

CASE REPORT



An unusual case of malignant fibrous histiocytoma presenting as multiple gingival masses: A rarity

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Abstract

Fibrous histiocytomas are soft-tissue tumors of both benign and malignant counterparts presenting in adults. Benign fibrous histiocytoma occurs mainly in the skin of the extremities, might involve the long bone. Its incidence in head and neck is extremely uncommon and comprises about only 1% fibrous histiocytomas whereas about 3–10% of malignant fibrous histiocytoma (MFH) occur in the head-and-neck region. Here, we are presenting a rare case of multiple gingival masses of MFH which adds to the literature about clinical and biological behavior of the rarity, emphasizing on the clinical correlation.

Keywords: Gingiva, Malignant fibrous histiocytoma, multiple sites, oral soft-tissue sarcomas, recurrence.

Introduction

Fibrous histiocytomas are benign soft-tissue tumors arising as fibrous mass. Malignant fibrous histiocytoma (MFH) is a malignant neoplasm of uncertain origin that arises from soft tissue and bone, histopathologically rich in histiocytes with a storiform pattern presenting as rarity in head and neck. Benign fibrous histiocytomas (BFHs) with gingival involvement documented only in seven cases.^[1] MFH in oral cavity is only 3% of all reported cases.^[2,3] BFHs of head and neck occur in adults over 25 years, mean age of 40; present as painless, slow-growing mass, usually more circumscribed than cutaneous form. MFH occurs in older adults – 5th–7th decade with male predilection,^[4] presents as rapidly growing fungating mass, and mostly occurs as a secondary metastatic tumor, sometimes as primary origin. BFHs are treated by wide surgical excision with minimal local recurrence of 5–10%.^[5] MFH needs invasive management of excision along with radiotherapy since more aggressive course as reported in a study where 5-year overall survival was 48% for patients with head-and-neck tumors, compared with 77% for patients with trunk/extremity tumors.^[6]

MFH first described by O'Brian and Stout in the 1960s and histopathological features were described first by Kempson and Kyriakos.^[7] Recently, the World Health Organization soft-

tissue tumors classification has modified the term “MFH” to include undifferentiated pleomorphic sarcoma, which is now the preferred term in the literature and fundamentally represents a diagnosis of exclusion. Clinical variants of MFH can be primary and secondary – arising as metastases from other part of body. Primary MFHs are extremely rare in oral cavity; curious pathologies to diagnose, monitor, and treat. Here, we document this peculiar case emphasizing clinical correlation in rarities. To the best of our knowledge, no case of MFH of gingiva occurring in multiple sites has been reported in literature.

Case Report

A female patient of 65 years of age came to our institution with a chief complaint of swelling and pain in the gums of upper front teeth for the past 20 days which was initially small and increased to attain present size. The associated pain occurred only during chewing. The patient was hypertensive and was under medication for the past 14 years. Intraorally, two swellings: A well-defined sessile growth of gingiva buccopalatally measuring 2 × 3 cm present in relation to teeth 11 and 21 [Figure 1a] and in gingiva of 33 and 34 measuring 0.5 × 0.5 cm with pale, smooth surface, firm consistency, and mild tenderness on palpation, without secondary changes. Hard tissue examination revealed Grade II



Figure 1: (a) Lesion in the gingiva of maxillary anterior. (b and c) The bone loss in intraoral periapical radiographs in the lesion site

mobility of teeth 11, 21, and 34. Mild bleeding on probing with periodontal pocket depth 7 mm was present. Intraoral periapical radiographs revealed horizontal alveolar bone loss in teeth 11, 21, 33, and 34 extending till middle and apical 1/3 of the roots with widening of periodontal ligament space [Figure 1b and c]. A provisional diagnosis of soft-tissue growth benign variants – irritation fibroma and fibrosed pyogenic granuloma was suggested.

After routine blood investigations, an excisional biopsy of gingival lesions and extraction of teeth 11 and 21 was done under local anesthesia, closed with 3-0 black silk suture. The specimens were sent for histopathological study. During the follow-up for suture removal, the excised region showed healing in progress. Both the specimens were studied and showed that soft-tissue section being highly cellular in nature, consisting of two cell types – spindle cells and round cells. The spindle cells were seen as short fascicles with some areas giving matted appearance. The round cells were presented with lobulated appearance in certain areas. Hyperchromatism and mitotic figures were present in spindle cells on the surface. In this section, the border of the lesion was clearly visible with no evidence of infiltrating picture [Figure 1].

Because of the multiple nature of similar lesion, deeper sections were taken from both the specimens and it exhibited aggressive nature of the proliferating cells, hyperchromatism, open faced nuclei, and atypical mitotic figures and diagnosed as “MFH” [Figure 3]. Hence, the patient was recalled and but when the patient reported 2 weeks later, the patient exhibited appalling recurrence of the lesion with size approximately 7 × 8 cm in the maxillary gingiva and 5 × 3 cm in the gingiva of the left lower premolar region, well defined, with surface showing mild erythema and ulcerations, hindering in function and nutrition [Figure 4a and b]. Following that, planning was done to excise the recurred lesion with wider margins. However, the patient did not report back and later we came to know that the patient died.

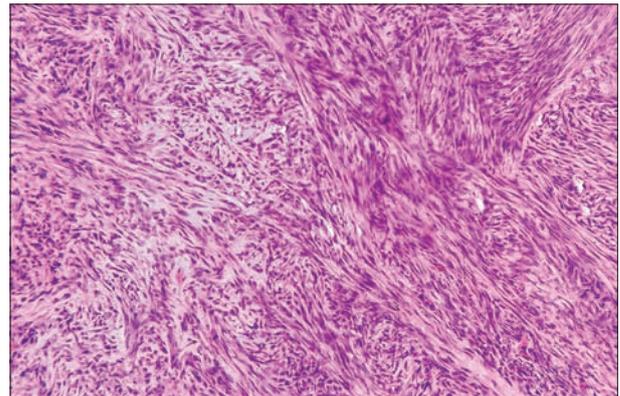


Figure 2: Histopathological picture presenting fibrocytes and histiocytes arranged in storiform pattern. Hematoxylin and eosin 40×

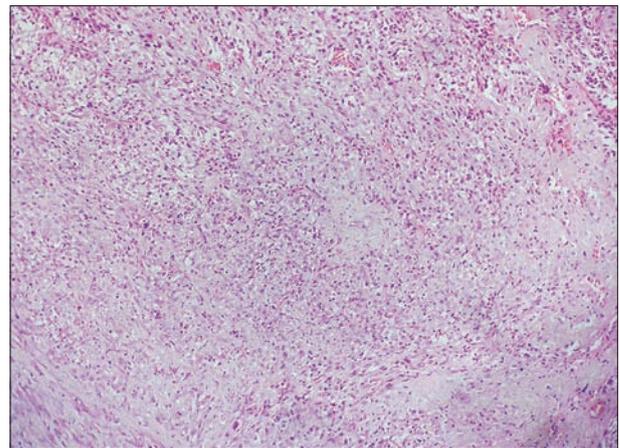


Figure 3: Serial section shows atypical mitotic figures, hyperchromatism, and cells with increased activity. Hematoxylin and eosin 40×. Malignant fibrous histiocytoma



Figure 4: (a and b) The alarming recurrence of the lesion

Discussion

MFHs are rare soft-tissue sarcoma affecting the oral cavity, common in buccal mucosa and vestibular region. Other sites reported are involving skull bones, maxilla and mandible, gingiva, and tongue. Gingiva being rarest of the sites; to our knowledge, only few cases have been reported in literature^[8] – Vijayalakshmi *et al.*, 2012, Park *et al.*, 2009, Agnihotri *et al.*, 2008, Boaz *et al.*,

2017, and Bali *et al.*, 2010. The diagnosis of fibrous histiocytomas solely depends on histopathology. Types of histopathological patterns are – storiform pleomorphic (70%), myxoid (20%), giant cell, and inflammatory. In the present case, multiple gingival masses with recurrence within a month are noted. The initial lesion was clinically showing hints of fibromatous lesion – peripheral giant cell granuloma and peripheral ossifying fibroma. This case is unique in ways – as it occurred in two sites, a variation from usual; the recurrence transpired rapidly with increase in size and hindering in nutrition in second follow-up (within weeks), even though healing was in progression the first follow-up; histologically – the presence of atypical mitotic figures, hyperchromatism, open faced nuclei reported as MFH, which was substantiated clinically when the patient presented with alarming recurrence. MFH has a poor prognosis and usually shows difficulty in diagnosis due to their varying clinical features.

Conclusion

This case is a rarity, probably the first case of MFH of gingiva in multiple sites to be documented. Since the diagnosis of malignant fibrous histiocytoma solely depends on histopathology, and does not show particular distinct clinical presentation, can even mimic their benign non-invasive counterpart clinically, it is crucial to follow up periodically and clinically correlate and intervene whenever indispensable. We hope that documenting this case in literature will add to reference of peculiar lesions of oral fibrous histiocytomas so it leads toward meticulous knowledge to the oral physician and surgeon community in diagnosis and management.

References

1. Prisse LA, Jayasooriya PR, Mendis BR, Lombardi T. Benign fibrous histiocytomas of the oral mucosa: Report on three cases and review of the literature. *Dermatopathology (Basel)* 2015;2:52-60.
2. Hardison SA, Davis PL III, Browne JD. Malignant fibrous histiocytoma of the head and neck: A case series. *Am J Otolaryngol* 2013;34:10-5.
3. Sreenivasan BS, Sankar VV, Mathew DG, George M. Undifferentiated pleomorphic sarcoma presenting as a gingival mass: A rare case. *J NTR Univ Health Sci* 2015;4:130-3.
4. Sarodea SC, Sarodea GS, Ingaleb Y, Ingalec M, Rajd AT, Patile S. Malignant fibrous histiocytoma of the mandible-a case report and review of published case reports. *J Oral Biol Craniofac Res* 2019;9:221-5.
5. Rajendran R. Benign and malignant tumors of oral cavity. In: Rajendran R, Sivapathasundaram B, editors. *Shafer's Textbook of Oral Pathology*. 7th ed. Noida, New Delhi: Elsevier; 2012. p. 163.
6. Sabesan T, Xuexi W, Yongfa Q, Pingzhang T, Ilankovan V. Malignant fibrous histiocytoma: Outcome of tumours in the head and neck compared with those in the trunk and extremities. *Br J Oral Maxillofac Surg* 2006;44:209-12.
7. Lambade PN, Lambade D, Saha TK, Bande CR, Ramakrishana A. Malignant fibrous histiocytoma: An uncommon sarcoma with pathological fracture of mandible. *J Maxillofac Oral Surg* 2015;14:283-7.
8. Zhurakivska K, Troiano G, Montella M, Lo Muzio L, Fiorillo L, Cervino G, *et al.* Oral health and molecular aspects of malignant fibrous histiocytoma patients: A systematic review of the literature. *Int J Environ Res Public Health* 2020;17:1426.