10.5005/jp-journals-10013-1228

CASE REPORT

Rare Nasal Mass: Inflammatory Myofibroblastic Tumor of Nose and Paranasal Sinuses

[1]Jafferhusein Sura, [2]Anoop Raj, [3]Praveen Kumar Rathore, [4]Divya Sharma, [5]Nita Khurana
[6]Vikram Wadhwa, [7]Eishaan Bhargava

ABSTRACT

Inflammatory myofibroblastic tumor (IMFT) also known as pseudotumor is a condition of unknown etiology, that mimmicks a neoplastic process clinically and radiologically. They occur all over the body with few exceptions. In the head and neck region, they are rare. The orbit, maxilla, nasopharynx and skull base are sites where they have been reported. We present a case of a 32-year-old male having a mass in the cheek, which presented a diagnostic challenge.

Keywords: Management, Myofibroblastic tumor, Paranasal, Sinuses.

How to cite this article: Sura J, Raj A, Rathore PK, Sharma D, Khurana N, Wadhwa V, Bhargava E. Rare Nasal Mass: Inflammatory Myofibroblastic Tumor of Nose and Paranasal Sinuses. Clin Rhinol An Int J 2015;8(1):38-40.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Inflammatory myofibroblastic tumor (IMFT) is an entity known by a variety of names like inflammatory pseudotumor[1] and plasma cell pseudotumor.[2] It is known to occur in almost every anatomic location, but is rare in the head and neck region. Here, we describe a case occurring in the nose and paranasal sinuses.

CASE REPORT

A 32-year-old male presented with complaints of nasal obstruction for 6 months. He had episodes blood tinged nasal discharge during the same period. He noticed a swelling over the right cheek for the last 4 months, which gradually increased in size though there was no pain. He then had pain in his right upper molars for the last 3 months and noticed the loosening of his teeth and the formation of an ulcer in the region of the upper alveolus, which gradually increased in size (Fig. 1).

On examination, he had a swelling over his right cheek of 4 × 4 cm which extended from the nasal ala to 2 cm in front-of the tragus and from below the inferior orbital margin to the upper-lip. The swelling had normal overlying skin, non-tender, normal temperature and was firm on palpation. The nasal bridge was depressed and on anterior rhinoscopy there was blood tinged nasal discharge in both nasal cavities. There was a mass seen in the right nasal cavity and upper part of left nasal cavity on nasal endoscopy, which was firm and bled on touch during probing. There was proptosis of both eyes. On oral examination, an ulcer measuring 2 × 2 cm was seen in the region of the right 1st to 3rd molars of upper alveolus which was firm and did not bleed on touch.

The patient underwent a computed tomographic scan, which revealed a mass involving the bilateral frontal sinuses, ethmoid sinuses, and right maxillary sinuses and right cheek and right side of hard palate (Fig. 2).

On clinical suspicion of malignancy, the patient underwent a biopsy, which was inconclusive. After two more repeat biopsies which were also inconclusive for malignancy. Even though the biopsies were not conclusive for malignancy, based on a strong clinical and radiological suspicion of malignancy, the patient was taken up for surgery. Intraoperatively, another biopsy was sent for frozen section, which was reported as an inflammatory swelling. At that stage, based on a clinical suspicion of inflammatory myofibroblastic tumor, a surgery excising all of the tumor was done. Postoperatively, the patient has no recurrence at 3 months after surgery.

Histopathological sections examined from various areas of the lesion showed hypocellular to moderately cellular lesion comprised of spindled to stellate shaped cells dispersed in a moderately to densely collagenised stroma. The cells showed normochromatic nuclei, inconspicuous nucleoli, infrequent mitosis. There were no areas of atypia or necrosis. The stroma showed few small calibre blood vessels and scattered inflammatory cells chiefly plasma cells, lymphocytes and eosinophils. Also noted were a

[1,4]Senior Resident, [2,3,5]Director and Professor, [6]Specialist
[7]Junior Resident

[1-3,6,7]Department of Otorhinolaryngology, Maulana Azad Medical College and Lok Nayak Hospital, New Delhi, India

[4,5]Department of Pathology, Maulana Azad Medical College New Delhi, India

Corresponding Author: Jafferhusein Sura, Senior Resident Department of Otorhinolaryngology, Maulana Azad Medical College and Lok Nayak Hospital, New Delhi, India, Phone: 9013588442, e-mail: dr.jafferhuseinsura@gmail.com

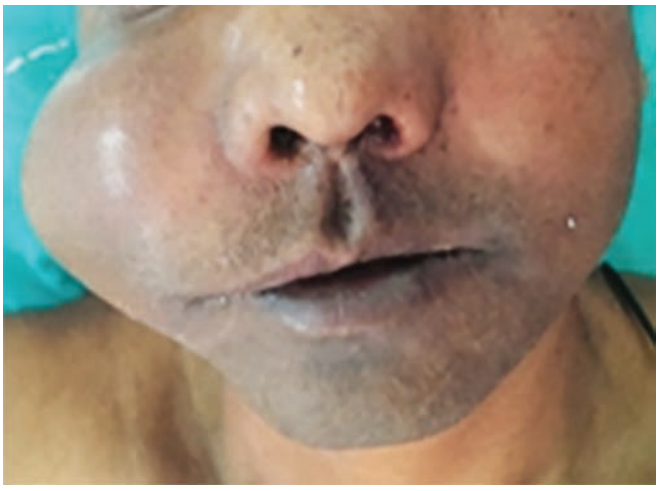


Fig. 1: Clinical profile

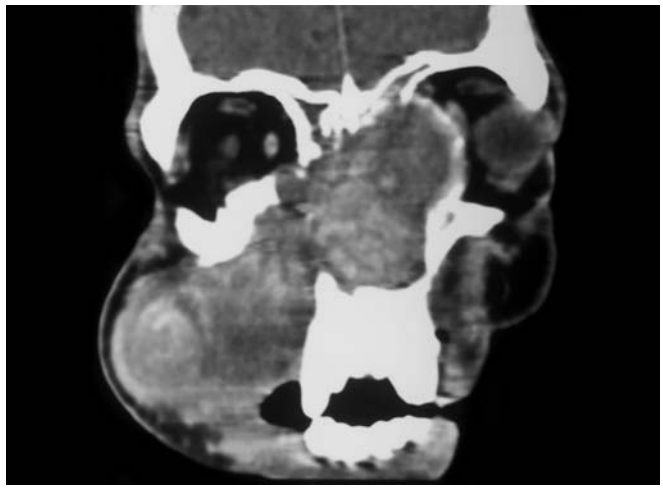


Fig. 2: Computed tomography scan of nose and PNS coronal sections showing the extent of tumor

few loose aggregates of plasma cells in the collagenised background. Overlying nasal mucosa was stretched out with foci of ulceration. Immunohistochemistry showed immunoexpression of Vimentin and smooth muscle actin in the spindle cells and both kappa and lamda in the plasma cells. Based on the histological and immunohistochemical features, a diagnosis of inflammatory myofibroblastic tumor was rendered (Figs 3 and 4).

DISCUSSION

The term IMFT was first proposed by Pettinato et al in 1990.[3] This lesion has been described in lung,[3] liver,[4] thyroid,[5] salivary glands[6] and temporal bone.[7] The most common sites in the head and neck region are the orbits[8] but it has also been reported in the maxillary sinus, infratemporal fossa, nasopharynx pterygopalatine fossa and skull base.[9-12] The most common extrapulmonary sites are the omentum and intestines.

Inflammatory pseudotumor of the cheek and oral cavity tends to occur more commonly in children and young adults, with the cheek and mandible being the sites of predilection.[13] It has been reported in the maxillary sinus but is extremely rare.[14] Compared to their counterpart in the abdominal viscera, the head and neck inflammatory pseudotumor does not produce systemic symptoms.

The cause of IMFT is not clearly known and microbiological cultures are usually negative. As the term IMFT suggests, these lesions are not clearly neoplastic. But evidence is there to suggest that these lesions are neoplastic albeit benign. Clonal cytogenetic abnormalities have been reported in some cases.[15-17] Histologically, inflammatory pseudotumor of oral cavity is similar to that of other organs. There are variable proportions of spindle cells and plasma cells and myofibroblasts and eosinophils. Cells are variably immunoreactive for smooth muscle actin and β-catenin, and were negative for ALK1, CD34, and EMA.[14,18]

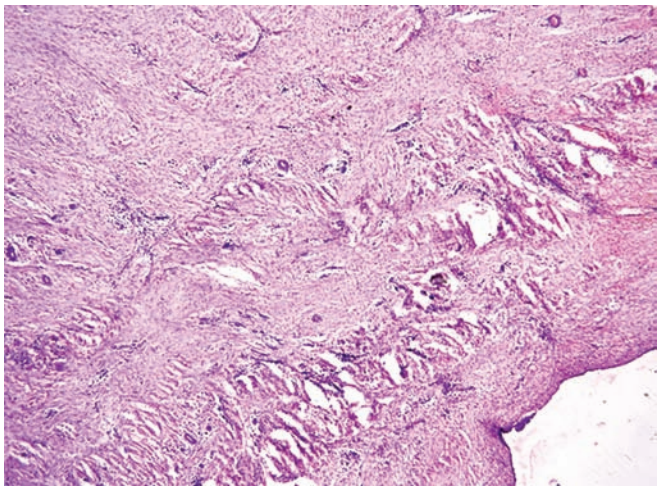
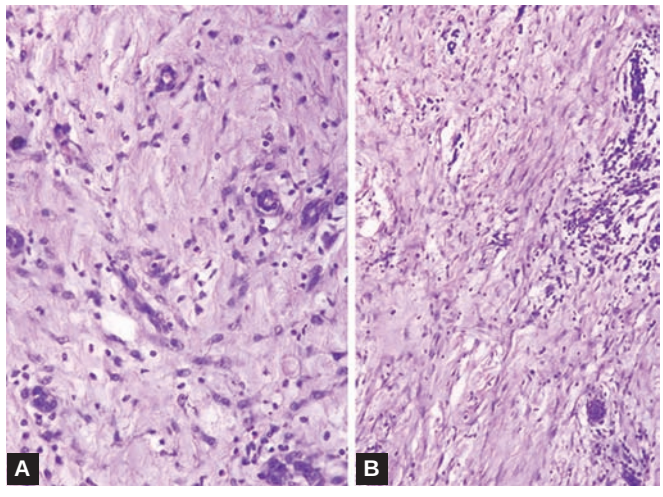


Fig. 3: Histopathology showing aggregates of plasma cells in the collagenized background



Figs 4A and B: Immunocytochemistry showing Vimentin and smooth muscle actin in the spindle cell swith kappa and lamda in the plasma cells

It is very difficult to clinically and radiologically differentiate IMFT from a truly malignant neoplasm.[8-12] In extraorbital head and neck locations of IMFT, it exhibits a more aggressive behaviour with bone erosion and bone remodelling clearly evident.[9-12] A recent study of 10 cases of hepatic IMFT also showed that imaging alone suggested the nature of the lesion to be intermediate grade malignancy. Hence, imaging alone could not be sufficient to diagnose the case.[19]

Based on the clinical features and radiological picture, it appears to be a malignant lesion, but biopsy is inconclusive for the same. Since, fine needle cytology is also not conclusive, the mainstay of treatment is surgical excisional biopsy. No postoperative radiotherapy is required, but a close follow-up of the patient is necessary.

CONCLUSION

Inflammatory myofibroblastic tumor of the maxillary sinus is extremely rare and tend to mimic a carcinoma clinically and radiologically. Since, it is difficult to diagnose histopathologically as well, a strong clinical suspicion for this lesion is essential when faced with such a patient, and a decision to operate must be taken as surgical excision provides a cure.

REFERENCES

1. Scully RE, Mark EJ, McNeely BU. Case records of the Massachusetts General Hospital. Case 13-1984. Cellular inflammatory pseudotumor involving ileal mesentery. N Engl J Med 1984;310:839.
2. Piscotto PT, Gray GF, Miller VR. Abdominal plasma cell pseudotumor. J Pediatr 1978;93:628.
3. Pettinato G, Manivel JC, De Rosa N, Dehner LP. Inflammatory myofibroblastic tumor (plasma cell granuloma): clinicopathological study of 20 cases with immunohistochemical and ultrastructural observations. Am J Clin Path 1990;94:430-435.
4. Shek TWH, Ng IOL, Chan KW. Inflammatory pseudotumor of liver. Am J Surg Pathol 1993;17:231-238.
5. Chan KW, Poon GP, Choi CH. Plasma cell granuloma of the thyroid. J Clin Pathol 1986;39:1105-1107.
6. Williams SB, Foss RD, Ellis GL. Inflammatory pseudotu, ors of the major salivary glands. Am J Surg Pathol 1992;16:896-902.
7. Gasparotti R, et al. Inflammatory Myofibroblastic tumor of temporal bone. Am J Neuroradiol 2003;24:2092-2096.
8. Weber AL, Romo LV, Sabates NR. Pseudotumor of the orbit: clinical, pathologic and radiologic evaluation. Radiol Clin North Am 1999;37:151-168.
9. Som PM, et al. Inflammatory pseudotumor of maxillary sinus: CT and MR findings in six cases. Am J Roentgenol 1994;163:689-692.
10. Du Vuysere S, et al. Extraorbital inflammatory pseudotumor of the head and neck: CT and MR findings in three patients. Am J Neuroradiol 1999;20:1133-1139.
11. Riberio AC, et al. Inflammatory myofibroblastic tumor involving pterygopalatine fossa. Am J Neuroradiol 2001; 22:518-520.
12. Han MH, Chi JG, Kim MS, et al. Fibrosing inflammatory pseudotumors involving the skull base: MR and CT manifestations with histopathologic comparison. M J Neuroradiol 1996;17:515-521.
13. Shek AWH, Wu PC, Samman N. Inflammatory tumor of mouth and maxilla. J Clin Path 1996;49:164-167.
14. Kim SY, Yang SE. Inflammatory myofibroblastic tumor of the maxillary sinus related with pulp necrosis of maxillary teeth: case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011 Nov;112(5):684-687.
15. Treissman SP, et al. Omental-mesenteric inflammatory pseudotumor. Cytogenetic demonstration of genetic changes and monoclonality of one tumor. Cancer 1994;73:1422.
16. Su L, Sheldon S, Weiss SW. Inflammatory myofibroblastic tumor: cytogenetic evidence supporting clonal origin. Mod Pathol 1995;8:12A.
17. Snyder CS, et al. Clonal changes in inflammatory pseudotumor of the lung: a case report. Cancer 1995;76:1545.
18. Karakok M, et al. Inflammatory myofibroblastic tumor (inflammatory pseudotumor) of the maxillary sinus mimicking malignancy: a case report of an unusual location (is that a true neoplasm?). Auris Nasus Larynx 2002 Oct;29(4): 383-386.
19. Xiao Y, at al. Radiological and histopathological features of hepatic inflammatory myofibroblastic tumour: analysis of 10 cases. Clin Radiol 2013 Nov;68(11):1114-1120.