

CASE REPORT

Inflammatory pseudotumor of maxillary sinus: Mimicking as an aggressive malignancy

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ABSTRACT

Inflammatory pseudotumor is a rare entity in the head and neck region. Clinically it behaves as an aggressive lesion with extensive destruction but considered to be a benign reactive process. Because of its extensive involvement and infiltrative nature, it is confused as a malignancy. Most of them respond very well to corticosteroid and surgical excision.

Key words: Immunohistochemistry, inflammatory pseudotumor, maxillary sinus

INTRODUCTION

Inflammatory pseudotumor (IPT) are clinicopathologically distinctive but biologically controversial entities, which have been described in the lungs, abdomen, retroperitoneum, and extremities, but rarely affect the head and neck region.^[1] It has been described under many appellations including plasma-cell granuloma, plasma-cell pseudotumor, inflammatory myofibroblastic tumor, inflammatory fibrosarcoma, and most commonly IPT.^[2] Exact etiopathogenesis is not known, though it is considered to be an exaggerated inflammatory reaction to tissue injury of unknown cause.^[3] Though it has a benign clinical course, it is said that at least a subset of IPTs represents true neoplasia rather than reactive myofibroblastic proliferation.^[1]

CASE REPORT

A 26-yr-old man presented with sensitivity in the upper-right back teeth since 1 year, accompanied by pain in the right side of face, forehead, and palate. Pain was severe, getting aggravated during the nighttime and relieved on medication. Diffuse swelling was evident producing mild midfacial asymmetry. On palpation, it was soft in consistency and tender. Right submandibular lymph nodes were also tender on palpation. On intraoral examination, palatal perforation

was seen on the right side. Orthopantomograph revealed haziness of right maxillary sinus. Computed tomography (CT) showed complete opacification of right maxillary sinus with erosion of medial wall and floor [Figure 1]. A provisional diagnosis of aggressive lesion of maxillary sinus was considered.

Incisional biopsies revealed fascicles of spindle cells along with chronic inflammatory cell infiltrate predominantly plasma cells and lymphocytes. No fungal organisms were appreciated, both with periodic acid schiff PAS and silver staining. Spindle cells showed positive expression for vimentin [Figure 2] and smooth muscle actin [Figure 3]. These cells were negative for caldesmon and CD-68. The final diagnosis of inflammatory myofibroblastic tumor was confirmed. The lesion responded very well to corticosteroids and decreased in its size enormously. It was surgically excised, and on follow up, there is no recurrence since last 24 months.



Figure 1: CT - Para nasal sinuses showing soft tissue growth in the right maxillary sinus with erosion of medial wall and floor

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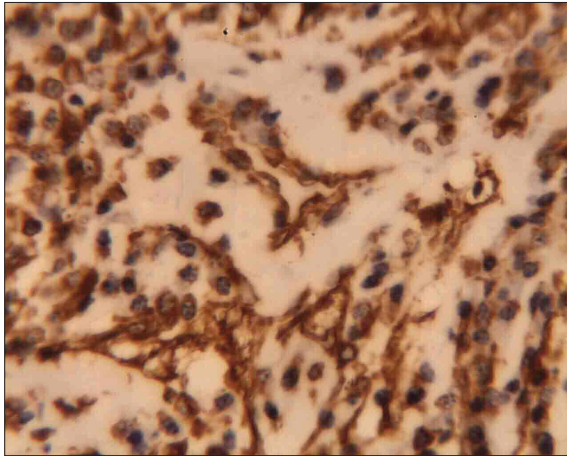


Figure 2: Spindle cells showing positivity to vimentin

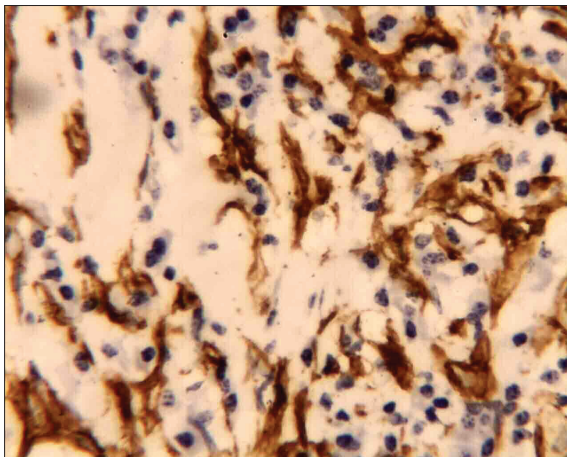


Figure 3: Spindle cells showing positive expression to alpha smooth muscle actin

DISCUSSION

IPT represents a spectrum of myofibroblastic proliferation that includes reactive to benign to malignant lesions. Some lesions are difficult to distinguish from the reactive process, where as others appear sarcomatous. Lesions may show neoplastic features such as persistent local growth, recurrence, and metastasis. Hence, few authors consider it as a low-grade sarcoma.^[2,4]

Histologically it mainly shows spindle cells in fascicles that represents myofibroblasts and inflammatory component which is predominantly lymphocytes and plasma cells.^[5,6] Spindle

cells showed positivity to vimentin, alpha-smooth muscle actin and negative to CD-68 (histiocytic), caldesmon (mature smooth muscle cells) proving these spindle cells originating from myofibroblasts.

Most of the IPTs reported in head and neck region showed a benign course. However cases related to paranasal sinuses seems to be showing highly aggressive behaviour, with poor response to surgery, radiotherapy, and chemotherapy, showing multiple recurrences and fatal outcome.^[1,2] In this case, lesion responded very well to corticosteroids and it was treated with wide surgical excision. The patient is being followed up and since last 24 months, there are no signs of recurrence.

CONCLUSION

Controversy still exists whether IPT is just a reactive process or a true neoplasia. However, it should be treated as a benign growth, unless it is showing marked destruction, cellular atypia, and frequent recurrences. Because of its unusual clinical and histopathological presentation, immunohistochemistry helps us to come to a definitive diagnosis and careful follow up of these cases are mandatory as its behavior is unpredictable.

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