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CASE REPORT

Inflammatory Myofibroblastic Tumor: A Rare Tumor in the Tongue

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ABSTRACT

Inflammatory myofibroblastic tumor (IMT) consists of myofibroblastic and inflammatory cell infiltration in the tissues. Recurrence and malignant transformation rate of this tumor is very unusual and regarded as benign fibroinflammatory disease. The etiological factor of this tumor is unknown but infection, trauma, and immunologic factors are blamed.

In this case report, we report a 30-year-old woman with a proliferative mass on her right side of tongue, which was diagnosed as "IMT." Inflammatory myofibroblastic tumor is very rarely found on tongue. This type of tumor may be misdiagnosed as malignant tumor because of its diagnostic difficulties. Wide local excision of tumor was done for an adequate treatment.

Conclusion: Clinical as well as radiological behavior of IMT is aggressive, so it may be misdiagnosed as malignant tumor. Therefore, its accurate diagnosis is vital to prevent unnecessary radical resection.

Keywords: Malignant tumor of tongue, Myofibroblastic tumor, Pseudotumor of tongue.

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INTRODUCTION

Inflammatory myofibroblastic tumor consists of myofibroblastic and inflammatory cell infiltration in the tissues.

Recurrence and malignant transformation rate of this tumor are very unusual and regarded as benign fibroinflammatory disease. The etiological factor of this tumor is unknown but infection, trauma, and immunologic factors are being accused.[1] Inflammatory myofibroblastic tumor is rarely found in head and neck region, although it is found anywhere in the body and is common in the abdominal cavity and extrimities.[2] We report a case of IMT of the tongue which is extremely rare.

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CASE REPORT

A 30-year-old woman presented to the ear, nose and throat outpatient department with painless mass on the right half of tongue for a period of 12 months which was associated with difficulty in speaking and chewing food. Oral examination revealed an approximately 6 cm × 3 cm sized, firm, and nontender mass at the right side of anterior two-thirds of tongue extending medially to midline with well-defined margin and ulceroproliferative growth at tongue lateral border (Fig. 1).

There was no clinically palpable lymphadenopathy in head and neck region and the other clinical examination was normal. Magnetic resonance imaging (MRI) scan (T2-weighted and short inversion time inversion recovery image) showed an approximately 52 × 29 × 29 mm^3 sized, irregular-shaped, heterogeneously hyperintense lesion with lobulated margin at anterior two-thirds of tongue lying predominantly on the right side of midline and extending into left side of tongue at anterior one-third region which appear hypointense on T1-weighted images (Fig. 2). Initially, the incisional biopsy was done from the lesion.

The histopathological examination showed subepithelial predominantly spindle to oval cells loosely arranged in hyalinized stroma admixed with prominent inflammatory cells comprising predominantly plasma cell, lymphocytes, and eosinophils, few mitotic figures with foci of sclerosed area and dilated and congested blood vessels, features suggestive of IMT (inflammatory

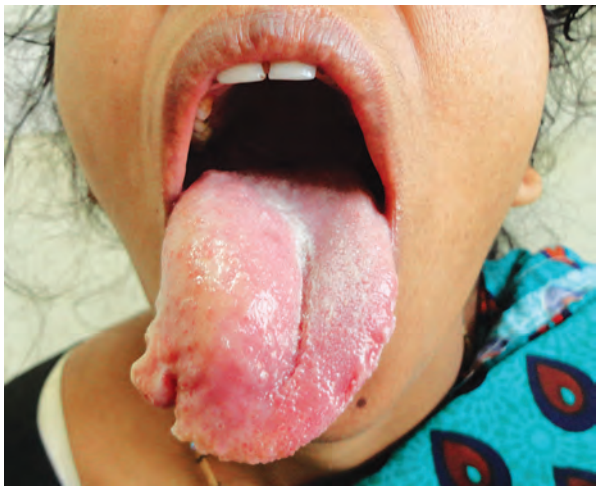
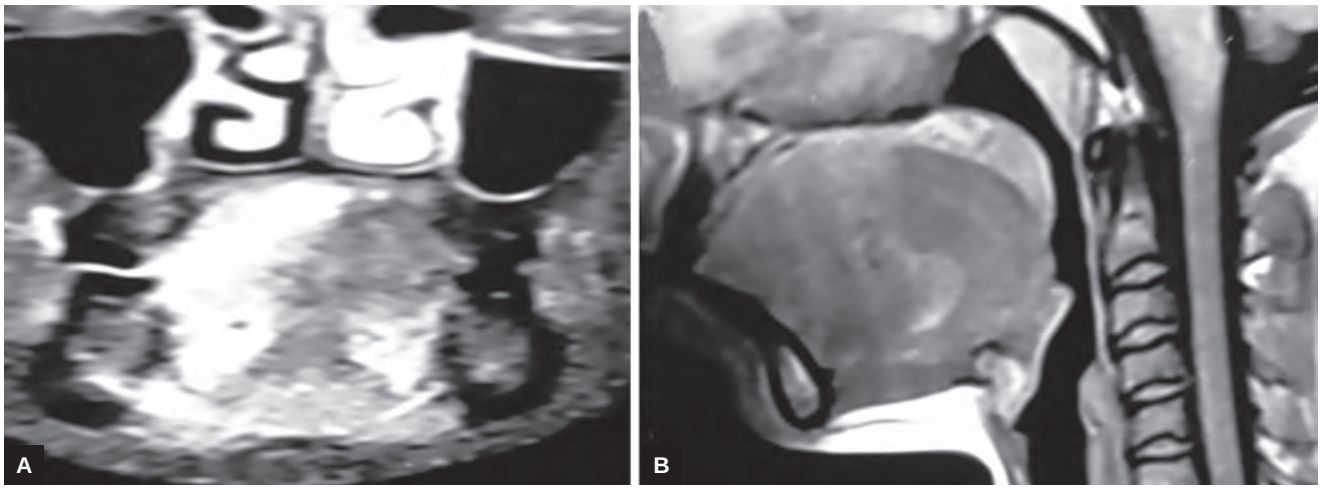


Fig. 1: Clinical photograph showing the lesion



Figs 2A and B: A coronal and sagittal MRI scan of oral cavity showing the lesion of the tongue

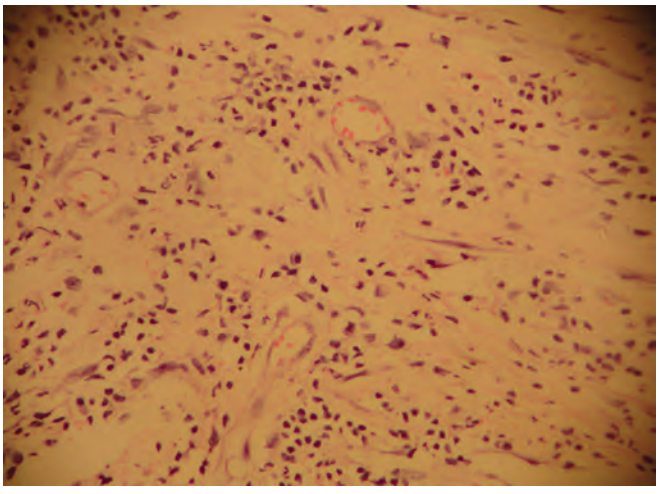


Fig. 3: Histological section showing a proliferation of spindle cells admixed with numerous chronic inflammatory cells (neutrophils, lymphocytes, macrophages, and plasma cells) in a fibrous and myxoid background (hematoxylin and eosin)

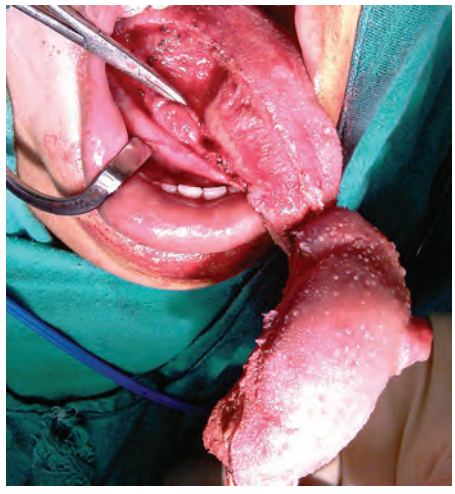


Fig. 4: Intraoperative photograph of the lesion

pseudotumor) (Fig. 3). Patient was managed with wide local excision of tumor with primary repair under general anesthesia (Fig. 4).

The surgically resected tumor was sent for histopathological examination. Again, the diagnosis of IMT was confirmed and the tumor was shown to have free margins. In 2 years follow-up, the patient is doing well and satisfied. There was no evidence of recurrence (Fig. 5).

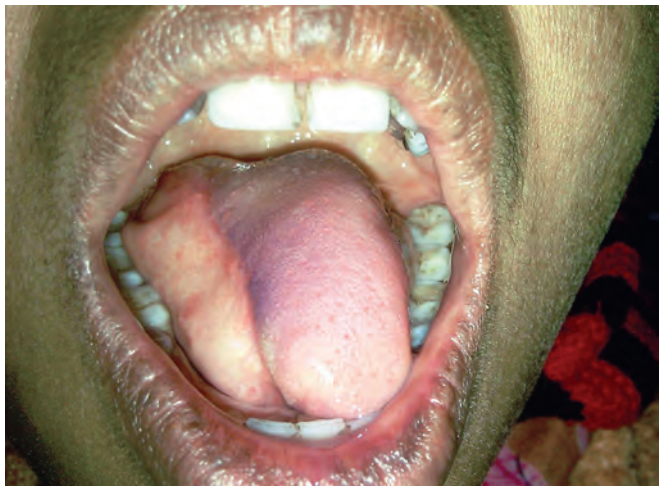


Fig. 5: The outcome after 1 year follow-up

DISCUSSION

Inflammatory myofibroblastic tumor of tongue is extremely rare constituted by a proliferation of myofibroblasts together with inflammatory cells intermingled within collagen fibers. Historically, this tumor is known by several names, such as plasma cell granulomas, inflammatory pseudotumor, and inflammatory myohistiocytic proliferation.[3] Inflammatory myofibroblastic tumor is rarely found in head and neck region, though it is found anywhere in the body and is commonly used to be found in the abdominal cavity and extremities.[2] Inflammatory myofibroblastic tumor may occur at any

age group but commonly occurs in the lung of young adults and children.[4]

This tumor very rarely occurred in the oral cavity. In a recent review of the literature, only 15 cases were reported by Brooks et al[5] in the various location of oral cavity. The common area of involvement in oral cavity is cheek and the mandible.[6] In our recent search of IMT, we have encountered very few cases located on the tongue[7-9] as the case herein presented.

Various pathogenic factors have been accused for the causes of the IMT. However, its cause remains unclear. It is hypothesized that stimulation of an inflammatory response with proliferation of reactive tissue myofibroblasts participation is repeated trauma or chronic irritation. According to this, oral IMT may be followed by repeated trauma to oral mucosal trauma.[10] Oral IMT does not have any systemic manifestation like visceral counterpart.[6]

Histologically, IMT consists of myofibroblastic cells in a collagenous stroma admixed with inflammatory cells, predominantly plasma cell and lymphocytes.[11] Diagnostic difficulty always exists due to relative proportion of myofibroblastic and inflammatory component, which may be confused with the reactive process of sarcomatous changes.

Immunohistochemistry is important for the final diagnosis and the spindle cells are usually positive for vimentin, desmin, muscle-specific actin, and smooth-muscle actin (SMA).[5,11] The tongue IMTs which we have reported having vascular and myxoid histological pattern and spindle cells were highly positive for vimentin and SMA.

Oral cavity IMT used to be treated as benign lesion with very good result.[5] Wide local excision of the tumor has showed excellent outcome without any recurrence. Steroid therapy, radiotherapy, curettage, and chemotherapy are the different modalities of treatment found in the literature with variable outcome.

Frequent follow up is mandatory as a cancer patient due to early detection of the tumor those truly behave like malignant lesion. Very few cases were reported as a spontaneous regression of IMT.[5,11] This case was also treated with wide local excision with primary repair with excellent outcome. In 1 year follow-up, the patient is doing well and satisfied without any evidence of recurrence.

CONCLUSION

Clinical as well as radiological behaviors of IMT are aggressive, so it may be misdiagnosed as malignant tumor. Therefore, its accurate diagnosis is vital to prevent unnecessary radical resection.

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