

Prevalence of oral hemangioma, vascular malformation and varix in a Brazilian population

Prevalência de hemangioma, malformação vascular e variz de boca numa população brasileira

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Abstract: Hemangioma, vascular malformation and varix are benign vascular lesions, common in the head and neck regions. Studies about the prevalence of these lesions in the oral cavity are scarce. The aim of this study was to estimate the prevalence of and to obtain clinical data on oral hemangioma, vascular malformation and varix in a Brazilian population. Clinical data on those lesions were retrieved from the clinical forms from the files of the Oral Diagnosis Service, School of Dentistry, Federal University of Minas Gerais, Brazil, from 1992 to 2002. Descriptive analysis was performed. A total of 2,419 clinical forms in the 10-year period were evaluated, of which 154 (6.4%) cases were categorized as oral hemangioma, oral vascular malformation or oral varix. Oral varix was the most frequent lesion (65.6%). Females had more oral hemangioma and oral varix than males. Oral vascular malformation and oral varix were more prevalent in the 7th and 6th decades, respectively. Oral hemangioma and oral varix were more prevalent in the ventral surface of the tongue and oral vascular malformation, in the lips. Oral hemangioma was treated with sclerotherapy (54.5%), and vascular malformation was managed with sclerotherapy and surgery (19.4% each). The data of this study suggests that benign vascular lesions are unusual alterations on the oral mucosa and jaws.

Descriptors: Hemangioma; Peripheral vascular diseases, epidemiology; Blood vessels, abnormalities; Mouth mucosa.

Resumo: Hemangioma, malformação vascular e variz são lesões vasculares benignas comuns na região de cabeça e pescoço. Estudos sobre a prevalência dessas lesões em boca são escassos. O objetivo deste estudo foi estimar a prevalência e realizar um levantamento de dados clínicos de hemangioma, malformação vascular e variz de boca. Dados clínicos dessas lesões foram consultados em fichas clínicas do arquivo do Serviço de Diagnóstico Oral da Universidade Federal de Minas Gerais, Brasil, no período de 1992 a 2002. Uma análise descritiva foi realizada. Foram obtidas neste período de 10 anos 2.419 fichas clínicas. Cento e cinquenta e quatro (6,4%) casos foram categorizados como hemangioma, malformação vascular e variz de boca. Variz de boca foi a lesão mais freqüente (65,6%). Mulheres foram mais acometidas pelos hemangiomas e varizes de boca. Malformação vascular e variz de boca foram mais prevalentes na 7^a e 6^a décadas de vida, respectivamente. A localização prevalente do hemangioma e da variz de boca foi a superfície ventral da língua, e da malformação vascular, os lábios. Hemangioma de boca foi tratado com escleroterapia em 54,5% dos casos e a malformação vascular com escleroterapia e cirurgia em 19,4% cada. Este estudo sugere que lesões vasculares benignas são alterações incomuns na mucosa bucal e nos maxilares.

Descritores: Hemangioma; Doenças vasculares periféricas, epidemiologia; Vasos sanguíneos, anormalidades; Mucosa bucal.

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Introduction

Benign vascular lesions are a consequence of blood vessel abnormalities or endothelial cell proliferation.¹³ The International Society for the Study of Vascular Anomalies (ISSVA), in 1996, approved a classification system modified from the one proposed by Mulliken, Glowacki²⁰ (1982). The diseases were subdivided into (a) tumors: hemangioma (HEM), pyogenic granuloma, rapidly involuting congenital hemangioma, noninvoluting congenital hemangioma, hemangiopericytoma, tufted angioma and kaposiform hemangioendothelioma; and (b) vascular malformation (VM).⁸

HEM is a benign proliferation of endothelial cells. It is the most common neoplasm of the infancy. HEM frequently is not present at birth and develops in three phases: proliferating, involution, and involuted.⁸ It presents as a red macula, papule or nodule, depending on the congestion degree and on how deep it is in the tissue. Although HEM is a benign lesion, in some cases, it may lead to compression of surrounding structures, formation of fissures, ulcers or hemorrhages, and functional and aesthetic problems.^{7,13} Oral HEM can be found in the lips, tongue or buccal mucosa. It is more common in white female, in twins and in premature infants.^{7,11,20}

VM is considered an abnormality of the embryonic development, a structural anomaly. VM may be composed by arteries, veins and/or capillaries. Clinically, they are similar to HEM; however, they are always present at birth and grow as the patient physically develops.²⁰ It does not spontaneously regress, remaining stable throughout life. Frequently, bone involvement is present as a radiolucent, multilocular and well-circumscribed image.⁷ The few studies on benign oral vascular lesions frequently do not distinguish between oral HEM and VM, or regard oral VM as a histological type of HEM, known as arteriovenous hemangioma.²

The treatments for benign vascular lesions are sclerotherapy, systemic corticosteroids, interferon α , laser, embolization, cryotherapy, and surgery. Whether they should be followed-up or treated depends on the patient's age, and on lesion site and size.^{2,7,15,26}

Varix (VAR) is an acquired benign vascular lesion, generally asymptomatic, and does not require treatment.²⁷ Age is a predisposing factor, as well as tissue loosening and increased venous pressure. Oral VAR is rare in infants, but very common in old adults.¹⁶ Oral VAR is characterized as a red to purple papule or nodule, commonly found on the tongue, lip or cheek, mainly in the seventh decade of life. Also, there are few studies on epidemiological data on oral VAR.^{16,27}

The aim of this study was to estimate the prevalence of and to report clinical data on benign oral vascular lesions (HEM, VM and VAR) in a Brazilian population, and also to relate these data with that found in the literature.

Material and Methods

The protocol of this study was approved by the Committee of Ethics in Research, Federal University of Minas Gerais (UFMG, number 190/02). Clinical forms from 1992 to 2002 were retrieved from the files of the Oral Diagnosis Service, School of Dentistry, Federal University of Minas Gerais (FO-UFMG; Belo Horizonte, MG, Brazil) and evaluated.

The Oral Diagnosis Service, FO-UFMG, uses criteria to distinguish between oral HEM and oral VM, as described by Mulliken, Glowacki²⁰ (1982). Oral HEM is a red, asymptomatic and well-circumscribed lesion that develops during late fetal stages or in infancy, which grows quickly and generally spontaneously regresses. Histologically, it presents hypercellularity in the proliferating phase and fibrosis and diminished cellularity in the involuting phase. Oral VM is a red and asymptomatic lesion with undefined limits. It is present at birth and grows as the patient develops.²⁰ Oral VM is characterized by hypocellularity and vascular channels lined by flat mature endothelium. Oral VAR is characterized by a red to purple papule or nodule, commonly found in the tongue, lip or cheek, mainly in the seventh decade of life.^{16,27} Oral VAR is morphologically composed by one to three extensive and tortuous blood vessels lined by flat mature endothelium.

Descriptive analysis (absolute and relative frequencies) was performed using the software EPI-INFO.⁵ Patient information (sex, age and race) and

Table 1 - Data regarding the 154 cases of benign oral vascular lesions (FO-UFGM, 1992-2002).

		Oral hemangioma		Oral vascular malformation		Oral varix	
		n.	%	n.	%	n.	%
Cases		22	14.3%	31	20.1%	101	65.6%
Sex	Male	6	27.3%	15	48.4%	38	37.6%
	Female	16	72.7%	16	51.6%	63	62.4%
Age (decades)	0 - 10	1	4.5%	2	6.5%	0	–
	11 - 20	3	13.6%	1	3.2%	1	1%
	21 - 30	4	18.2%	1	3.2%	5	5%
	31 - 40	2	9.1%	4	12.9%	14	13.9%
	41 - 50	3	13.6%	3	9.7%	10	9.9%
	51 - 60	3	13.6%	7	22.6%	28	27.7%
	61 - 70	2	9.1%	9	29%	26	25.7%
	71 - 80	4	18.2%	2	6.5%	9	8.9%
	81 - 91	0	–	1	3.2%	8	7.9%
	N.I.	0	–	1	3.2%	0	–

N.I.: No information.

Table 2 - Data regarding the 154 cases of benign oral vascular lesions (FO-UFGM, 1992-2002).

		Oral hemangioma		Oral vascular malformation		Oral varix	
		n.	%	n.	%	n.	%
Race	White (Caucasian)	12	54.5%	13	41.9%	57	56.5%
	Non-white	2	9.1%	3	9.7%	6	5.9%
	N.I.	8	36.4%	15	48.4%	38	37.6%
Symptoms	Asymptomatic	17	77.2%	22	71%	89	88.1%
	Symptomatic	2	9.1%	4	12.9%	1	1%
	N.I.	3	13.6%	5	16.1%	11	10.9%
Localization	Upper lip	2	9.1%	8	25.8%	3	3%
	Lower lip	5	22.7%	6	19.4%	3	3%
	Floor of the mouth	0	–	0	–	9	8.9%
	Ventral surface of the tongue	7	31.8%	4	12.9%	77	76.2%
	Buccal mucosa	4	18.2%	7	22.6%	6	5.9%
	Others	4	18.2%	6	19.4%	2	2%
	N.I.	0	–	0	–	1	1%

N.I.: No information.

disease information (diagnosis, symptom, location, number, size and treatment) were obtained.

Results

A total of 2,419 clinical forms were evaluated in the studied period (ten years). Of these, 154 (6.4%) were oral HEM, VM or VAR. Oral HEM

was diagnosed in 22 cases (0.9%), oral VM, in 31 cases (1.3%), and oral VAR, in 101 cases (4.2%). Oral VAR was the most frequent lesion (101 cases, 65.6%), followed by oral VM (31 cases, 20.1%) and HEM (22 cases, 14.3%). Data regarding the 154 cases of benign oral vascular lesions are shown in Tables 1, 2 and 3.

		Oral hemangioma		Oral vascular malformation		Oral varix	
		n.	%	n.	%	n.	%
Number of lesions	Single	19	86.4%	25	80.6%	8	7.9%
	Multiple	2	9.1%	4	12.9%	57	56.4%
	N.I.	1	4.5%	2	6.5%	36	35.6%
Size (cm)	0.1 - 1	0	–	1	3.2%	0	–
	1.1 - 2	1	4.5%	2	6.5%	0	–
	2.1 - 3	0	–	1	3.2%	0	–
	3.1 - 4	3	13.6%	5	16.1%	0	–
	> 4	4	18.2%	1	3.2%	0	–
	N.I.	14	63.6%	21	67.7%	101	100%
Treatment	Sclerotherapy	12	54.5%	6	19.4%	3	3%
	Surgery	3	13.6%	6	19.4%	2	2%
	No treatment	0	–	6	19.4%	5	5%
	N.I.	7	31.8%	13	41.9%	91	90%

N.I.: No information.

Table 3 - Data regarding the 154 cases of benign oral vascular lesions (FO-UFGM, 1992-2002).

Discussion

The clinical parameters from the classification of Mulliken, Glowacki²⁰ (1982) were used to distinguish oral HEM from VM in our sample. Since the majority of studies on benign oral vascular lesions do not use this classification, differences in epidemiological data on these diseases are common.^{9,17,21,26} However, since oral HEM is a neoplasm and oral VM is a disturbance of development, the distinction between these two entities is important for the knowledge of the clinical behavior and management of these illnesses.^{7,20} The use of the Mulliken, Glowacki²⁰ (1982) classification for benign oral vascular lesions has also standardized the studies and epidemiological data on these lesions.

It was observed that benign oral vascular lesions represented 6.4% of all the diseases diagnosed in the Oral Diagnosis Service, FO-UFGM, and that oral VAR was the most prevalent lesion. Jainkittivong *et al.*¹⁴ (2002) observed that oral VAR represented 59.6% of the oral mucosa conditions diagnosed in patients older than 60 years. Kovac-Kovacic, Skaleric¹⁷ (2000) found a relative frequency of 16.2% for oral VAR, with higher frequency (93%) in patients in their 7th and 8th decades of life. Few studies and with variable relative frequency of oral HEM and VM are found in literature. In the study of Corbet

*et al.*⁶ (1994), oral HEM represented 2% of the lesions of the oral mucosa in patients with ages ranging from 65 to 74 years. Al-Khateeb *et al.*¹ (2003), in a study with infants, reported a relative frequency of 0.9% for HEM. Paltiel *et al.*²³ (2000) reported on the relative frequency of HEM and VM, and found that HEM was predominant in 53% and VM, in 56% of the cases. In our study, oral VM was more frequent than oral HEM. Unlike Paltiel *et al.*²³ (2000), our study was performed in a sample of jaw diseases.

Oral HEM and VAR were more frequent in females, as shown by the studies of Ettinger, Manderson¹⁰ (1974), and Donnelly *et al.*⁷ (2000). Jackson *et al.*¹³ (1993) observed a female : male ratio of 4:1 in patients with HEM. Jackson *et al.*¹³ (1993) and Barrett, Speight² (2000) observed that VM affects equally females and males, as in our study.

Oral VAR was more frequently diagnosed in patients in their 6th and 7th decades of life. Ettinger, Manderson¹⁰ (1974) also observed a higher occurrence of oral VAR as age increased, stating that, with it, tissue loosening occurs, which is an important factor in the development of oral VAR. The incidence of this alteration in our study was 70.2% in patients older than 60 years, similar to that observed by Bean^{3,4} (1952; 1956), and Miles¹⁸ (1972).

Otherwise, according to Kovac-Kovacic and Skalecic¹⁷ (2000), oral VAR is more common in patients between 51 and 60 years.

Jackson *et al.*¹³ (1993) observed that HEM is the most common benign neoplasm in infancy, the majority arising between the 1st and the 4th week of life. At five and seven years of age, respectively 50% and 70% of the infants no longer have their lesions. This is confirmed by the studies of Mulliken, Glowacki²⁰ (1982), and Mulliken¹⁹ (1992). VM is present at birth, grows together with the patient, and does not spontaneously regress.^{19,20} Hence, the clinical behavior of HEM and VM may explain the broad age range of these alterations found in our study, since our patients sought care in a later moment of their lives. Moreover, previous studies^{13,19,20} analyzed HEM and VM located in other anatomic regions, in which functional and/or aesthetic problems are more pronounced than in the jaws.

Previous studies did not refer to the race of the patients when analyzing benign vascular lesions^{7,10,13}. In our sample, all three lesions were more frequent in Caucasian patients. Parra *et al.*²⁴ (2003) observed that in the Brazilian population, approximately 39% of the population has European genes, 33% has Amerindian genes, and 28% has African genes. In addition, the Brazilian population census of 2000¹² showed that 54% of the population is Caucasian. Hence, the higher prevalence of benign oral vascular lesions in Caucasian Brazilians is in accordance with the country's race distribution.

Jackson *et al.*¹³ (1993) described HEM and VM as asymptomatic lesions. In our analysis, both alterations were also asymptomatic. Regarding oral VAR, studies in the literature do not describe symptoms.^{10,27} However, we observed that oral VAR are predominantly asymptomatic.

Oral VAR was located more frequently in the ventral surface of the tongue. Oral VM was more frequent in the upper lip, buccal mucosa and lower lip. Also, Barrett, Speight² (2000) observed that oral VM is more frequent in the lips and buccal mucosa. Oral HEM was more common in the ven-

tral surface of the tongue, followed by the lower lip and buccal mucosa. Nguyen *et al.*²² (2004) observed that among HEM lesions of the head and neck, oral HEM was mostly located in the buccal mucosa.

As to the number of lesions, Takahashi *et al.*²⁸ (1994) reported that oral HEM is a single lesion. Barrett, Speight² (2000) stated that oral VM is always a single disease without systemic implication, which is also true in our study. In contrast, oral VAR presented predominantly as multiple lesions, as in the studies by Kleinman¹⁶ (1967) and Ettinger, Manderson¹⁰ (1974).

Benign oral vascular lesions may be treated by sclerotherapy, systemic corticosteroids, interferon α , laser, embolization, cryotherapy, and surgery. Management and treatment decisions depend on the patient's age, and on the lesion's site and size.^{15,25} Sadeghi, Gingrass²⁵ (1989) stated that surgery is not indicated for bigger lesions due to post-surgical hemorrhages. In such cases, arterial embolization is satisfactory.²⁵ Satisfactory results are seen with sclerotherapy in the treatment of small benign vascular lesions, including oral diseases.¹⁵ In our sample, oral HEM and VM were treated with sclerotherapy or surgery. For oral VAR, treatment was carried out in five cases, because lesions were single and with unusual localization (lips and buccal mucosa). Moreover, they were satisfactorily treated by sclerotherapy or surgery.

Conclusions

Benign oral vascular lesions are unusual alterations on the oral mucosa and jaws.

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