The clinical challenges and dilemma in the management of uncommon maxillary sinus tumors – A report of two cases

Siddhanth Jain¹, Shivakumar Thiagarajan¹, Poonam Panjwani², Pranav Sathe¹, Mukta Ramadwar²

Departments of ¹Head and Neck Surgical Oncology and ²Pathology, Tata Memorial Center, Homi Bhabha National Institute, Mumbai, Maharashtra, India

Abstract Maxillary sinus is the common site for the nose and paranasal sinus tumors with diverse histopathological types and the treatment for each may differ. Making a histopathological diagnosis on occasion can be challenging. We had two patients presenting with upper alveolus growth in whom establishing the histopathological diagnosis was challenging. Through clinical evaluation, imaging (computed tomography and/or magnetic resonance imaging) and identification of key histopathological features helped in the management of these patients.

Keywords: Giant cell tumor, management, maxillary sinus, myofibroblastic tumor, tumor

Address for correspondence: Dr. Shivakumar Thiagarajan, Department of Head and Neck Surgical Oncology, Tata Memorial Center, Homi Bhabha National Institute, Mumbai - 400 012, Maharashtra, India.

E-mail: shivakumarthiagarajan@gmail.com

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INTRODUCTION

The maxillary sinus is the most common site of the nasal cavity and paranasal sinus tumors.^[1] There are a plethora of histopathological types arising from this site which include both benign and malignant.^[1] It is very important to establish the histopathological diagnosis for these tumors as this could dictate their further management.

CASE REPORT

We had two patients presenting to us with upper alveolus growth of a few months' duration following a dental procedure. Both patients were evaluated with imaging. In Case 1 [Figure 1a], magnetic resonance imaging (MRI) revealed an ill-defined heterogeneously enhancing soft-tissue lesion with central scarring and calcifications involving the maxillary sinus, retro-maxillary space and infratemporal fossa

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suggestive of reparative giant cell granuloma [Figure 1b]. Biopsy was suggestive of giant cell rich lesion, with no evidence of malignancy. Brown's tumor was one of the differential diagnoses which were ruled out after appropriate investigations. A decision to perform an excisional biopsy was taken in the tumor board; however, the procedure could not be completed due to profuse bleeding during the procedure for which angioembolization was necessitated. The report of the biopsy done during this procedure did not add to the diagnosis. Hence, a complete surgical excision of the tumor with appropriate reconstruction was done. Histopathologically, it was a giant cell-rich lesion with evenly distributed multinucleated osteoclasts. The stroma showed areas of hemorrhage and fibroblastic proliferation. No stromal atypia or abnormal mitotic figures were seen [Figure 1c]. The overall features were favoring a giant cell tumor (GCT) [Figure 1d].

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Figure 1: (a) Clinical image of right upper alveolus growth in case.1, (b) MRI of case.1 showing the extensive involvement of the right maxilla, (c) Microphotograph of the tumor with hematoxylin and eosin stains at 400× showing evenly distributed multi nucleated osteoclasts, (d) Microphotograph of the tumor with hematoxylin and eosin stains at 200x showing areas of hemorrhage and fibroblastic proliferation

In case. 2 [Figure 2a] MRI showed an ill-defined mass in the left maxillary alveolar region [Figure 2b]. The maxilla appeared eroded on imaging and the biopsy was suggestive of spindle cell lesion (low grade) with myofibroblastic proliferation. A decision to perform surgery was taken in the tumor board, and an infrastructure maxillectomy with reconstruction was performed. Microscopically, it was moderately cellular spindle cell tumor, composed of stellate/spindle cells situated in a myxoid stroma [Figure 2d]. There was no necrosis with very occasional mitotic activity and no atypical mitoses. In immunohistochemistry, the cells showed focal expression for desmin [Figure 2c]. The overall features were consistent with a low-grade myofibroblastic tumor, FNCLCC grade 1.

DISCUSSION

Some of the differential diagnoses for GCT are giant cell granuloma, Brown's tumor, aneurysmal bone cyst, phosphaturic mesenchymal tumor and tenosynovial GCT (TSGCT).

A giant cell granuloma shows unevenly distributed giant cells with areas of hemosiderin deposition and reactive bone.^[2-5] The giant cells are usually seen arranged around the hemorrhagic areas. It is a benign process, and the common sites are the jaw and craniofacial bones. These tumors are usually unencapsulated.

Brown's tumor of hyperparathyroidism shows multiple bony lesions with similar histologic features such as giant cell granuloma.^[6] It is essential to perform serum PTH and serum alkaline phosphatase in these patients.



Figure 2: (a) Clinical image of the left upper alveolus growth in case.2, (b) MRI picture of case.2 showing the upper alveolus growth with bone erosion, (c) Microphotograph of the tumor with the cells showing focal expression for Desmin on IHC, (d) Microphotograph of the tumor with hematoxylin and eosin stain at 400× showing stellate/spindle cells situated in a myxoid stroma

An aneurysmal bone cyst is also known to occur in the facial bone and shows air-fluid levels on radiology. Histological features are similar to giant reparative granuloma.^[6]

Phosphaturic mesenchymal tumor has a typical clinical association with rickets and osteomalacia.^[6] These tumors also show osteoclastic giant cells and a characteristic grungy type of calcification, areas of hemorrhage and ossification. These are usually benign and associated with low levels of serum phosphate and Vitamin D.

TSGCT, also considered to be a fibro-histiocytic tumor is known to affect the temporomandibular joint.^[6,7] Histologic features include osteoclastic giant cells mixed with macrophages, and histiocytic cells containing hemosiderin pigment.

The histopathological diagnosis in our second case was low-grade myofibroblastic tumor. Myofibroblastic tumors are usually seen as deep intramuscular tumors in the extremities and heads-and-neck region. These are tumors having myofibroblastic differentiation, with at least one marker of myogenic differentiation positive.^[8] These may mimic other benign fibroblastic proliferation like fibromatosis and nodular fasciitis.^[8] There were no features suggestive of a high grade sarcoma.

The histologic features among the various nasal cavity and peripheral nervous system tumors may overlap. A thorough clinical evaluation along with appropriate imaging, which includes computed tomography and/or MRI are essential. The correlation of the clinical, imaging findings and key histologic findings are important to arrive at a definite diagnosis or at least have a few.differential diagnoses based on which treatment for these patients can be planned.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/ have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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