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CASE REPORT

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Benign fibrous histiocytoma of the tongue: a case report

Histiocitoma fibroso benigno em língua: relato de caso

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ABSTRACT

The fibrous histiocytoma is a soft tissue neoplasm that affects the dermis and the subcutaneous tissue, rarely is found in the oral cavity and perioral regions, and is originated from the proliferation of fibroblasts and histiocytes. The objective of this paper is to report a case of Benign Fibrous Histiocytoma in a 30-year-old male patient, complaining of a painless nodule in the tongue for about six months. With diagnostic clinical hypotheses of Fibrous Hyperplasia, Neurofibroma, Traumatic Neuroma, Fibrous Histiocytoma, Granular Cell Tumor or Ectomesenchymal Chondromyxoid Tumor a excisional biopsy was performed. The histopathological examination revealed a non-encapsulated proliferation of spindle cells with some giant multinucleated cells in the periphery of the lesion. Immunohistochemical reactions were performed, staining only for vimentin in the spindle cells and for CD68 in the multinucleated giant cells. According to these characteristics, the final diagnosis was Benign Fibrous Histiocytoma. The correct diagnosis of spindle shaped cell neoplasia must be performed with the aid of histopathological analysis and immunohistochemistry, mainly because the morphological similarities with other benign and malignant lesions.

KEYWORDS

Fibroblasts; Benign fibrous histiocytoma; Malignant fibrous histiocytoma; Histiocytes, Tongue diseases.

RESUMO

O Histiocitoma Fibroso é uma neoplasia de partes moles que acomete a derme e o tecido subcutâneo, raramente é encontrado na cavidade oral e regiões periorais, e tem origem a partir da proliferação de fibroblastos ou histiócitos. O objetivo deste artigo é relatar um caso de Histiocitoma Fibroso Benigno em um paciente masculino, 30 anos de idade, com um nódulo indolor, bem delimitado, com duração de cerca de seis meses, localizado no dorso anterior da língua. Com as hipóteses clínicas diagnósticas de Hiperplasia Fibrosa, Neurofibroma, Neuroma Traumático, Histiocitoma Fibroso, Tumor de Células Granulares e Tumor Condromixoide Ectomesenquimal uma biópsia foi realizada sob anestesia local e a lesão foi fixada em formol a 10% e enviada para análise histopatológica. O exame histopatológico revelou uma proliferação não-encapsulada de células fusiformes com algumas células gigantes multinucleadas na periferia da lesão. A marcação imunohistoquímica foi positiva para CD68 nas células gigantes multinucleadas e para vimentina nas células fusiformes. O diagnóstico final foi de Histiocitoma Fibroso Benigno. Para um diagnóstico correto, este deve ser feito correlacionando características clínicas, análise histopatológica e imunohistoquímica devido à similaridade microscópica do Histiocitoma Fibroso com outras lesões com aspecto fusocelular, assim como similaridade clínica com outras lesões benignas e malignas.

PALAVRAS-CHAVE

Fibroblastos; Histiocitoma fibroso benigno; Histiocitoma fibroso maligno; Histiócitos; Lesões em língua.

LITERATURE REVIEW

Benign fibrous histiocytoma (BFH) is a rare neoplasm of soft tissue, which affects the dermis and subcutaneous tissue, has origin as from the fibroblasts and histiocytes. [1] It was first described by Stout and Lattes in 1967. [2] Before 1960, the BFH was not considered a distinct clinic-pathological entity. But, with electronic microscopy and immunohistochemistry advancement, it was possible to differentiate this diagnosis. [3] Per present a variable nature this type of neoplasm has many synonyms as: Nodular Fibrosis Subepidermic, Sclerosing Hemangioma and Simple Fibroid when it involves the skin. [4]

The BFH occurs most frequently on the skin surface of the extremities, however can develop in deeper tissues, in tissue of the oral mucosa or even in bone. [5] This neoplasm must be differentiated of Malignant Fibrous Histiocytoma that often presents a more aggressive course, can present recurrences and metastases. [6]

Little cases of HFB occurs in the oral cavity and perioral regions. The intraoral sites most commonly of involved soft tissue are the gums, lips lower and upper, soft palate, floor of the mouth and tongue being one of the least reported locations. [4]

This type of neoplasm is reported at any age with predominance during the third and fourth decades of life. Its distribution by gender varies between different populations. The clinical features of oral HFB are a painless solitary tumor, slow growth, 2-3 cm to over 10 cm, along a period of several months. [7] The symptoms are specifically caused by interference with the normal anatomy and physiology of the area in which it appears, when the mass is located on the tongue difficulty speaking may be present, for example. [3,8]

CASE REPORT

Male patient, 30 years of age, was referred for evaluation of a nodule in the tongue for the last six months. During the anamnesis he did not report any symptoms such pain or difficulty to speak. The physical examination revealed a small, asymptomatic and sessile white nodule in the anterior dorsum of the tongue (Figure 1). The diagnostic hyphoteses was fibrous Hyperplasia, Neurofibroma, Traumatic Neuroma, Fibrous Histiocytoma, Granular Cell Tumor and Ectomesenchymal Chondromyxoid Tumor. An excisional biopsy was performed under local anesthesia and the lesion was fixed on formaldehyde 10% and sent for histopathological analysis.

The histopathological examination revealed a non-encapsulated proliferation of spindle-shaped cells somewhat disposed in a storiform pattern. In addition, some giant multinucleated cells occupied the periphery of the lesion (Figure 2 A,B,C and D). Immunohistochemistry reactions were performed for cytokeratin AE1/AE3, vimentin, S100, smooth muscle actin, desmin and CD68, being positive only for CD68 in the giant multinucleated cells and for vimentin in the spindle-shaped cells (Figure 3 A and B). According to clinical, histopathological and immunohistochemical analysis, it was established a final diagnosis of Benign Fibrous Histiocytoma, once there was not any sign of malignant changes. In a follow up of 2 years, no evidence of recurrence was observed.

Figure 1 - Clinical Aspects: intraoral exam revealing a white nodule in the anterior dorsum of the tongue.

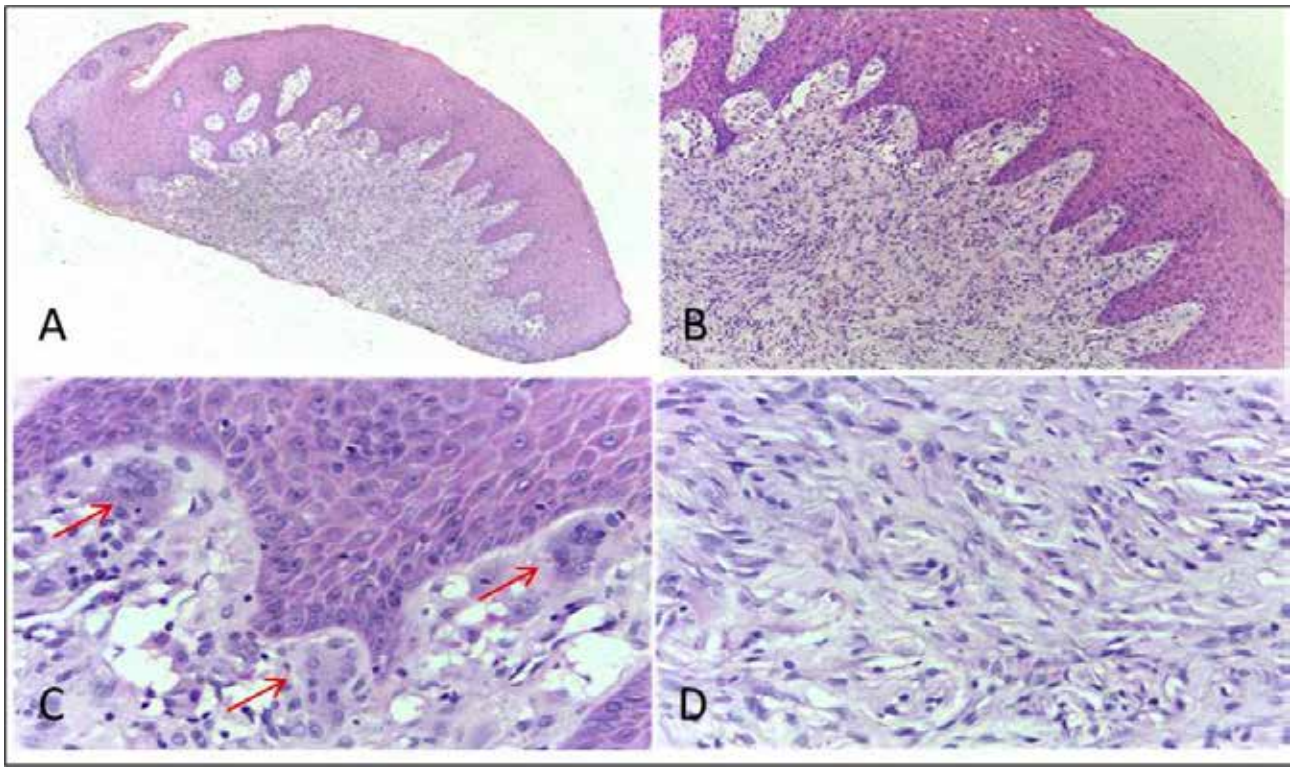

Figure 2 - Histopathological characteristics: A- General aspect in larger magnification (HE – 40 x); B- Subepithelial proliferation of spindle cells (HE – 100x); C- Presence of giant multinucleated cells on the periphery of the lesion (HE – 400 x); D- Spindle cells in a storiform arrangement (HE – 400 x).

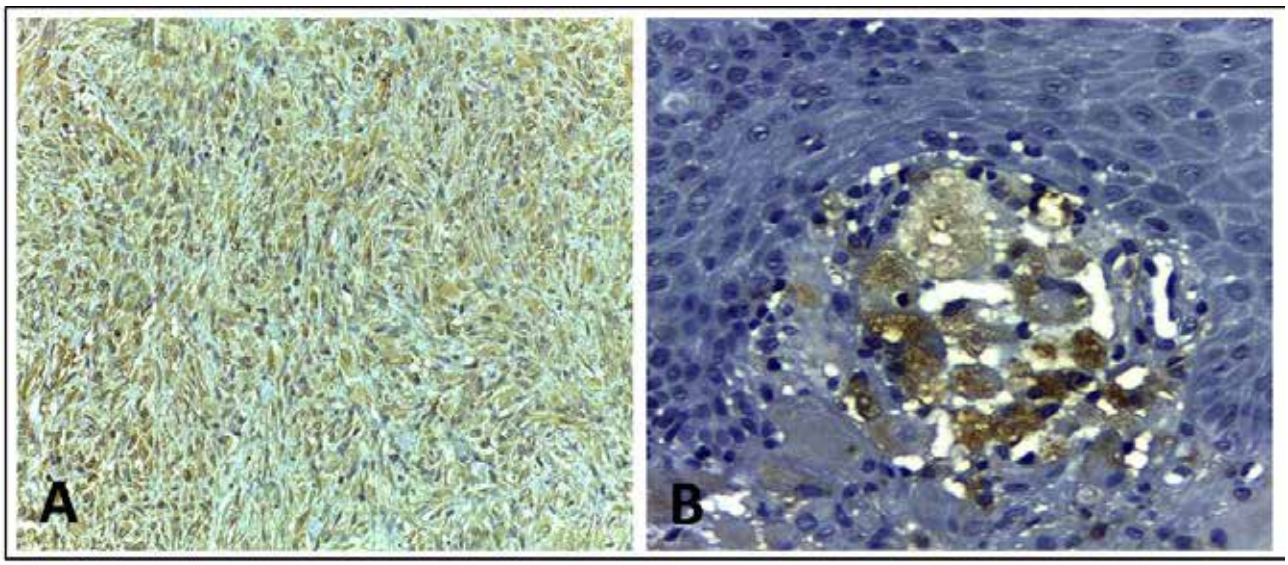

Figure 3 - Immunohistochemical Aspects: A- Positive staining for vimentin in spindle cells (Immunoperoxidase, 200 x); B- Positive staining for CD68 in multinucleated giant cells (Immunoperoxidase, 400 x).

DISCUSSION

The etiology of Fibrous Histiocytoma is not well established, is questionable whether it is a reactive proliferation to an inflammatory stimulus or neoplastic process. [4] Some cases are reported after local injuries, such as trauma, insect bites or folliculitis, strengthening the conception of inflammatory origin. MacLeod and Jones [9] reported a case of oral HFB in lip than occurred after continuous trauma to bite. In opposition, some examples support the idea clonal expansion, strengthening the conception of neoplastic origin. [4]

Due to uncertainty about the natural history of fibrohistiocytic lesions, the HFB was not identified as a distinct clinical entity until years 1960. [3] Still today, this type of lesion often can be confused clinically with other benign tumors commonly found in the oral cavity, such as fibroma or lipoma. [10] It is probably that there are also small cases, as the present one, not diagnosed because the patient or the clinical dentist even noticed or because the diagnosis is performed only based on the clinical exam.

It is a lesion with a predilection for skin of the extremities and areas exposed to the sun, but can affect bones, deep tissues and the oral cavity. [1,5,11] For the best of our knowledge, there are 49 cases of BFH involving the oral cavity previously reported in the English-language literature (Table I). Gender distribution was similar and included 26 female and 22 male (54.1% and 45.8 %, respectively). Age ranging from 6 months to 80 years. BFH mainly affected mandible (14 cases, 28%) and buccal mucosa (10 cases, 20%), Of the 49 cases, 7 were located on the tongue, as the case described here. Oral BFHs are usually present as nodular painless masses, but in some cases, they can interfere and cause discomfort in speaking. [12]

Histologically the HFB lesions are similar independently the site on what emerge showing spindle cells similar to fibroblasts and also histiocytes. The findings of Pandey et al. [13] are equivalent to this study, wherein was described a case of HFB in to tongue what microscopically showed spindle cells and histiocytes. Jo et al. [10] described the same pattern storiform of spindle cells in a buccal mucosa lesion. Even intra osseous lesions in the mandible, described by De-ming et al. [14] and Shoor et al. [15] revealed spindle cells and giant cells with positive staining for vimentin and CD68 respectively. Immunohistochemistry for CD68 can be used in these cases because it acts by marking the various cells of the macrophage lineage including monocytes, histiocytes, giant cells and osteoclasts. [15] Lesions such as Nodular Fasciitis, Fibrous Tumor Solitaire, Neurofibroma and Dermatofibroma are the main microscopic differential diagnosis for BFH. [11]

BFH should be differentiated from Malignant Fibrous Histiocytoma, that microscopically presents elevated presence of mitotic activity, atypical cells, cellular pleomorphism, prominent areas of hemorrhage and necrosis. Tanaka et al. [16] described a case of malignant transformation of Fibrous Histiocytoma in mandible, shows atypical spindle cells, giant cells with pleomorphic nucleus and positive immunohistochemistry results for cell proliferation markers such as Ki-67 and also to the p53, suggesting malignant transformation. This type of transformation occurs rarely, in approximately 1% of all BFHs. [16]

Treatment for oral BFH is complete excision of the lesion, the prognosis is excellent, only 5 to 10% are reported recurrence in cases of tumors of large dimensions or when the excision is incompletely. [10] Metastases are not reported in benign lesions and treatment with radiotherapy and chemotherapy have no indication in these cases. [13]

Table 1 - Summary of benign fibrous histiocytoma of the oral region reported in the English-language literature

Authors	Year of publication	Location	Gender	Age
Hoffman et al.(17)	1981	Buccal Mucosa Mandible Gum	M F	08 12
Cale et al. (18)	1989	Maxilla	M	13
MacLeod and Jones (9)	1992	Lower Lip	F	22
Gray et al. (19)	1992	Upper Lip Buccal Mucosa Buccal Mucosa Body of Mandible Tongue Tongue Buccal Mucosa Lower Lip Maxillary Vestibule Buccal Mucosa Mandibular Ridge Mandibular Vestibule Buccal Mucosa Mandibular Vestibule Body of Mandible Cortex Mandible Mandibular Vestibule Maxillary Gum	M M M M F F F F M F F M F F F F F F	45 42 65 59 37 50 71 45 49 70 12 60 68 - 59 - 66 37
Hong et al. (20)	1999	Floor of the mouth	F	74
Femiano et al. (21)	2001	Buccal Mucosa	M	32
Ide an Kusama (22)	2002	Lower Alveolar Ridge	F	50
Yamada et al. (23)	2002	Upper Lip Lower Lip	M M	6 months 52
Alves et al. (10)	2003	Buccal Mucosa	F	26
Heo et al. (24)	2004	Mandible	M	42
Kishinoet al. (25)	2005	Mandible	F	49
Katagiri et al. (26)	2008	Condylar Process	M	48
Menditti et al. (27)	2009	Mandibular Vestibule Tongue	M M	44 34
Giovani et al. (28)	2010	Buccal Mucosa	M	36
Tanaka et al. (16)	2011	Mandible	M	80
López-Jornet et al. (29)	2011	Tongue	F	08
De-ming et al. (14)	2012	Mandible	M	31
Rullo et al. (30)	2012	Buccal mucosa Tongue	M -	42 9 months
Bindhu et al. (31)	2012	Palate	F	20
Rajathi et al. (32)	2013	Mandibular Gum	M	23
Pandey et al. (13)	2013	Tongue	M	26
Priya et al. (12)	2013	Tongue	F	30
A.Gaffar et al. (33)	2014	Mandibular Gum	M	12
Saluja et al. (34)	2014	Maxilla	F	23
Gupta et al. (35)	2014	Maxilla	F	19
Dias et al. (36)	2015	Lower Lip	F	07
Shoor et al. (15)	2015	Mandible	F	30
Jo et al. (10)	2015	Buccal Mucosa	F	36

CONCLUSION

In summary, BFH is a rare neoplasm in the oral cavity, only with the clinical and histopathological features is not possible to establish the diagnosis. It is necessary performing immunohistochemical reactions for determining the origin of the cells, since many others neoplasm both benign and malignant, can present similar morphological characteristics.

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