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Fibrous histiocytoma of the tongue: A case report

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Case Report

Abstract

BACKGROUND AND AIM: Benign fibrous histiocytoma (BFH) is a rare lesion in the head and neck with a slow, single, and painless growth that consists of fibroblasts and histiocytes. In this study, a BFH case was reported and examined from clinical, microscopic, and immunohistochemical aspects.

CASE REPORT: A 36-year-old man with a red nodule on the dorsal surface of his tongue was referred to the oral medicine department. The appearance of this nodule was similar to the adjacent tissue in its surface. According to the same microscopic view of this lesion with other soft tissue tumors, immunohistochemistry test confirmed the diagnosis. It was treated with en-bloc surgical resection. In four follow-ups up to one year, there was no recurrence. Considering the results of these cases and comparing them with other cases, although there is a slim chance of recurrence in one year, follow up is recommended.

CONCLUSION: Clinical view of FH is not characteristic and tumors with microscopic spindle-shaped appearance are challenging in diagnosis. IHC is obligatory to reach a prompt diagnosis and due to recurrence, follow-up is recommended.

KEYWORDS: Histiocytoma; Benign Fibrous; Tumors; Tongue Disease; Oral Cavity

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Fibrous histiocytoma (FH) is a tumor with mesenchymal origin.[1] FH is divided into benign and malignant categories.[2] The malignant type describes a soft tissue sarcoma known as a histiocytic tumor and fibroblasts with malignant potential, most of which being in the upper and lower limbs, orbit, pelvis, knee, head, and neck.[3-5] Through electronic microscopes and immunohistochemistry (IHC) experiments, the benign type is easily distinguished from the malignant one.[6]

Some of the assumptions about cellular origin are considered as fibroblastic and histiocytic, and others have considered a dendrocyte origin of the cell based on the presence of factor XIIIa.[7,8]

Benign fibrous histiocytoma (BFH) may be cutaneous or non-cutaneous, with the cutaneous type usually found in the skin exposed to sunlight. However, the non-cutaneous BFH only contains 1% of benign lesions that are commonly found in the tissues of the lower extremities (50%) and less in the upper extremities (20%).[9] BFH is divided into superficial and deep forms, with the deep form being very rare and involving subcutaneous tissues. This type of BFH is most common in lower extremities and less in the head and neck.[3,10] BFH is rare in the oral cavity and it was reported in buccal mucosa, tongue, gingiva, alveolar mandibular ridge, maxilla, upper and lower lip, soft palate, and floor of the mouth.[9]

BFH is a painless tumor with a slow growth between 2-3 to 10 cm over a period of

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several months.[2] The symptoms of this lesion occur due to the interference with normal anatomy and physiological functions, for example, difficulty in speech due to masses on the tongue.[11,12]

Its treatment is en-bloc surgical resection. Its prognosis is good and there is no possibility of recurrence in the mouth unless the resection is incomplete and no metastasis has been reported. However, follow-up is recommended for these patients.[3,7,13-15] The purpose of this study is to report a case of BFH in the tongue of the 36-year-old man.

Case Report

The 36-year-old male patient, visited by an otorhinolaryngologist with a feeling of discomfort and firmness in the central region of the dorsum of his tongue. He had a history of about 20 days accompanied by discomfort and interference with speech, without any pain or burning sensation. At the time of examination, a firm well circumscribed red nodule was observed in the dorsum of the tongue with a smooth surface (Figure 1). There was no history of systemic illness and the patient was a non-smoker with no history of alcohol consumption.

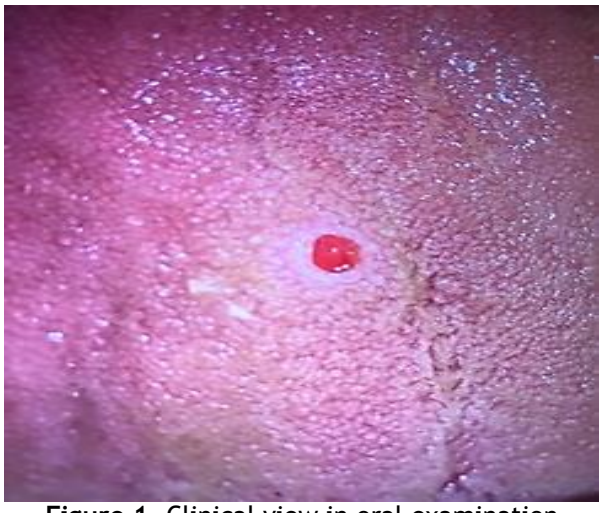

Figure 1. Clinical view in oral examination

The clinical diagnosis of mesenchymal tumor was confirmed. Excisional sampling was performed and histopathologic diagnosis of HF was approved. For further evaluation, the patient was referred to an oral and maxillofacial medicine specialist and was examined about 1 week after the sampling.

A slight firmness in the examination was felt which was diagnosed as to be associated with the scar of the biopsy. A histopathologic review was requested for the second time. Microscopic evaluation of hematoxylin and eosin (H&E) stained slides showed stratified squamous mucosa with a flat-topped subepithelial nodular proliferation of bland-looking spindle or polygonal cells with oval and occasional elongated reniform vesicular nuclei and inconspicuous nucleoli with clear, foamy, or pale eosinophilic cytoplasm filled subjacent connective tissue (Figure 2).

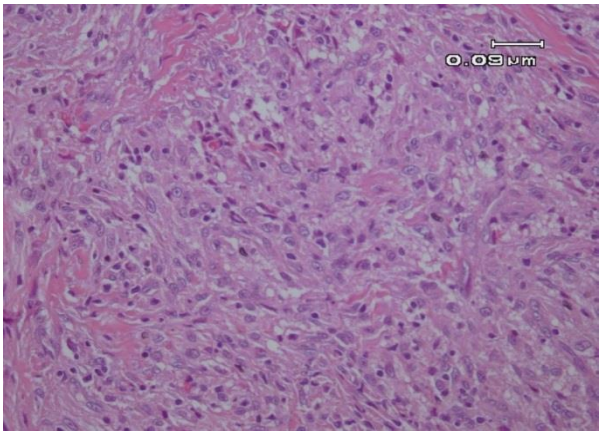

Figure 2. Haphazardly arranged, bland looking spindle cells with no evidence of mitotic figures or cellular atypia, H&E ×100

No abnormal mitosis or necrosis was seen among the neoplastic cells. The immunohistochemical study of tumor cells showed diffuse positivity for CD68. In addition, the tumor cells were negative for both CD1a (Figure 3) and S100 markers (Figure 4). However, S100 marker can be used in determining neural tumors, ki67 (Figure 5) commonly shows proliferative activity, CD1a demonstrates langerhans cells (LC), and CD68 (Figure 6) is positive in cells with fibrohistiocytic origin.

Finally, BFH was rendered as the definite diagnosis. In the four follow-ups up to one year, there was no recurrence. Therefore, the patient was asked to come back in a year.

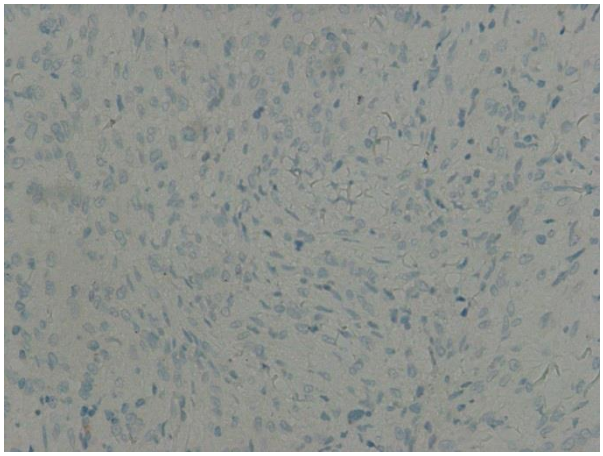

Figure 3. Negative immunohistochemistry (IHC) for CD1a, × 400

Discussion

The cause of BFH has not been detected yet. It can be a proliferative reaction to the source of inflammatory response or a neoplastic process.

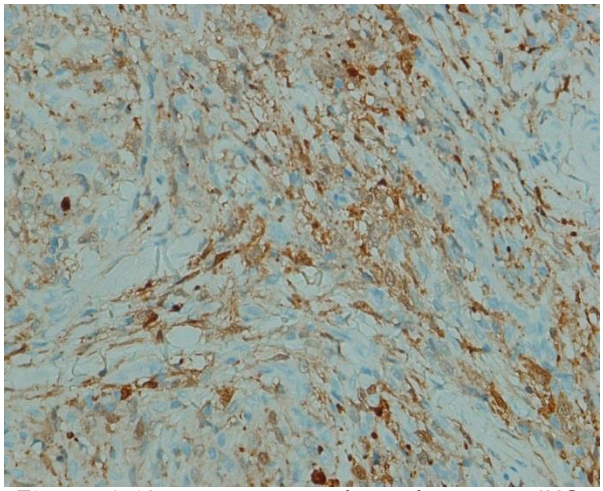

Figure 4. Negative immunohistochemistry (IHC) for S100, × 400

Several cases were created after local injury such as trauma or insect bite or folliculitis that support the inflammatory response theory and other cases support the idea of clonal expulsion associated with the neoplastic process.[7] Clinical diagnosis of oral BFH has features such as slow well-circumscribed enlargement and non-invasive behaviors with intact mucous membranes, and it should be noted that at the clinical level, the disease is not differentiated with other soft tissue neoplasms.[2,7]

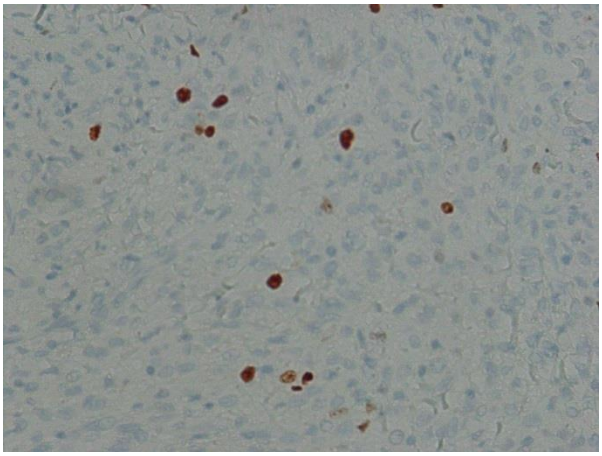

Figure 5. Positive immunohistochemistry (IHC) for Ki67, × 400

There are a few reports on oral BFH mostly indicating a painless nodule on the surface of different parts of the oral cavity like the anterior dorsum of the tongue in subjects under 50 years old, especially in the male patients. It usually takes no longer than six months from presence to diagnosis with discomfort or pain as the most reported complaint.[16-19]

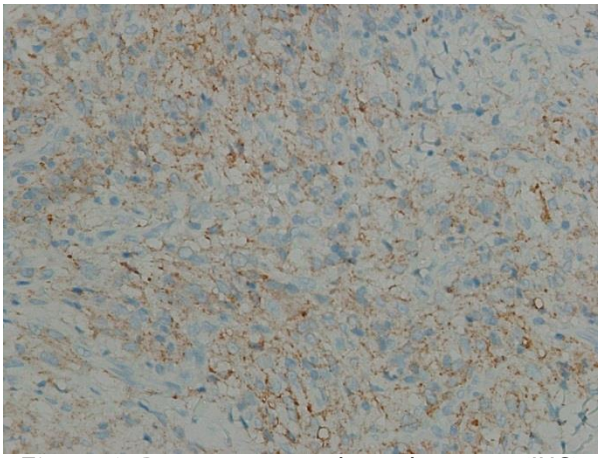

Figure 6. Positive immunohistochemistry (IHC) for CD68, × 400

The present case is the same as other reports in terms of gender and age without any special discomfort due to its slow growth and size.[16,17]

In a review study,[18] the histopathologic view usually showed a non-infiltrating fibrohistiocytic lesion composed of spindle cells. They had vesicular nucleus arranged in

a typical storiform pattern. Oral BFH contained proliferated histiocytes, spindle shaped tumor cells or round histiocyte-like cells, multinucleated giant cells, lipid-containing xanthoma cells, and scattered lymphocytes.[17,20]

Tumors with microscopic spindle-shaped appearance are challenging diagnosis including a spectrum of neoplasms originated from neural tissue (neurofibroma), fibroblastic tissue (fibromatosis), and fibrohistiocytic tissue. In spite of several microscopic findings including wavy-appearance and haphazardly-arranged cellular pattern, IHC staining is necessary to confirm final diagnosis. S100 marker can be used in determining neural tumors, ki67 commonly shows proliferative activity, CD1a demonstrates LC, and CD68 is positive in cells with fibrohistiocytic origin.[16,17]

Today, there is no specific marker for BFH,[9] but with immunohistochemical tests that include low mitosis, atypical cells, vimentin positive, CD68(+), S100(-), CD117(-), LEU7 (-), desmin (-), and SMA (-) can be detected from neurofibroma, leiomyosarcoma, and dermatofibroma.[16,17]

Diagnoses of BFH from malignant fibrous histiocytoma (MFH) is conducted with high cellular polymorphism, high mitotic activation, capsule penetration into the tissue, and revealing hemorrhage and necrosis.[17,21]

Prisse et al. reported BFH to develop in late adulthood and cutaneous form in younger adults since 1975.[22] Recently, most of the lesions in tongue occurred between 20-40-year-old patients and the average age for BFH is 28.5[20,22,23] taken place since 1975. BFH occurs more often in middle age or older years of life, while the cutaneous type occurs in young adults.[7] Moreover, in some studies, considering problems associated with the tongue and by constraining time (from 2000 to date), the dominant age was 20-40 years old,[20,22,23] and the mean of BFH age in the oral mucosa was reported as 28.8 years old.[20,22]

BFH occurs in the oral mucosa more often in women,[22] but in some studies in different populations, the disease is reported to vary in both sexes.[2]

In the study by Kumar conducted on 66 cases of BFH in the period of 1964 to 2016, there were only 7 cases with tongue lesions with 4 male patients. This study showed that 40% and 20% of the lesions occurred respectively in the dorsum and in the anterior part of the tongue.[20]

In a review study, all the BFH patients were treated with local surgical resection or CO_2 laser. Application of the CO_2 laser showed a recurrence which was treated with hemiglossectomy and those with surgical treatment were without recurrence.[23] In another review,[20] the surgical en-bloc treatment was used to treat BFH with a three-year regular following-up period. Since excellent prognosis secondary to a good resection was reported by many authors,[20,22] in the present case, after precise resection and 1,3 ,6, and 12 month follow-ups, no problem emerged.

Conclusion

In this study, a BFH case was reported and examined from clinical, microscopic, and immunohistochemical aspects. Given that BFH is a rare tumor in the mouth, it is essential to perform IHC tests to rule out other lesions. Considering the results of these cases and comparing them with other cases, although there is a slim chance of recurrence in one year, follow up is recommended.

Conflict of Interests

Authors have no conflict of interest.

Acknowledgments

None.

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