyndromic oral clefts of this study. From 126 children, 71 (56.35%) were males and 55 (43.65%) were females (M/F ratio 1:1.3). Oral clefts were found to be more frequent in Caucasians than in non-Caucasians (85.72% and 14.28% respectively). Gender was not a predictive factor for prevalence. As recommended by the World Health Organization (WHO), 85% of the patients received treatment before the age of one year. The other patients (15%) received treatment after two up to ten years of age. Interestingly, most patients were characterized as belonging to the lowest socioeconomic class. CLP accounted for the highest proportion of the cases (39.68%), followed by CL in 48 patients (38.09%) and CP in 28 patients (22.23%) (Table 2). The prev-

Table 1 - Distribution of the 126 patients affected by nonsyndromic oral clefts of this study according to gender and race.

Evaluated Variables		n	Prevalence (%)	Chi-Square
Gender	Male	71	56.35	$\chi^2 = 2.02$ $p = 0.15^*$
	Female	55	43.65	
Race	Caucasian	108	85.72	$\chi^2 = 64$ $p < 0.001^{**}$
	Non-Caucasian	18	14.28	

*non-significant difference. **significant difference.

alence of isolated CL and isolated CP was similar between genders, but CLP prevalence in males was 2.57 times higher than in females (Table 2). A heterogenous distribution was clearly evident as demonstrated by the Chi-Square test.

Complete and unilateral CLP (26.19%) was the most prevalent oral cleft, followed by incomplete and unilateral CL (23.81%) and incomplete CP (18.26%) (Table 3). Incomplete and unilateral CL was the most frequent type of cleft in this group, comprising 62.5% of the isolated clefts in the lip (Table 3). Furthermore, unilateral CLs were 7 times more frequent than bilateral CLs, whereas an incomplete involvement of the lip was 2.2 times more frequent than a complete involvement. Incomplete CP was 4.6 times more frequent than complete CP, accounting for 82.14% of the CP cases (Table 3). Unilateral clefts of the lip and palate simultaneously and completely were more frequent than the bilateral occurrence of this type of malformation (66% and 34% respectively) (Table 3). In table 3, all differences were significant.

Table 2 - Prevalence of nonsyndromic oral clefts in children according to gender.

	Male n (%)	Female n (%)	Total n (%)	Chi-Square
Cleft Lip-Palate	36 (72)	14 (28)	50 (39.68)	$\chi^2 = 7.04$ $p = 0.0296^*$
Cleft Lip	25 (52.08)	23 (47.92)	48 (38.09)	-
Cleft Palate	14 (50)	14 (50)	28 (22.23)	-

*significant difference.

Discussion

It is now well established that the several types of oral clefts are epidemiologically different, which is probably dependent on ethnicity.¹¹ Although a great number of epidemiological studies on oral clefts have been published around the world, there are few that have been carried out in Brazil. Furthermore, studies such as the present one provide insight into the prevalence of oral cleft of different population groups, which can be useful for government planning of new strategies for medical care of oral cleft patients.

The findings of the present study reveal that, of the 126 children with nonsyndromic oral clefts, the prevalence of CLP and isolated CL was quite similar, with 50 patients (39.68%) having CLP and 48 patients (38.09%) having CL. The remaining 28 patients (22.23%) had CP. In most published studies, the percentage of subjects with CLP has been higher compared to that of CL or CP alone, including the Brazilian studies.^{1,3} Barbosa *et al.*¹ (2003) described an incidence significantly higher of CLPs (65.5%) compared to that of isolated CLs (18.2%) and isolated CPs (16.3%). Similarly, another recent Brazilian study demonstrated a higher prevalence of complete CLP, either unilateral or bilateral, compared to that of isolated clefts.³ These differences may be due to the race blending between African-Brazilian and Caucasian individuals, which is very common in Brazil; however, a conclusion based on small samples in association with limited knowledge on the etiology of the oral clefts is tenuous.

Table 3 - Overall and group prevalence of the different types of oral clefts.

Type		n	Prevalence in the group (%)	Overall Prevalence (%)	Chi Square
Isolated Cleft Palate	Complete	5	17.86	3.97	$\chi^2 = 11.57$ $p < 0.001^*$
	Incomplete	23	82.14	18.26	
Isolated Cleft Lip	Complete Unilateral	12	25	9.52	$\chi^2 = 40.5$ $p < 0.001^*$
	Incomplete Unilateral	30	62.5	23.81	
	Complete Bilateral	3	6.25	2.38	
	Incomplete Bilateral	3	6.25	2.38	
Cleft Lip-Palate	Complete Unilateral	33	66	26.19	$\chi^2 = 5.12$ $p = 0.0237^*$
	Complete Bilateral	17	34	13.49	

*significant difference.

The study presented here also revealed that the unilateral involvement of the CLP is more frequent than the bilateral involvement, as well as the unilateral involvement in isolated CL. There was a significantly higher number of patients with incomplete CL, i. e. involvement of either lip or alveolar bone, compared to the number of patients with complete CL. Interestingly, Freitas *et al.*³ (2004) found that complete CL was the most frequent type of cleft in this group, comprising 20% of the clefts on the left side and 15.3% on the right side. In the group of patients with isolated CP, an incomplete involvement of either hard palate or soft palate was more prevalent than a complete involvement of both palates, a finding which is similar to that presented by Freitas *et al.*³ (2004) and other studies.^{8,10}

Gender distribution showed an overall male predominance (M/F ratio 1:1.3), which was significantly increased in the CLP group (M/F ratio 1:2.57). Furthermore, Caucasians represented more than 85% of the patients affected by nonsyndromic oral clefts in this study. A tendency towards the Caucasian male prevalence has been reported previously in several studies including a Brazilian one.^{2,3} On

the other hand, isolated CP has been described as significantly more prevalent in Brazilian females,^{1,3} which was not observed in our study.

Conclusions

In summary, the present study showed that complete and unilateral CLP was the most frequent oral cleft among a group of Brazilians treated in a reference hospital in the state of Minas Gerais, Brazil. Furthermore, the prevalences of CLP and isolated CL were very similar, but higher than that of isolated CP, and Caucasian males were more frequently affected than females. Our study also points to the need to establish a national cleft registry to ensure reliable recording of patients with these congenital anomalies.

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