Case Report

# Benign fibrous histiocytoma of the lower lip

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## Abstract

The fibrous histiocytoma is a soft-tissue neoplasm of the biphasic cell population of fibroblasts and histiocytes that affects the dermis and the subcutaneous tissue. The objective of this article is to report a case of benign fibrous histiocytoma (BFH) of the lower lip in a 32-year-old female patient with a chief complaint of swelling in the lower lip for the past 1 month. With diagnostic clinical hypothesis of fibrous hyperplasia, fibrous histiocytoma and mucocele, an excisional biopsy was performed. The histopathological examination revealed a nonencapsulated proliferation of spindle cells with some giant multinucleated cells in the periphery of the lesion. Multinucleated giant cells and lymphocytes were noted throughout the lesion. Immunohistochemical reactions were performed, staining only CD68 in the multinucleated giant cells. According to these characteristics, the final diagnosis was BFH.

**Keywords:** Benign fibrous histiocytoma, histiocytes, immunohistochemistry, lip, multinucleated giant cells

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## INTRODUCTION

It is a rarely heard lesion among the pathologies of the oral cavity. According to the literature, it can be rarely malignant and may involve both soft and hard bony tissues.[1] Nowadays, benign fibrous histiocytoma (BFH) is included in the so-called "fibrohistiocytic tumors of the soft tissues" that are divided into cutaneous and noncutaneous types, and in the "fibrohistiocytic tumors of the bone".[2] The World Health Organization in 1960 recognizes the term "fibrohistiocytic" to merely denote a lesion which is composed of cells that resemble round histiocytic and spindled fibroblastic morphology.[3]

Patients having a history of sun exposure, trauma, or chronic infection, suggesting that BFH is a reactive lesion and with predilection in males as compared to females (2.5:1).[2] It has been reported in the age group of older than 25 years with a mean age of 40 years. The clinical features of the oral BFH are of painless solitary tumor, slowly enlarging, from 2 to 3 cm up to more than 10 cm in size and over a period of several months. As far as the oral cavity is concerned, majority of the BFHs are found in the soft tissues of the buccal mucosa, gingiva, lower and upper lip, soft palate and floor of the mouth. The other sites reported to be commonly affected by this tumor are the upper and lower limbs, orbit, retroperitoneum, pelvis, knee, head and neck. Symptoms include dysphagia and dyspnea and when the mass is located in the tongue, difficulty to speak may be present.[1]

## CASE REPORT

A 32-year-old female patient reported to the Department of Oral Medicine and Radiology, Seema Dental College

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and Hospital for a nodular growth on the lower lip that had been present for 1 month [Figure 1]. There was no relevant medical history of any systemic disease or medication. No history of trauma to the jaw, surgical intervention, or any recent dental procedures was given. Intraoral examination revealed a slightly darker color near the vermilion border in the midline of the lower lip, and clinical examination shows well-circumscribed, moderately mobile, nontender fibroelastic lump of approximately 2 cm in diameter.

Then, the patient was referred to the Department of Oral and Maxillofacial Surgery where she underwent an excision of the mass under general anesthesia with primary closure. After surgery, the biopsy was referred to the Department of Oral and Maxillofacial Pathology for the histopathological diagnosis.

Macroscopic examination shows smooth, well-circumscribed encapsulated lesion of 8 mm × 6 mm × 5 mm in size. The specimen appeared oval in shape, creamish-brown in color [Figure 2]. The H&E-stained sections revealed a non-infiltrating fibrohistiocytic lesion with grenz zone in relation to an overlying stratified squamous epithelium [Figure 3]. The lesion was chiefly composed of interlacing fascicles of spindle cells having a plump and vesicular nucleus with tapered and blunt ends arranged in a storiform pattern. Focal areas of few pleomorphic cells, round histiocytes, lipid-laden macrophages, lymphocytes and multinucleated giant cells were noted. The special stain of Masson trichrome was also done revealing the storiform pattern of collagen fibers [Figure 4]. For the final confirmation of diagnosis, tissue blocks were sent for immunohistochemical (IHC) studies [Figure 5]. The tumor cells showed a high intensity for CD 68. Based on the H&E and IHC findings, a final diagnosis of BFH was made.

## DISCUSSION

Fibrous histiocytoma represents a benign but diverse group of neoplasms which exhibit both fibroblastic

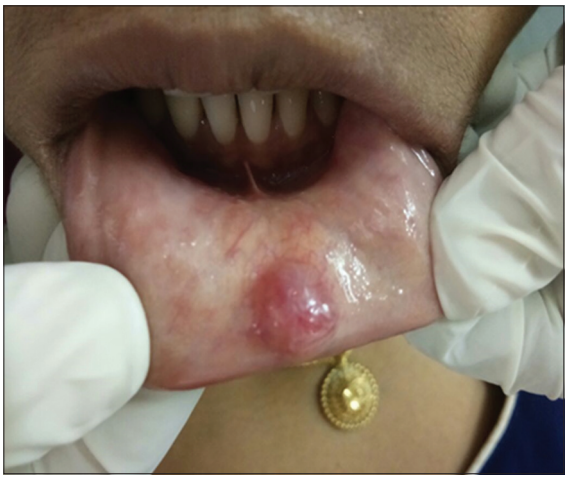

**Figure 1:** Clinical photomicrograph of patient showing swelling w.r.t left lower lip

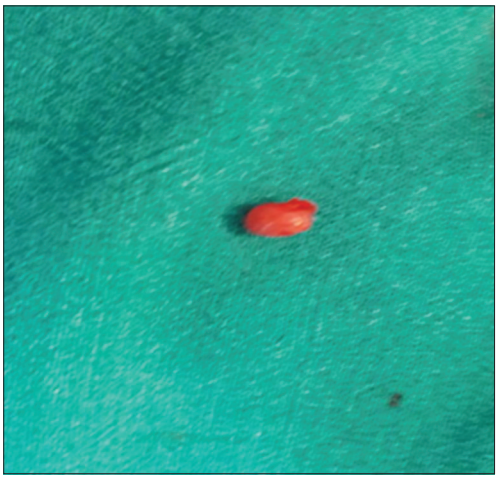

**Figure 2:** Photomicrograph of excised specimen

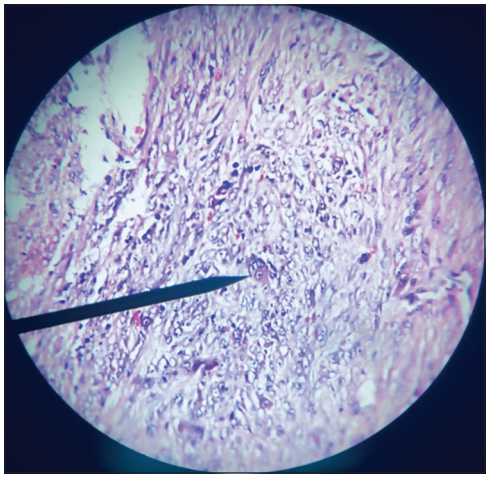

**Figure 3:** Photomicrograph showing H&E staining with giant cell (×10)

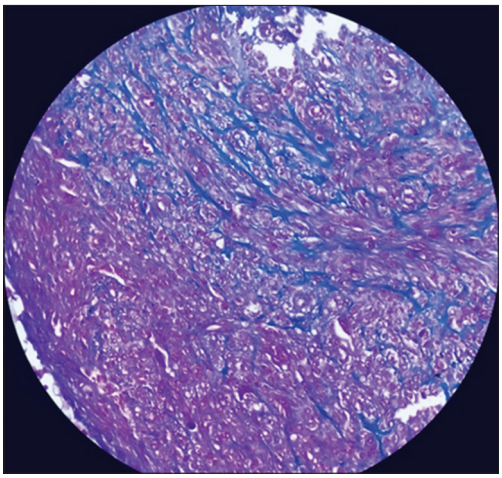

**Figure 4:** Photomicrographh showing Masson Trichrome staining showing intense amount of Collagen fibers (×10)

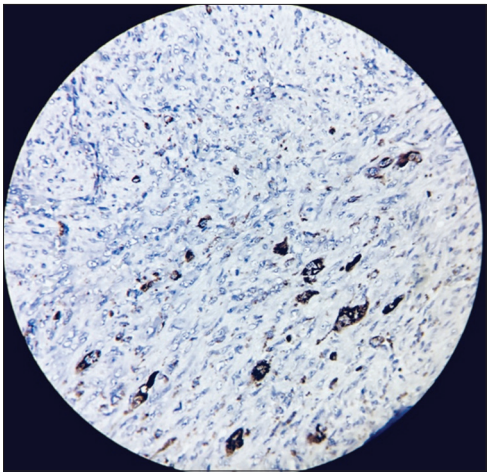

**Figure 5:** Photomicrograph showing IHC staining of CD68 showing (×10)

and histiocytic differentiation.[4] It was first described by Stout and Lattes in 1967. Before 1960, the BFH was not considered a distinct clinicopathological entity. However, with electronic microscopy and IHC advancement, it was possible to differentiate this diagnosis.[5] The histogenesis of this tumor is controversial. On account of the tissue culture evidence, it was initially proposed that it has a histiocytic origin, in which the histiocytes act as facultative fibroblasts.[6] Later on, results of electron microscopic and ultrastructural studies suggested that the histiocytic and fibrocytic cell lines are derived from the small numbers of undifferentiated mesenchymal cells. Apart from this, cell marker studies have supported a fibroblastic origin for these tumors.[7]

Gray *et al*. found that the mean age of patients was 55 years ranging from 12 to 71 years.[8] Bielamowicz *et al*. in their study of BFH in the head-and-neck region found an M:F ratio of 2.5:1.[9] The clinical appearance is related to the location of the tumor [Table 1]. Oral BFH presents as a solitary, painless and well-circumscribed nodule.[7]

Histiocytes/macrophages are derived from monocytes and play an important role in the regulation of immune functions. They are involved in different aspects of host defense and tissue repairs, such as phagocytosis, cytotoxic activities, regulation of inflammatory and immune responses and wound healing.[10] Disorders of monocytes and histiocytes/macrophages are divided into three major categories: (1) functional defects, (2) reactive responses and (3) neoplastic proliferations. Functional defects are mostly hereditary, such as lysosomal storage diseases. Reactive responses are nonneoplastic conditions associated with hypoplasia, hyperplasia or hyperactivation of the monocytic/histiocytic system. Neoplastic disorders are the result of the clonal proliferation of monocytic/histiocytic cells.[11]

The term histiocyte was originally used to designate a large cell normally found in lymph nodes and spleen that was morphologically nonspecific but had voluminous, granulated cytoplasm, sometimes containing ingested particles and one or more round to irregularly shaped pale nuclei. Subsequently, the term histiocyte was taken as synonymous with the fully differentiated end cells of the monocyte/macrophage lineage, including sinusoidal macrophages in the spleen, alveolar macrophages in the lung and Kupffer cells in the liver.[12]

H&E-stained section reveals stratified squamous epithelium along with connective tissue stroma. Connective tissue stroma shows a dual cell population of fibroblasts and histiocytes. The fibroblasts are spindle shaped and arranged in a storiform pattern (loosely arranged whorls of elongated spindle fibroblast cells).[13] The differential histological diagnosis includes neurofibroma, leiomyosarcoma and dermatofibroma, so-called atypical-BFH.[2]

The cells do not show any sign of malignancy as no mitotic activity is seen. Many of the nuclei are large, and a multinucleated Touton-type giant cell is seen. The Touton-type giant cell is a giant cell with a ring of nuclei with foamy cytoplasm around. The stroma is densely fibrous and shows areas of myxoid change or focal hyalinization.[14] Special stain such as Masson trichrome stain was also performed. The stained section shows the storiform pattern of collagen fibers of light blue color and dark blue-stained fibroblast and histiocytes.

IHC acts as a useful aid in diagnosis by confirming the origin of cells. CD68 is a transmembrane glycoprotein that is particularly useful as a marker for the various cells of the macrophage lineage, including monocytes, histiocytes and giant cells. In the current case, the positivity for CD68 demonstrates the histiocytic nature of the cells.[10]

## CONCLUSION

Benign fibrous histiocytoma is a mesenchymal tumor composed of cells with fibroblastic and histiocytic characteristics. The tumor is rare and presents a clinical and histopathological challenge. After 1 year of follow-up, the patient did not have any specific problems. Proper diagnosis and treatment plan with long-term follow-up is of utmost importance in the management of these tumors.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have

given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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