

Inflammatory myofibroblastic tumor of the upper alveolus: A rare entity presenting as a jaw swelling

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ABSTRACT

Inflammatory myofibroblastic tumor (IMT) is a rare tumor of borderline nature that can clinically present as a malignant neoplasm. It commonly occurs in the lungs, and a very few oral IMTs have been reported in the literature. IMT consists of inflammatory cells and myofibroblastic spindle cells. The diagnosis of IMT requires histopathological examination with immunohistochemical staining to look for the expression of smooth-muscle actin for confirmation of the diagnosis. The objective of this paper is to report an IMT on the upper alveolus with clinic-a pathological similarity with a malignant lesion and its management. Though oral IMTs are rare, it should be considered in the differential diagnosis of tumors of the upper jaw. Complete surgical excision of alveolar IMT is the treatment of choice because of its unpredictable clinical behavior. The patients with oral IMTs require periodic post-surgical follow-up for recurrence.

Keywords: Benign tumor, inflammatory myofibroblastic tumor, jaw swelling, upper alveolus

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a distinct borderline lesion composed of myofibroblastic cells with a variable admixture of inflammatory cells, including mature lymphocytes, histiocytes, plasma cells and eosinophils, and collagen. Inflammatory pseudotumor was first described by Brunn in 1939. Umiker and Iverson have first described it as a tumor.^[1] IMT was earlier known as inflammatory pseudotumor, which was later coined as IMT by Scott *et al.* in 1988.^[2] IMT commonly occurs in the lungs. IMT has a clinical and radiological behavior similar to that of a malignancy.^[3] IMT occurs anywhere in the body including the oral cavity and the major salivary glands.^[4-6] The etiology and pathogenesis of IMT are not well known. Here, we describe a case of IMT on the upper alveolus and its management.

CASE REPORT

A 36-year-old female presented with a progressive and painless swelling on the left upper jaw of 1 month duration. There was no suggestive history of intake of medications known to cause

gum hypertrophy. On intraoral examination, there was a large lobulated, pinkish color swelling on the left upper alveolus abutting the adjacent hard palate [Figure 1]. Mucosa over the swelling was intact. The swelling was firm in consistency and nontender. There were no palpable neck nodes. Computed tomogram scan (CT scan) of the maxillo-facial region showed a heterogeneously enhancing soft tissue mass lesion on the alveolus with lytic destruction of the upper alveolus [Figure 2a]. The maxillary antrum and the orbital floor were free from the mass [Figure 2b].

Incision biopsies were taken from the growth for 2 times under local anesthesia and both the biopsies showed only hyperplastic squamous epithelium with inflamed fibro-collagenous tissue without any evidence of malignancy. As the lesion was growing rapidly, and no definitive pathological diagnosis could be established, the patient was considered for up front surgery. Weber-Fergusson's incision without the infra-orbital extension was made for the exposure for excision of the tumor [Figure 3a]. Intra-operatively mass was limited to the upper alveolar only without extension to adjacent palate and the floor of the maxillary antrum. Hence, the procedure done was a partial

upper alveolectomy. A tumor of 7 × 4.5 × 3 cm in size was removed in toto [Figure 3b]. After excision of the tumor as there was no oro-antral fistula, and the resultant wound was allowed for a second-intention healing. The specimen was sent for the final histopathological examination. Microscopic examination of the section from the specimen showed low grade collagenous and focally myxoid spindle cell neoplasm [Figure 4a]. Immunohistochemical (IHC) staining of the tissue section was done for smooth muscle actin (SMA), S-100 protein, CD 30, cytokeratin and Ki67. IHC was positive for the expression of SMA [Figure 4b]. IHC expression was negative for S-100 protein, CD 30, Ki67 and cytokeratin. The diagnosis of IMT was confirmed after combination of the clinical features, radiological images, histopathological examination and multiple IHC staining.

The patient is currently on follow-up, and there is no recurrence or associated morbidity like trismus at the 1½ years postsurgery follow-up [Figure 5].

DISCUSSION

Inflammatory myofibroblastic tumors are classified as tumors of intermediate biological behavior because of its propensity for local recurrences and rarely nonhead and head visceral IMTs have been shown to metastasize.^[7] Extra pulmonary IMT is rare and few cases have been reported in the literature. Synonyms of IMT were inflammatory pseudo tumor, plasma cell granuloma, plasma cell pseudo tumor, pseudosarcomatous lesions/tumors.

Different hypothesis for origin of IMTs has been suggested like infection, auto immune, and trauma. The pathogenesis and etiology of IMT are not clear. Swain *et al.*, have shown that IMTs may have a different etiology and clinicopathologic features from inflammatory pseudo tumor in the central nervous system.^[8] The presence of human herpesvirus-8 DNA sequences and the over expression of interleukin 6 and cyclin D1 have been reported in IMTs.^[9]

The common head and neck site of IMT is the larynx and the intra oral site for IMT is the buccal mucosa and it also occurs in other head and neck sites such as the tonsils, para-pharyngeal space, sino-nasal tract, and the trachea.^[7] However, in our case IMT occurred in the upper alveolus abutting the adjacent hard palate. IMT of the upper jaw has been reported in an 11-year-old female by Rautava *et al.*^[10] IMTs of upper aero-digestive tract are common in adults (median age of 59) and more often seen in the males.^[11] In the present case the patient is a female of 36 years old. Binmadi *et al.* have shown that in the upper alveolus it presented as a small sessile nodule.^[12] However, in our case it presented as a large multi lobulated swelling on the upper alveolus. IMTs are characterized by ill-defined infiltrative bony erosion on CT scan,^[2] and in the present case there was a heterogeneously enhancing mass with destruction of the upper alveolus on the CT scan.

The histological differential diagnosis of IMT includes benign tumors like nodular fasciitis, solitary fibrous tumor, benign



Figure 1: Intraoral picture of the patient showing the large multi lobulated swelling on the upper jaw and adjacent palate

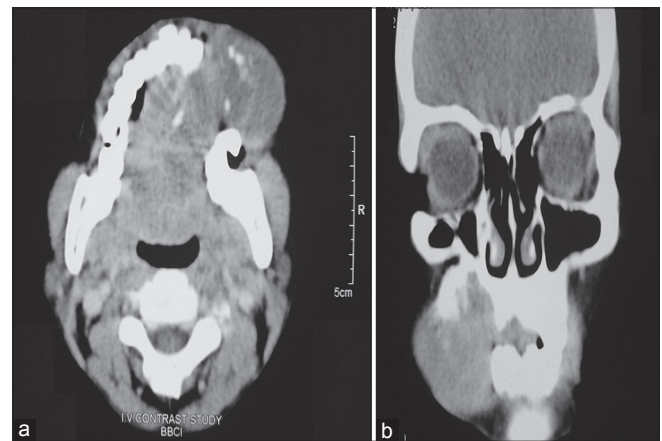


Figure 2: (a) Computed tomogram scan (CT scan) image showing the destruction of bone of the upper jaw, (b) coronal CT scan image showing the intact orbital floor

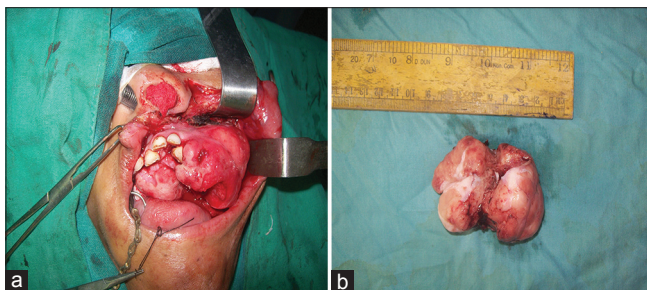


Figure 3: (a) Intraoperative picture showing the exposure for removal of the tumor, (b) the postoperative specimen of the tumor removed in toto

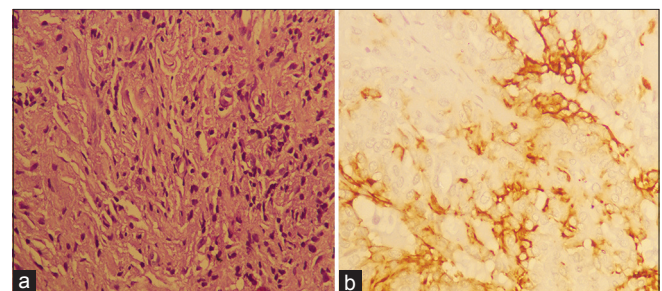


Figure 4: (a) Photomicrograph of H and E, ×40 showing features of inflammatory myofibroblastic tumor, (b) picture showing the immunohistochemical positivity for the expression of smooth muscle actin

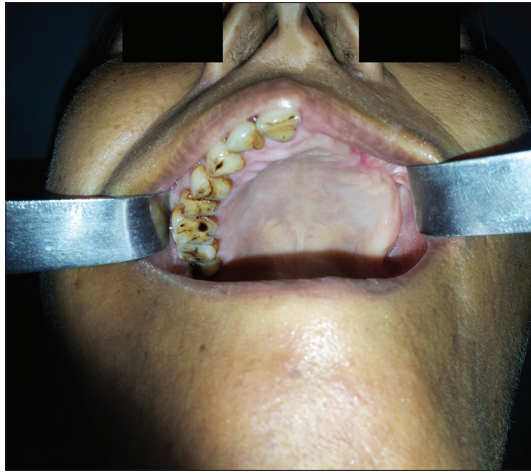


Figure 5: Intraoral picture of the patient at the 1 years post excision follow-up

fibrous histiocytoma, follicular dendritic cell tumor, myofibroma and malignant tumors like, malignant peripheral nerve sheath tumor (MPNST), anaplastic large cell lymphoma (ALCL), fibrosarcoma and leiomyosarcoma. In nodular fasciitis, there is less inflammatory infiltrate than IMT and mucin rich stroma.^[13] IHC for the present case was positive for the expression of SMA, which is a diagnostic feature on IHC for IMT. In the diagnosis of IMT epi-myoeithelial islands should not be detected, even by IHC staining for cytokeratin like in the present case.^[14] Histologically the present case was a low-grade lesion, so the diagnosis of MPNST, inflammatory fibroid polyp, ALCL, and malignant fibrous histiocytoma were not considered. However, low grade neural lesion was ruled out by negativity of S-100 marker. Gallego *et al.* has also demonstrated that in IMT there is faint positivity of Ki67 on IHC study.^[15] However, in the present case IHC for Ki67 was negative for nuclear stain.

Non, surgical management of IMT, includes chemotherapy in the form of cyclosporine, methotrexate, azathioprine, and cyclophosphamide but it has little role and corticosteroids is not so effective in the treatment of IMTs of the head and neck region.^[4] Moreover, in the present case conservative treatment with corticosteroids could not be started due to the lack of definitive diagnosis of the mass preoperatively. In the present case the lesion was clinically and radiologically mimicking a malignant tumor and such an aggressive behavior of upper jaw IMT has also been shown by Gale *et al.*^[16] Follow-up for 10 years to detect local recurrence following simple excision of oral IMTs has been suggested.^[15] In the present case, only surgical excision was done, and there was no local recurrence at the follow-up of 1½ years after treatment.

CONCLUSION

Authors would like to add an unusual presentation of IMT of the upper alveolus to the literature. Though alveolar IMTs are extremely rare, it can be considered in the differential diagnosis

of tumors of the upper jaw. Complete surgical excision of alveolar IMT is the treatment of choice because of its unpredictable clinical behavior. The patients with oral IMTs require periodic postsurgical follow-up to detect local recurrence.

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