ATYPICAL BENIGN FIBROHISTIOCYTOMA IN THE ORAL CAVITY: A CASE REPORT.

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ABSTRACT

The benign fibrous histiocytoma (BFH) is a pseudo-tumoral lesion, common in skin and with rare presentation in the mouth. Although its etiology is unclear, it is associated with trauma, sun exposure or chronic infections. A mother attends with his 10 years old son, consulting for a lingual swelling with an erythematous halo and ulcerated interfering when chewing occurs. An incisional biopsy was performed that resulted in benign fibrous histiocytoma. Subsequent excisional biopsy confirms the diagnosis. A review of the literature on the issue was done, the most important clinical characteristics are presented and the relevance of different immunohistochemistry as Ki67, CD68 and smooth muscle actin in the context are discussed for diagnosis and define appropriate treatment of the lesion.

INTRODUCTION: THE BENIGN FIBROUS HISTIOCYTOMA (BFH)

Is a pseudotumoral lesion with fibroblastic and histiocytic differentiations that can be present in benign or malignant form, the malignant variant can affect soft and hard tissues. Its cellular origin is uncertain; its appearance is associated with trauma, sun exposure[1] or chronic infections.

It is common on the skin (extremities) and occurring anywhere on the body, including the viscera or skeleton. Is extremely rare entity in the oral cavity, and it can appear anywhere covered with mucosa, preferring the buccal and vestibular mucosa.[2,3,4,5]

It is most common in adults of middle and old ages. Its intraoral variety has a slight predilection for women aged 30 to 40, rarely affecting young people.[6]

The BFH presents as an increase in volume with painless growth, without associated lymph node involvement.[5,6] It is detected when they interfere with function, due to displacement, interposition or compression.[7]

Because of the highly nonspecific symptoms, it can be confused with benign lesions such as fibromas or lipomas, or malignant when presenting a painless ulcer for a long period.

The clinical study can guide a diagnostic hypothesis, but its diagnosis is eminently histopathological, and is crucial for an adequate surgical treatment.[8]

Although rare in the oral cavity, it is a differential diagnosis in fibrous lesion of the oral mucosa.

CASE DESCRIPTION

A 10-year-old male patient without systemic history presented at the Finis Terrae University Faculty of Dentistry. The patient's mother notices an increased tongue volume and difficulty chewing.

Clinically, a tumor lesion is observed on the left edge of the lingual dorsum of approximately 20x10 mm with an erythematous halo and ulcerated in its most prominent portion, asymptomatic and with one month of evolution. **Fig. 1.**

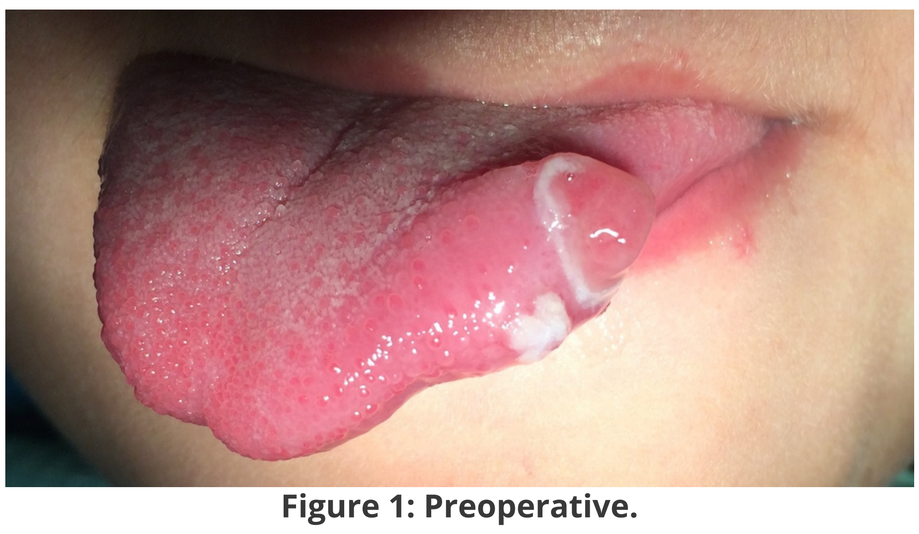

Figure 1: Preoperative.

An incisional biopsy is performed. Hematoxylin-eosin histopathology is not conclusive and is complemented with immunohistochemical tests.

The smooth muscle actin test shows about 80% positivity. Ki67 staining shows 10% cell proliferation. CD68 staining shows positive giant cells. Other positive mononuclear cells, close to 60%.

The final diagnosis was "Atypical benign fibrohistiocytoma in the oral cavity". The pathologist suggests total excision of the lesion with a safety margin, given the presence of cellular activity. **Fig. 2**

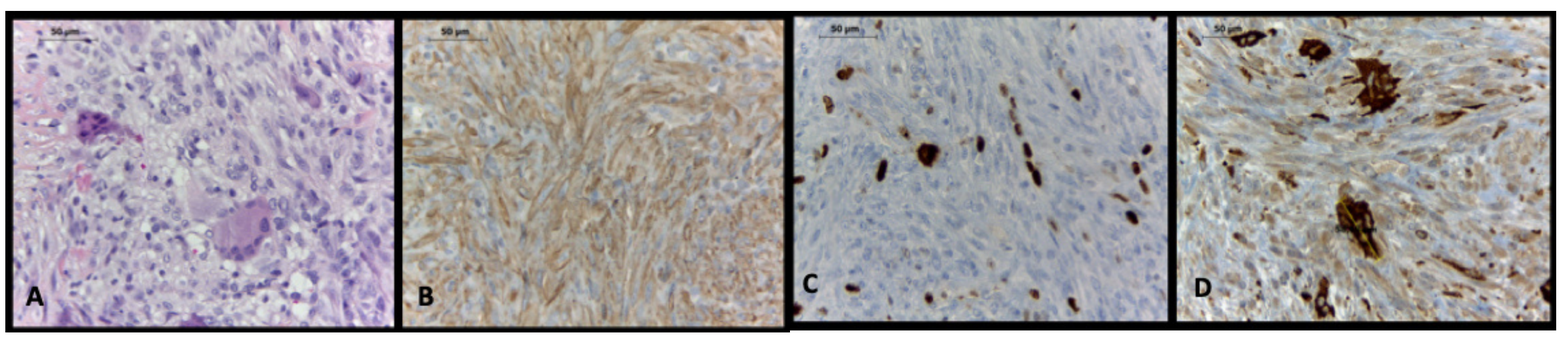

Figure 2: A) Hematoxylin-Eosin staining. B) Smooth muscle actin. C) Ki-67 staining. D) CD-68 staining.

In the Maxillofacial Surgery service of the San Juan de Dios Hospital, exeresis was performed under general anesthesia with a safety margin, which, although it was well differentiated, did not establish a clear limit with the muscular plane.

Histopathology subsequently confirmed the initial diagnosis. **Fig. 3.** At 45 days after the surgery, there is no evidence of recurrence. Lingual function contributes to anatomical remodeling. **Fig. 4.**

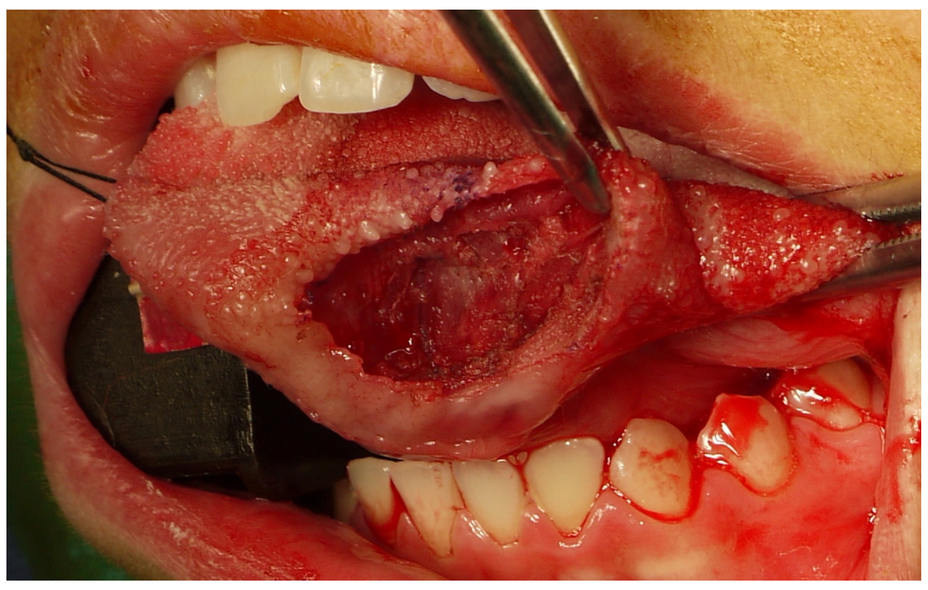

Figure 3: Excision with margin of safety.

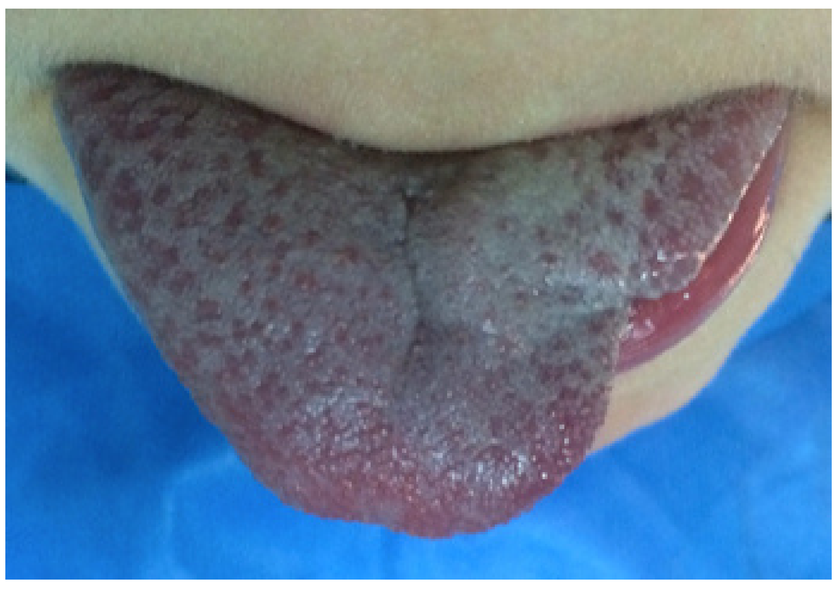

Figure 4: Postoperative.

DISCUSSION

It is difficult fot the pathologist to diagnose, both histologically and morphologically it resembles other soft tissue tumor lesions[2,3] and we can find coexistence of multiple cell types, predominantly fibroblasts and histiocytes, occasionally multinucleated giant cells, xanthomatous cells with lipid content and lymphocytes are observed. Fibroblasts have a fused shape with a pattern in the form of small bundles.

The sample does not usually contain cellular atypia, nor many mitoses. The stroma is densely fibrous and may show hyaline to myxoid changes in some areas. Smooth muscle actin is useful to identify and observe the pattern of fused cells,[9] helping to establish the type of lesion, as in this case.

According to the literature,[2,3,7] a large number of biopsied specimens show positive staining by immunohistochemical techniques with vitamin and CD-68, as in this case; however, the positive reaction to certain markers can vary over time. Ki-67 provides key information on growth (aggressiveness) as it is a specific marker for cell proliferation, CD-68 guides the diagnosis as it is marker for histiocytes.[7]

Although cell proliferation in BFH is usually minimal to null, this case showed a positive reaction of about 10% of the sample to Ki-76.[9] Together with the atypical location and the rich vascular supply of the area, added to the early presentation, it was determined to carry out the total excision of the lesion with a safety margin, despite the benign nature of classic BFH.

CONFLICT OF INTEREST: None

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