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. These lesions most often arise on the skin but may rarely occur in deep tissues. The involvement of the paranasal sinuses is very rare.

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

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.

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.

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.

CASE REPORT

A 45-year-old man presented with a 6-month history of nasal obstruction, epistaxis, 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.

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...
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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. The etiology of oral BFH is obscure. Chronic irritation, continuous trauma, and spontaneous development have been reported for those located within the oral cavity. According to the WHO, BFH is rare, with <100 reported cases. Patients have ranged in the age from 6 to 74 years at diagnosis.

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...
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.[10] 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
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.

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Conflicts of interest
There are no conflicts of interest.