Case Report

Malignant fibrous histiocytoma of the mandible: A rare case report with diagnostic challenge and treatment approach

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Abstract

Malignant fibrous histiocytoma (MFH), now termed undifferentiated pleomorphic sarcoma (UPS), is a high-grade tumour, mostly affecting the extremities and retroperitoneum. It is a rare entity in the mandible. After a literature search in the PubMed database, only 15 cases have been reported, indicating how uncommon it is in the mandible. In this instance, we document an unusual occurrence of MFH, affecting the mandible in a 37-year-old male patient. Histopathology of the biopsy specimen along with the strong positivity to immunohistochemical markers such as Vimentin and CD68 confirmed the diagnosis of MFH. It is a rare entity presenting diagnostic and therapeutic challenges requiring a multidisciplinary approach for optimal management and treatment.

Keywords: CD68, high-grade tumour, malignant fibrous histiocytoma, mandible, storiform, vimentin

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INTRODUCTION

Malignant fibrous histiocytoma (MFH) is a rare cancer that can develop in various soft tissues throughout the body. Its occurrence in the mandible is less common compared to other locations.[1,2] While it can manifest at any age, it primarily affects adults, with a slight male predominance.[3] About 70% of cases present as primary tumours, while 30% arise from pre-existing conditions, notably in individuals who have undergone radiation therapy. Diagnosis typically involves a combination of clinical examination and imaging techniques like panoramic radiography, computed tomography (CT), or magnetic resonance imaging (MRI), along with histopathological analysis of biopsy samples. Histologically, MFH is characterized by pleomorphic spindle cells demonstrating histiocytic and fibroblastic features. Here, we describe a case of MFH involving the mandibular region.

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CASE PRESENTATION

A 37-year-old male patient reported to the Oral Pathology Department with the chief complaint of swelling in the lower front region of the jaw for the past one year. The patient gives a history of the primary lesion in the same region for which complete lesional excision was performed. The patient gives no history of associated pain. Extraoral examination revealed no gross asymmetry [Figure 1a]. Right and left submandibular lymph nodes were palpable, approximately 1.5 × 2 cm, soft to firm in consistency, mobile and roughly oval, non-tender. Intraoral examination

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reveals differentiated growth seen over gingiva both buccally and lingually in relation to lower right canine to first molar extending anteroposteriorly from mesial of lower right canine to distal of first molar, superoinferiorly from occlusal level to depth of vestibule [Figure 1b]. On inspection, the size of the lesion was 4 × 2 cm, roughly oval, pinkish white in colour, margins were diffused, and borders were regular and smooth in the surface. On palpation, tenderness was absent, soft to firm in consistency, and was not fixed. The clinical differential diagnosis was made as pyogenic granuloma and malignancy of lingual alveolar mucosa in 41–46 region. Investigations advised were orthopantomogram (OPG), CT, ultrasound sonography (USG), and biopsy. OPG revealed a radiolucent image with ill-defined, irregular borders extending from lower right 3 to the distal of 6 [Figure 1c]. CT report revealed an ill-defined intensely enhancing mass lesion seen in the right buccal mucosa near the alveolar ridge and adjacent to the tongue [Figure 1d]. The lesion measured 4.5 × 2.2 cm. There was the presence of bilateral submandibular lymphadenopathy largest measuring 1 × 0.6 cm in the right submandibular region. Features suggested carcinoma of the right buccal mucosa with the above-mentioned extensions and lymphadenopathy. USG neck showed no relevant or contributory findings. A new biopsy was performed, and the microscopic evaluation revealed the presence of a storiform arrangement of proliferating spindle-shaped neoplastic cells interspersed with pleomorphic histiocytes with foamy cytoplasm [Figure 2a]. Multiple multinucleated giant cells suggestive of touton giant cells were also appreciated [Figure 2b]. For confirmatory diagnosis, immunohistochemistry markers were applied. The tumour cells showed significant expressions of vimentin and CD68, but cytokeratin (CK), S100, and epithelial membrane antigen (EMA) were negative leading to a final diagnosis

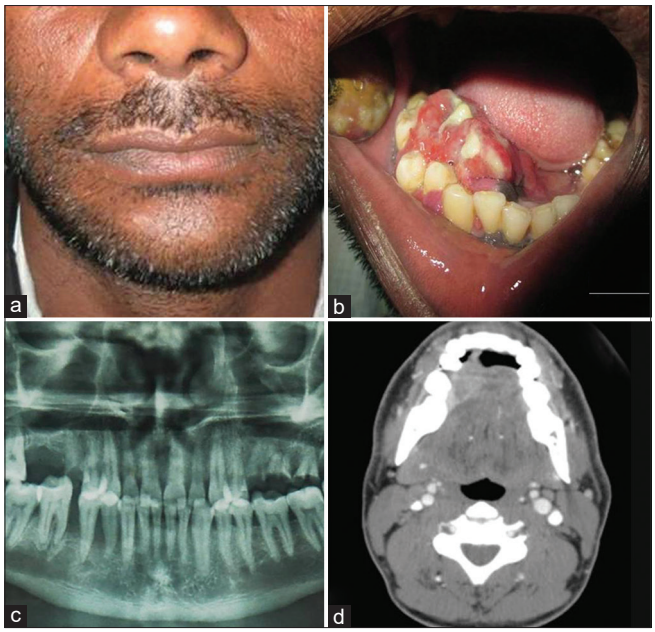

Figure 1: (a) Extraoral photograph, (b) intraoral growth in the lower right quadrant, (c) orthopantamogram (OPG) showing radiolucency in the lower right quadrant. (d) Computed tomography (CT) revealed an ill-defined intensely enhancing mass lesion seen in the right buccal mucosa near the alveolar ridge and adjacent to the tongue

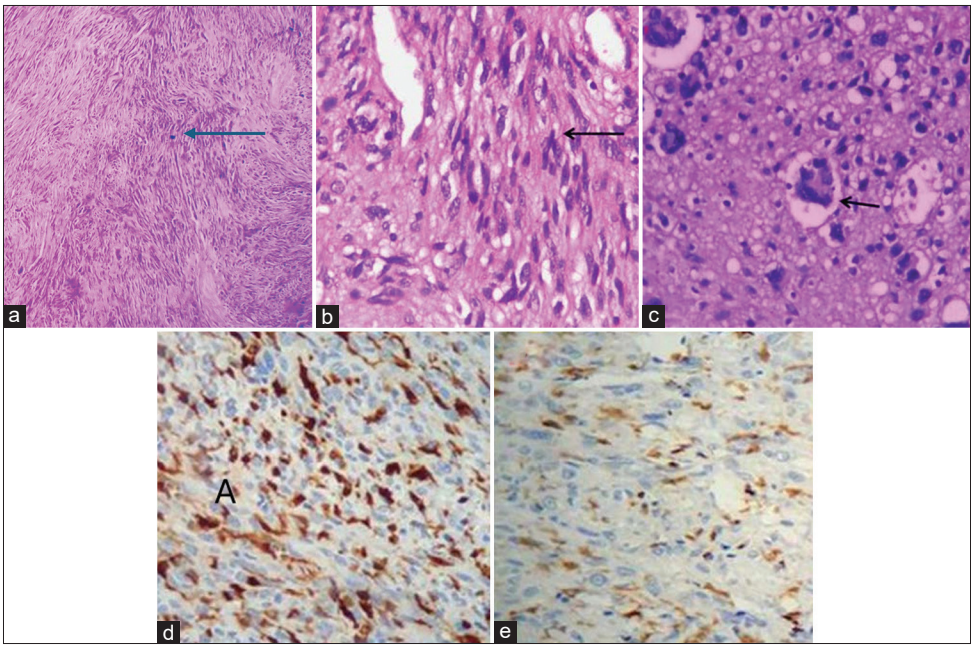

Figure 2: (a and b) Haematoxylin and eosin (H and E) stained section shows pleomorphic spindle-shaped cells arranged in storiform pattern (4X,10X magnification). (c) H and E-stained section shows multinucleated giant cell (10X magnification). (d) The immunohistochemistry (IHC) section shows strong positivity to vimentin. (e) The IHC section shows positivity to CD68

of malignant fibrous histiocytoma [Figure 2c and d]. The patient underwent a right segmental mandibulectomy procedure.

DISCUSSION

MFH remains a frequently encountered lesion with an elusive origin, affecting both soft tissue and bone. Since its introduction by Kauffman and Stout in 1961, controversy has surrounded its classification.[4] In 2002, the World Health Organization (WHO) reclassified MFH as an undifferentiated pleomorphic sarcoma (UPS), removing its formal diagnostic status.[5] Notably, individuals aged between 50 and 70 years exhibit a higher incidence of MFH, with the youngest reported case involving mandibular region involvement at 15 years old.[6,7] A male predominance is observed in 65% of cases.[8] Clinically, MFH typically presents as a progressively enlarging mass, often painful over time. Our patient, a 37-year-old man, presented with an asymptomatic swelling on the right side of the lower jaw persisting for one year. Although trauma accounts for 20% of MFH cases, our patient had no history of such trauma, fuelling debates regarding MFH's association with trauma without substantial evidence.[9,10] While primary lesions predominate, secondary lesions are less common but strongly associated with prior radiation exposure.[11,12] Histologically, MFH exhibits various subtypes, with the giant cell variant displaying the poorest prognosis due to its aggressiveness. Recurrence rates are high, influenced by aggressive tissue infiltration. Storiform-pleomorphic type predominates, consistent with our case, as supported by immunohistochemistry (IHC) findings showing vimentin and CD68 positivity.[13-15] Management typically involves a multidisciplinary approach encompassing surgical resection, adjuvant chemotherapy, and radiation therapy. In this case, the patient underwent segmental mandibulectomy to achieve complete tumour resection with wide margins. Close follow-up examinations are essential for early detection of recurrence or metastasis and long-term surveillance.[11]

CONCLUSION

MFH involving the mandible is relatively rare. It presents diagnostic and therapeutic challenges requiring a multidisciplinary approach for optimal management and treatment.

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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Nil.

Conflicts of interest

There are no conflicts of interest.

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