

Inflammatory Pseudotumor of Premaxillary Subcutaneous Tissue

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ABSTRACT

Inflammatory pseudotumor also known as plasma cell granuloma is a space occupying lesion which occurs in wide variety of somatic tissue and visceral organs, including lungs, liver, thyroid and many other organs. Recently, it has been described in the major salivary glands and buccal tissue in children.¹ We have described here the clinical, radiological and histological findings of inflammatory pseudotumor in premaxillary subcutaneous tissue in an adult, which is a rare site.

Keywords: Inflammatory pseudotumor, Plasma cell granuloma.

INTRODUCTION

Inflammatory pseudotumor is a tumor-like lesion characterized histologically by fascicles of plump spindle cells in a background of chronic inflammatory cells, notably plasma cells. Other synonyms have been used to describe similar lesions, such as plasma cells granuloma, histiocytoma, xanthomatous granuloma, inflammatory myofibroblastic tumor and spindle cell pseudotumor.¹

These lesions are benign; however, misdiagnosis as malignant tumor can lead to vigorous treatment and serious consequences. Its correct recognition by the pathologist is important to avoid unnecessary radical and potentially mutilating surgery. Prognosis is excellent after complete excision.²

CASE REPORT

A 25-year-old male presented in ENT OPD of MGM hospital with one month history of swelling over the left cheek. The swelling was insidious in onset, gradually progressed to involve the whole maxillary region of the left side of face extending from infraorbital region to the upper lip on left side and was associated with mild pain and fever for initial two days.

After one week of conservative treatment with anti-inflammatory drugs and antibiotics, swelling reduced considerably in size with some remnant swelling in left lower maxillary region near the nasolabial fold. After few days, patient noticed a small, irregular, painless growth in the left upper gingivobuccal sulcus that sometimes bled while brushing teeth.

On external examination, diffuse swelling was present over left lower half of maxilla, near the nasolabial fold, measuring approximately 3 × 3 cm in dimensions with smooth surface, no visible scars, sinuses or redness.

On palpation, a hard swelling approximately 2.5 × 2.5 cm was palpable which was fixed and nontender. Skin over the swelling was free.

Intraoral examination revealed 1cm granulation tissue, present in the upper left gingivobuccal sulcus adjacent to first and second premolar tooth, which was tender and bled on palpation. Underlying bony elevation was palpable.

Dental examination was normal. Anterior rhinoscopy showed hypertrophied inferior turbinate on the left side. Bilateral nasal cavities were equally patent with no discharge or swelling. Diagnostic nasal endoscopic findings were normal. No cervical lymphadenopathy. Systemic examination was normal.

On FNAC of intraoral tissue, highly cellular smear showed numerous histiocytic clusters, macrophages, plasma cells and osteoclastic giant cells against the background of polymorphonuclear cells. Few benign squames were also seen. Some cells showed atypia (high N:C ratio, prominent nucleoli). Findings were suggestive of inflammatory giant cell lesion with differential diagnosis of reparative giant cell granuloma.

Plain computed tomography (CT) scan of the paranasal sinus coronal sections (Figs 1A and B) showed well-defined subcutaneous fat density lesion in the left cheek region adjacent to left canine teeth, mild mucosal inflammatory changes involving left maxillary sinus and mild deviation of nasal septum to the right side with hypertrophy of bilateral inferior turbinate narrowing nasal airway.



Fig. 1A: Plain CT scan of the paranasal sinus coronal section showing well-defined subcutaneous fat density lesion in the left cheek region adjacent to canine teeth

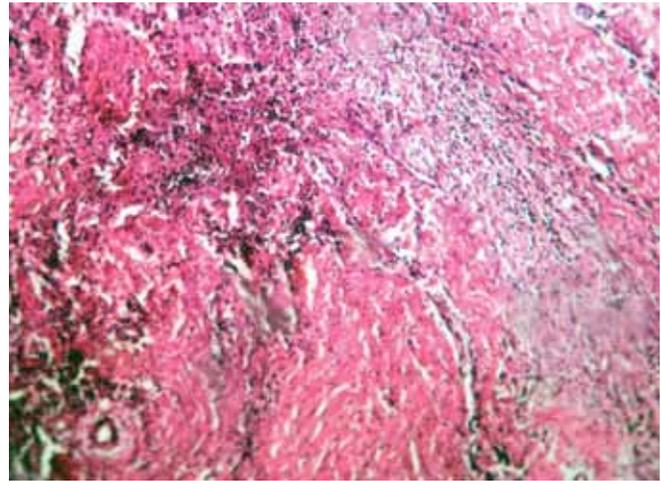


Fig. 3A: Low power histopathological microphotograph

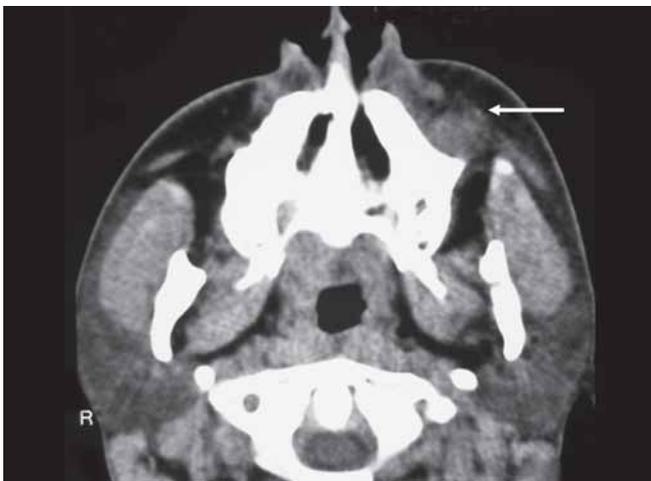


Fig. 1B: Plain CT scan of the paranasal sinus coronal showing well-defined subcutaneous fat density lesion in the left cheek region and mild mucosal inflammatory changes involving left maxillary sinus with intact anterior bony wall of maxilla

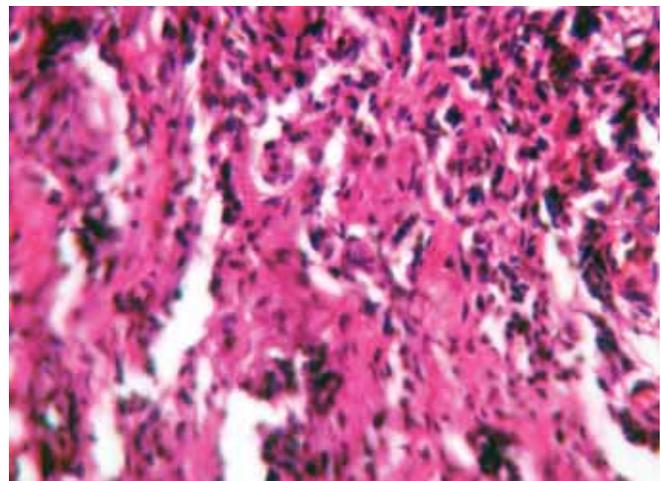


Fig. 3B: High power histopathological microphotograph



Fig. 2: Intraoperative picture showing firm tissue in the premaxillary subcutaneous tissue with granulation tissue over buccal surface

oval-shaped lesion in outer aspect of left maxillary alveolar arch with diffuse skin and subcutaneous tissue surrounding inflammatory changes.

Routine blood investigations were in normal limits.

Surgical exploration of the swelling was performed by intraoral approach. Incision was given in the left gingivobuccal sulcus surrounding the granulation tissue extending from the left canine tooth to the 2nd premolar (Fig. 2). Firm tissue was found in the subcutaneous tissue with the granulation tissue over the buccal surface, which was excised completely. There was no bony defect on the anterior wall of maxilla. The wound was sutured with catgut suture. Postoperative, oral antibiotic, anti-inflammatory drugs and gargles were given for 7 days.

On histopathological examination (Figs 3A and B), the microscopic H and E sections studied showed proliferation of the spindle cells (fibroblasts) with focal myxoid areas along with dense mixed (acute and chronic) inflammatory infiltrate. Numerous budding capillaries were seen along with few congested blood vessels. No evidence of malignancy was seen.

Patient was asymptomatic after 6-month follow-up.

Magnetic resonance imaging (MRI) of the face, using T1, T2 and FLAIR sequences, showed focal STIR hyperintense

DISCUSSION

First described in 1905 by Birch-Hirschfield, inflammatory pseudotumor remains somewhat of an enigmatic disease entity despite multiple otolaryngologic, radiologic and pathologic reports.³ The term pseudotumor was coined because these lesions mimic expansive, invasive, malignant tumors both clinically and radiologically. Inflammatory pseudotumors are clinically and histologically diverse group of lesions characterized by a tumor mass of acute and chronic inflammatory cells with a variable fibrous response.⁴ There are many synonyms, such as histiocytoma, xanthogranuloma, plasmocytic cell granuloma and inflammatory myofibroblastic proliferation, due to its wide histological presentation spectrum.⁵⁻⁷ These tumors have been reported to occur in every site in body. In the head and neck region, it is commonly found in the orbit, nasal cavity, nasopharynx, maxillary sinuses, larynx and the trachea.⁸ Inflammatory pseudotumor of the subcutaneous tissue in the premaxillary region is extremely rare.

The exact etiology of these lesions is not clear. However, various stimuli may act as triggers for its development, such as unrecognized microorganisms, minor trauma, smoking and chronic irritation caused by cocaine abuse.^{9,10}

It may represent a chronic response or a delayed presentation related to remote or undetected trauma.¹¹ It has been postulated that they might be the result of a postinflammatory repair process, a metabolic disturbance or an antigen-antibody interaction with an agent that was no longer identifiable in aspiration or biopsy material.¹⁻³

Inflammatory pseudotumor of the head and neck region is generally not associated with nonspecific systemic symptoms, such as unexplained fever, weight loss and laboratory abnormalities. CT scan or MRI of inflammatory pseudotumor in the head and neck region often suggests infiltrative growth and can mimic meningioma, malignant neoplasms, Wegner's granulomatosis and sarcoidosis. Histologically, these tumors are composed of myofibroblastic spindle cells, admixed with a prominent infiltrate of lymphocytes, plasma cells and mixed inflammatory cells. Three basic histological patterns have been described namely: (1) Myxoid/vascular pattern, resembling inflammatory granulation tissue; (2) Compact spindle cell pattern with fascicular and/or storiform areas and variation of cellular density and (3) Hypocellular pattern, densely collagenized and reminiscent of a fibrous scar.⁸ Immunohistochemistry confirms the myofibroblastic phenotype of spindle cells.⁹

Inflammatory pseudotumor is usually diagnosed by exemption since clinical and histopathologic findings are sometimes vague and inconsistent. It is generally accepted that a definite cytological diagnosis of inflammatory pseudotumor cannot be made by fine needle aspiration cytology because the predominant cell pattern is nonspecific.^{13,14} Total resection of the tumor is advisable even if it is small in size and asymptomatic because of its aggressive behavior in terms of destruction of surrounding structures. A case of pseudotumor of paranasal sinus with

fatal outcome as a result of extensive intracranial spread of tumor has also been reported.¹⁵

In this article, we present a case of inflammatory pseudotumor in premaxillary subcutaneous tissue which is a rare site. Inflammatory pseudotumor is a distinct pathological entity characterized by fascicles of spindle cells in an inflammatory background rich in plasma cells. Cytologic diagnosis on fine needle aspiration can rarely reach a definitive diagnosis and excisional biopsy is usually required. Treatment is complete surgical resection which is curative. Its correct recognition by surgical pathologist is, therefore, vital in order to avoid unnecessary extensive and radical surgery.

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