

Inflammatory myofibroblastic tumor of anterior maxillary gingiva: An unusual clinical presentation

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Abstract

Inflammatory myofibroblastic tumor (IMT) is a rare benign neoplasm with variable clinical presentation. We hereby present a case of a 27-year-old female who presented to us with an unsuspecting gingival overgrowth in her anterior gingiva. This article aims to describe IMT as a rare intraoral entity which may involve the anterior maxillary gingiva, involving or sparing the underlying bone, as only a handful of such cases have been reported so far. Surgical excision is the mainstay of treatment. Biopsy and histopathological examination of even the smallest lesions is a must. Timely diagnosis and intervention can prevent severe morbidity that can arise if these lesions are left undiagnosed as aggressive management of lesions that become extensive is mandatory.

Keywords: Inflammatory myofibroblastic tumor, myofibroblast tumour, myofibroblastoma, oral manifestations, reactive fibroma

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Submitted: 08-Aug-2020, **Revised:** 28-May-2021, **Accepted:** 31-May-2021, **Published:** 28-Feb-2022

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is an infrequently reported, rare growth with indefinite etiopathogenesis with a varied clinical presentation.^[1] It is rare in the oral cavity^[2] and presents with a wide differential diagnosis and some cases are misdiagnosed as spindle cell tumors. Histologically in a variably myofibrotic background, inflammatory infiltrate is present.^[3]

The World Health Organization classifies IMT as tumor of intermediate potential attributed to their high recurrence potential and low risk of distant metastasis. Its clinical and radiological representation in larger lesions mimics a malignancy.^[1]

This case requires publication for its uniqueness as a rare case of IMT occurring on anterior maxillary gingiva with different clinical manifestations mimicking a simple gingival overgrowth. Furthermore, the importance of histological and immunohistochemical findings of any lesion; however, insignificant has been stressed upon, at the same time highlighting the importance of complete excision for preventing recurrence.

CASE REPORT

A 27-year-old female presented with a month-old history of a slow and painless growth in anterior maxilla. She

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How to cite this article: Goyal L, Rao S, Reddy GS, Agarwal P. Inflammatory myofibroblastic tumor of anterior maxillary gingiva: An unusual clinical presentation. J Oral Maxillofac Pathol 2022;26:S73-6.

Access this article online

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DOI:

10.4103/jomfp.jomfp_338_20

was systemically healthy with no history of medications. She had undergone surgery for it 8 months earlier but had recurred. She had diastema between her maxillary central incisors where a growth was evident. It was well-circumscribed, round, sessile and of the same color as adjoining mucosa [Figure 1a]. There was no evidence of bleeding or exudates. Radiographic examination was insignificant [Figure 1b].

A clinical diagnosis of gingival fibroma was made with differential diagnosis of peripheral ossifying fibroma/peripheral giant cell lesion. After written informed consent, the lesion was excised *en bloc* with adequate margins and open flap debridement with osteoplasty was performed under local anesthesia [Figure 2a]. The soft tissue specimen was forwarded for histopathological examination. The postoperative healing of the site was uneventful.

Macroscopic examination of the specimen revealed soft tissue mass about 2.0 cm × 1.0 cm × 0.5 cm. Microscopic examination using hematoxylin and eosin stains revealed highly cellular connective tissue stroma with plasma cell infiltrate [Figure 2b]. The sub-epithelium was composed of spindle cells (plump, elongated with vesicular nuclei) arranged in fascicles with focal areas of collagen deposition. Dense inflammatory exudate was evident with few eosinophils and occasional histiocytes. The immunohistochemistry was positive for anaplastic lymphoma kinase (ALK) [Figure 2c].

The lesion was thus diagnosed as IMT. Patient has been on regular follow-up postoperatively for over 2 years and has no evidence of recurrence [Figure 3].

DISCUSSION

This article aims to describe intraoral IMT as a rare entity as only a handful of cases involving anterior maxillary gingiva and sparing the underlying alveolar bone have been reported.^[2]

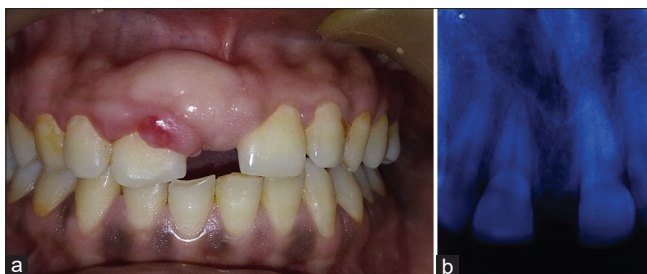


Figure 1: (a) Preoperative clinical presentation: Labial view showing gingival growth with midline diastema between two central incisors. (b) Preoperative radiographic presentation: Intraoral periapical radiograph showing no radiopacity or radiolucency

IMT has a prevalence of 0.04%–0.7%.^[4] It is commonly found in lungs, but among extrapulmonary sites with larynx is the most common.^[2,5,6] Oral IMT is rare with 0.0012% incidence with buccal mucosa and mandible being commonly involved.^[5-7]

Anterior maxillary gingiva is a rare site. Normally, there is a scarcity of muscle fibers in the labial frenum region. The presence of spindle cells with collagen in IMTs highlights the importance of the mimetic muscles present in the labial frenum in 35% of individuals, contributing to the myofibroblastic component of the lesion and explaining its presence in this unusual location and may be further investigated.^[7]

IMT is a neoplastic process resulting from chromosomal translocations that frequently lead to overexpression of ALK kinase, identified using immunohistochemistry.^[1] The majority of IMTs occur due to ALK gene rearrangements and less commonly due to viral infections such as EBV, HIV, HHV8, IgG4 related diseases, trauma, chronic inflammation, or auto-immune diseases.^[6] IMT is used as the universal terminology to address intermediate soft tissue tumors which comprise spindle cells with myofibroblast differentiation. It also presents with numerous inflammatory cells, plasma cells and/or lymphocytes.^[4]

The diagnosis of IMT is based on histopathological presentation. Differential diagnoses should include pyogenic granulomas, giant cell lesions, myofibroma, solitary and benign fibrous tumors, fibrosarcoma, leiomyoma, and mesenchymal odontogenic tumor.^[8]

Surgical excision and curettage is the management of choice but recurrence is high (15%).^[5,7,9] In localized gingival lesions, surgical excision has shown good results with the absence of recurrence in 10-year follow-up. Excision should be done down to the bone for prevention of recurrence or relapse of diastema. Other treatments include surgical excision with additional use of CO₂,^[7] steroid therapy, radiotherapy, or radical surgery.^[10] These modalities are considered for invasive, recurrent variants when there are signs of malignancy or positive margins. In 8%–18% cases malignant transformation occurs with metastasis in <5%. Its histological presentation is variable, so sometimes, multiple biopsies have to be taken and diagnosis has to be further confirmed by immunohistochemical staining.^[6]

Patient perspective should be kept in mind and the difference between this lesion and malignancy should

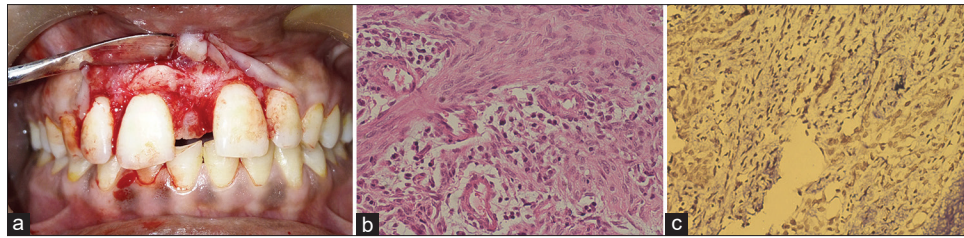


Figure 2: (a) Open flap debridement after excision of gingival growth on labial aspect of right central incisor. (b) Diagnostic method- Histopathological hematoxylin and eosin photomicrograph of lesion showing spindle cell bundles with inflammatory cell infiltrate. (c) Immunohistochemistry showing staining positive for anaplastic lymphoma kinase



Figure 3: Follow-up 2 year postoperatively with no recurrence

be explained while managing such lesions. Psychiatric counseling should be done whenever aggressive management is required. It is important to educate and motivate the operated patients to come for follow-up as recurrences are common even after clear surgical excision of tumor margins. Cultured tumor cells have stem cell properties and are the probable cause of recurrence.^[7] Due to its high recurrence potential, this pseudotumor has a disputed nomenclature and there is no fixed consensus about its treatment plan.

The strengths of this article include the unusual clinical presentation of the lesion and the importance of histopathological evaluation even for seemingly insignificant growths. Limitation of this case was the inability to esthetically rehabilitate the patient in terms of diastema closure, due to patient unwillingness.

IMTs should be considered a differential of common gingival pathologies such as pyogenic granulomas and giant cell lesions due to similar presentation. Regular monitoring is mandatory to rule out recurrence, malignant transformation, or metastasis.^[2] Smallest of gingival outgrowths should be sent for histopathological examination for the early diagnosis. Proper history, prompt biopsy, and excision are important to prevent complications

following subsequent extensive bony involvement. Timely diagnosis and intervention prevent morbidity that can arise following aggressive management of the larger/advanced lesions. Incorrect diagnosis and inability to understand its variable presentation may lead to unnecessary aggressive surgical treatment and functional disability that inadvertently follows.

CONCLUSION

IMT is a rare differential of seemingly insignificant anterior gingival outgrowths. The early diagnosis and excisional biopsy are the mainstay of treatment. Follow-up is crucial to prevent recurrence due to the aggressive nature of this disease.

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.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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