Case Report

Benign fibrous histiocytoma of the maxilla: A rare case report and literature review

ABSTRACT

Benign fibrous histiocytoma (FH) is a benign soft-tissue tumor that can present as a fibrous tissue mass anywhere in the body. The involvement of the paranasal sinuses is extremely rare, and very few cases have been reported in literature till date. We here report a case of benign FH localized in the maxillary sinus. The clinical and histological features of the lesion are discussed with a brief literature review of this pathology in the paranasal sinuses.

Keywords: Benign fibrous histiocytoma, head-and-neck histiocytoma, maxillary sinus, paranasal sinus

INTRODUCTION

Fibrous histiocytoma (FH) is a benign tumor composed of a mixture of fibroblastic and histiocytic cells.[1] These lesions most often arise on the skin but may rarely occur in deep tissues. The involvement of the paranasal sinuses is very rare.[1-4]

FH is reported to present at any age with predominance in male adults (2.5:1) with a mean age of 40 years.[2,5]

The diagnosis of FH may be clinically difficult and challenging because of the spectrum of features, which frequently overlaps with other benign and malignant tumors, especially when the lesion is located in the deep tissues and is confirmed after the biopsy.[6-8]

The development of immunohistochemical techniques and electronic microscopy during the past 30 years has allowed us to differentiate between malignant and benign forms; consequently, benign FH (BFH) and malignant FH became a new clinical entity.[9]

We herein report a rare case of BFH of the maxillary sinus in a 45-year-old man who presented with nasal obstruction, epistaxis, and proptosis. The initial biopsy and later immunohistochemistry after proper surgical excision revealed BFH.

Access this article online	
Website: www.njms.in	Quick Response Code
DOI: 10.4103/njms.NJMS_85_18	

CASE REPORT

A 45-year-old man presented with a 6-month history of nasal obstruction, epistaxis, proptosis, and left cheek swelling. On clinical examination, a firm fleshy polyp-like mass was seen in the left nasal cavity. Diagnostic nasal endoscopy of the right nasal cavity revealed that choanae were free. No lymphadenopathy was seen. Rest of general physical examination was normal. There was no history of trauma or irradiation [Figure 1].

Ophthalmologic consultation revealed normal visual acuity according to age and normal extraocular muscle functions. Computed tomography (CT) of the paranasal sinuses revealed soft-tissue density measuring 5 cm × 4 cm × 3 cm involving

Jitendra Pratap Singh Chauhan, Abhay Kumar, Prateek Kumar Porwal, Rajesh Kumar Thakur[1], Kailash Mittal[2], Harshita Singh[3]

Departments of ENT, [1]Dental and [2]Radiotherapy, UPUMS, Etawah, Uttar Pradesh, [3]Department of ENT, Tata Main Hospital, Jamshedpur, Jharkhand, India

Address for correspondence: Dr. Prateek Kumar Porwal, Room No. 101, SR Hostel, DTH, UPUMS, Saifai, Etawah - 206 130, Uttar Pradesh, India.
E-mail: mbbsbtech@gmail.com

Received: 14 November 2018, **Revised:** 03 January 2019, **Accepted:** 30 April 2019, **Published:** 16 December 2020

How to cite this article: Singh Chauhan JP, Kumar A, Porwal PK, Thakur RK, Mittal K, Singh H. Benign fibrous histiocytoma of the maxilla: A rare case report and literature review. Natl J Maxillofac Surg 2020;11:298-301.

the left maxillary sinus and few anterior and posterior ethmoidal air cells. Mass was pushing the nasal septum to the right side and eventually extending into the pterygopalatine fossa, masseteric space, and infratemporal fossa with the bony destruction of the pterygoid plates and medial, lateral, posterior, and superior walls of the maxillary sinus with the erosion of the orbital floor [Figure 2].

The lesion had been increasing slightly in size over the past 6 months.

A diagnostic biopsy was performed by hematoxylin and eosin staining at low power (×200) which showed fibrocollagenous tissue lined by cuboidal epithelium which comprises mildly pleomorphic plump to spindle cell arranged in short intersecting fascicle forming wage storiform pattern. High-power (at ×400) view showed pleomorphic spindle-shaped fibroblasts which had features of hyperchromatism, altered nuclear–cytoplasmic ratio, and mitotic figures with multinucleated giant cells with thin elongated nuclei and pointed ends. Large atypical pleomorphic histiocytes were seen in the background [Figure 3].

After proper investigations and preanesthetic checkup, we posted him for functional endoscopic sinus surgery (FESS) and endoscopic medial maxillectomy under general anesthesia. Grossly, left maxillary bone was involved with tumor size of 5 cm × 4 cm × 3 cm on CT of the paranasal sinuses [Figure 4]. The cut surface was gray white, fleshy with infiltration of bone. Sections from routine formalin-fixed paraffin-embedded tissue stained with hematoxylin and eosin revealed ovoid-to-spindle-shaped fibroblastic cells in fascicles as well as a storiform pattern. Histiocytes and multinucleated giant cells were also noted.

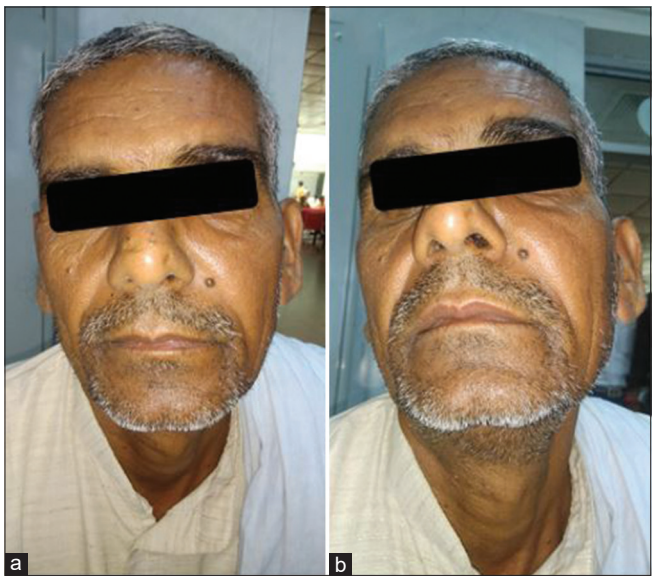

Figure 1: (a) Swelling of the left cheek and (b) proptosis of the left side

On immunohistochemistry, tumor cells were positive for vimentin, CD34, and SMA. They were negative for desmin, S-100, and Ki-67 [Figure 3].

Clinicoradiological correlation along with cytology, histopathology, and immunohistochemistry helped us to establish the final diagnosis of BFH.

The patient underwent radiotherapy and was doing well as on the 6th month.

DISCUSSION

BFH was unknown as a clinical entity before 1970; now, as a result of the development of immunohistochemical techniques and electronic microscopy, differential diagnosis became more feasible.[1] The etiology of oral BFH is obscure. Chronic irritation, continuous trauma, and spontaneous development have been reported for those located within the oral cavity.[5] According to the WHO, BFH is rare, with <100 reported cases. Patients have ranged in the age from 6 to 74 years at diagnosis.[10]

According to the WHO histological classification of the tumors, BFH is defined as a benign lesion with rare mitosis and the absence of cellular atypia and composed of spindle-shaped fibroblasts arranged in a unique storiform

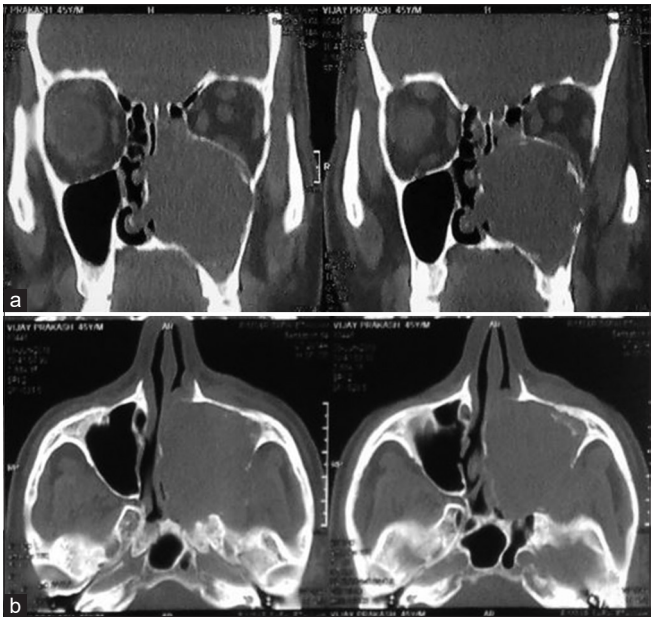

Figure 2: Computed tomography scan showing (a) coronal section revealing soft-tissue density measuring 5 cm × 4 cm × 3 cm involving the left maxillary sinus, few anterior and posterior ethmoidal air cells, pterygopalatine fossa, and infratemporal fossa with the bony erosion of the superior wall of the maxillary sinus and orbital floor. (b) Axial section mass pushing the nasal septum to the right side and extending into the pterygopalatine fossa and erosion of the medial, lateral, and posterior walls of the maxillary sinus

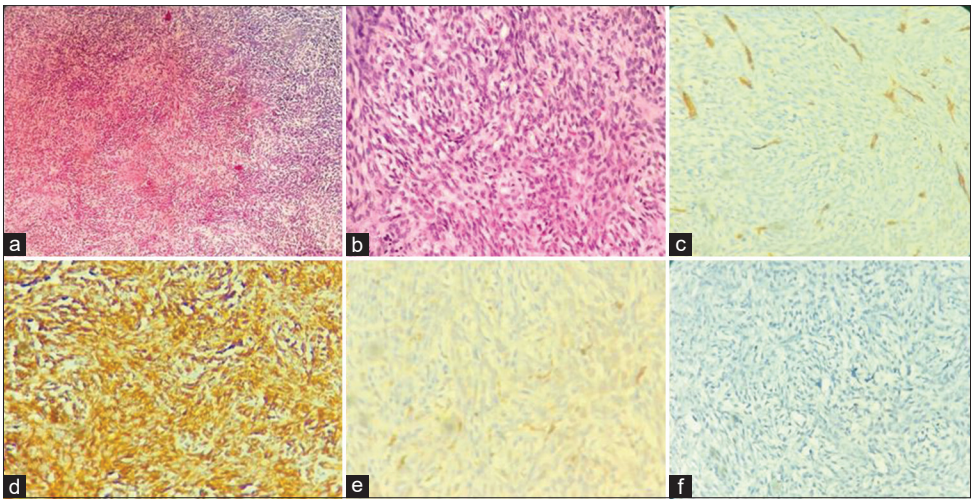

Figure 3: (a) Benign fibrous histiocytoma showing a mixture of fibroblastic, myofibroblastic-like, and histiocytic cells arranged in a vague storiform pattern (H and E, ×200). (b) Benign fibrous histiocytoma showing spindle cells having scant cytoplasm, thin elongated nuclei, and pointed ends (H and E, ×400). (c) Immunohistochemistry for smooth muscle antigen is positive in blood vessels (×400). (d) Immunohistochemistry for vimentin is diffusely and strongly positive (×400). (e) Immunohistochemistry for CD34 is positive in blood vessels. (f) Immunohistochemistry for Ki67 shows very low proliferation seen as nuclear positivity in <1% cells (×400)

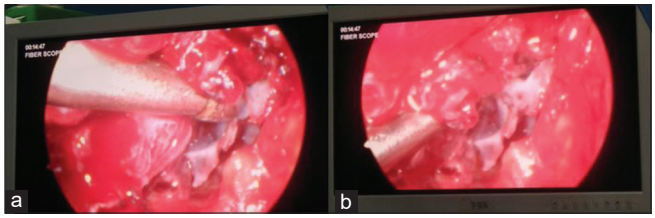

Figure 4: Endoscopic medial maxillectomy showing a mass in (a) the maxillary antrum, (b) excising small tissue for histopathology examination

pattern with a variable admixture of small, multinucleated, osteoclast-like giant cells. Foamy cells (xanthoma), chronic inflammatory cells, stromal hemorrhages, and hemosiderin pigment are also commonly present. In our case, it showed spindle-shaped fibroblasts arranged in a storiform pattern, histiocytes, and giant cells.[8,9]

Immunohistochemical staining was done in our cases which showed that the tumor cells were positive for vimentin, CD34, and SMA and negative for S-100 protein, epithelial membrane antigen, cytokeratin, and desmin. The positivity for CD34, CD68, and vimentin indicated that the lesion is composed of histiocytic cells and fibroblast-like cells on immunohistochemistry, and the negativity for SMA and S-100 showed that the lesion could be differentiated from leiomyosarcoma and neurogenic tumors.[2] Positivity with vimentin and negative staining with S-100 protein were consistent with the findings of Kanazawa *et al*. and Menditti *et al*.[2,5]

CT of the paranasal sinuses should be requested if the involvement of bone is suspected. Magnetic resonance imaging studies are used in case of soft-tissue involvement. Remarkably, few studies have shown the involvement of the maxillary sinuses [Table 1].

The primary treatment for FH is surgical excision with or without radical neck dissection.[5,12] As the incidence of regional lymph node metastasis is relatively low (4%–17%), elective neck dissection is performed only when there is evidence of metastasis.[5] In our patient, FESS with medial maxillectomy was carried out without radical neck dissection as there was no evidence of regional lymphadenopathy.

The prognosis of oral BFH is excellent. Metastases have not been reported. Typically, the treatment plan for BFH consists of wide surgical resection as mentioned in the literature, but considering the location (involving orbital floor) and extension of the lesion, we decided to do FESS with endoscopic medial maxillectomy along with curettage instead of complete open resection by open approach.

In this case, surgery with wide excision remains the primary treatment which can be followed by radiotherapy.

CONCLUSION

A rare case of BFH occurring in the maxillary sinus of a 45-year-old male is reported. As the lesion has histopathologic similarity with other lesions, clinical and radiographic correlation is essential for differential diagnosis. The prognosis for BFH is excellent, and recurrence is very rare. The treatment plan will vary from curettage to wide excision, depending on

Table 1: Various studies showing the involvement of the maxilla with fibrous histiocytoma

Author	Location	Age/Sex/ Number of cases	Operation	Publication Date/ Study period
Cale *et al.*[3]	Maxilla	13 year/M	Surgical excision	1989
Besly *et al.*[11]	Maxilla	2 cases	Surgical excision	1993
Shahoon *et al.*[12]	Maxilla	36 year/M	Surgical excision	2001-2009
Kasat *et al.*[13]	Maxilla, hard palate	46 year/F	Surgical excision	2014
Saluja *et al.*[14]	Maxilla	23 year/F	Surgical excision	2014

various factors such as the area involved and the age of the patient.

Consent

Written informed consent was obtained from the patient for publication of this case and accompanying images.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Rice DH, Batsakis JG, Headington JT, Boles R. Fibrous histiocytomas of the nose and paranasal sinuses. Arch Otolaryngol 1974;100:398-401.
2. Kanazawa H, Watanabe T, Kasamatsu A. Primary malignant fibrous histiocytoma of the mandible: Review of literature and report of a case. J Oral Maxillofac Surg 2003;61:1224-7.
3. Cale AE, Freedman PD, Kerpel SM, Lumerman H. Benign fibrous histiocytoma of the maxilla. Oral Surg Oral Med Oral Pathol 1989;68:444-50.
4. Heo MS, Cho HJ, Kwon KJ, Lee SS, Choi SC. Benign fibrous histiocytoma in the mandible. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;97:276-80.
5. Menditti D, Laino L, Mezzogiorno A, Sava S, Bianchi A, Caruso G, *et al.* Oral benign fibrous histiocytoma: Two case reports. Cases J 2009;2:9343.
6. Weiss SW, Enzinger FM. Malignant fibrous histiocytoma: An analysis of 200 cases. Cancer 1978;41:2250-66.
7. Crissman JD, Henson SL. Malignant fibrous histiocytoma of the maxillary sinus. Arch Otolaryngol 1978;104:228-30.
8. Meister P, Höhne N, Konrad E, Eder M. Fibrous histiocytoma: An analysis of the storiform pattern. Virchows Arch A Pathol Anat Histol 1979;383:31-41.
9. Meister P, Konrad E, Höhne N. Incidence and histological structure of the storiform pattern in benign and malignant fibrous histiocytomas. Virchows Arch A Pathol Anat Histol 1981;393:93-101.
10. Fletcher CD, WHO classification tumors of soft tissue and bone. 4th ed. Lyon: International agency for research on cancer; 2013.
11. Besly W, Wiesenfeld D, Kleid S, Allan P, Poker I. Malignant fibrous histiocytoma of the maxilla – A report of two cases. Br J Oral Maxillofac Surg 1993;31:45-8.
12. Shahoon H, Esmaeili M, Nematollahi M. Eight-year follow-up of malignant fibrous histiocytoma (undifferentiated high-grade pleomorphic sarcoma) of the maxilla: Case report and review of the literature. J Dent Res Dent Clin Dent Prospects 2009;3:32-5.
13. Kasat VO, Saluja H, Rudagi BM, Kalburge JV, Sachdeva S. Malignant fibrous histiocytoma of maxillary alveolar ridge extending to the hard palate. J Cancer Res Ther 2014;10:422-4.
14. Saluja H, Kasat VO, Rudagi BM, Dehane V, Kalburge JV, Nikam A. Benign fibrous histiocytoma of the maxilla: A case report and review of literature. Indian J Dent Res 2014;25:115-8.