Giant odontogenic fibroma of maxilla

Brajendra Baser, Arvind Kinger, Geeti V. Mitra, Manya Thakur Roy
Departments of ENT and Oral and Maxillofacial Surgery, SAMC and PG Institute, Indore, Madhya Pradesh, India

Address for correspondence:
Dr. Brajendra Baser, Department of ENT, SAMC and PG Institute, Indore, Madhya Pradesh, India.
E-mail: baserbv@gmail.com

ABSTRACT

Odontogenic fibroma is a benign ectomesenchymal tumor classified as central and peripheral on the basis of its location and as an epithelium rich or epithelium poor based on its histological features. Radiological findings consist of radiolucent areas with well-defined bony margins. The lesion is detected early because of its location and usually treated with surgical excision and curettage. We present a case of giant odontogenic fibroma of right maxilla presenting as gross facial deformity and posing a dual challenge of excising the tumor mass and reconstructing the ensuing defect.

Keywords: Giant, maxilla, odontogenic fibroma, reconstruction

INTRODUCTION

Odontogenic fibroma is a benign mesodermal tumor originating from the dental follicle, periodontal ligament or dental papilla.[1] WHO has defined this lesion as a rare neoplasm consisting of collagenous fibrous tissue with varying amount of odontogenic epithelium.[2] It is classified on the basis of its location as central or intraosseous and peripheral or extraosseous.[3] The lesion occurs most commonly in mandible although cases have been reported in maxilla as well. The lesion involves maxilla in its anterior region and mandible in its posterior region.[4] The age of afflicted persons ranges from 4 to 80 years with a female predilection. The lesion grows slowly and becomes symptomatic only when it reaches a considerable size. Initial clinical presentation can be a slight expansion of the buccal or lingual cortical plate. Very large lesions are present with external facial deformity.

Radiological features in smaller lesions may show unilocular radiolucent area with well-defined borders. Larger lesions may show multifoculation with scalloped margins and may cause cortical expansion or tooth loosening or displacement.[5] Depending on the amount of epithelium seen histologically, the histological classification of odontogenic fibroma as epithelium poor (simple type) and epithelium rich (complex or WHO type).[2] The treatment includes surgical excision and curettage. The recurrence rate is very low as reported in the literature. We present a case of giant odontogenic fibroma presenting as gross facial deformity with upward and outward displacement of orbit and downward and medial displacement of palate. Such a large tumor was a dual challenge in excising the tumor mass and in reconstruction of the surgical defect.

CASE REPORT

A 20-year-old male patient reported to the Department of ENT at SAIMS Medical College with the chief complaint of painless swelling in the right maxillary region, which was slowly growing for the past 3 years causing marked facial disfigurement with palatal and orbital displacement [Figure 1]. There was gaping between teeth of right upper alveolus. Externally, the swelling was approximately 10 cm × 13 cm in size, firm, nontender, and nonfluctuant in nature. It was superiorly encroaching on the right orbit and displacing it in a superolateral direction. Posteroinferiorly, it was going up to the angle of mandible. Anteroinferiorly, it displaced the upper alveolus and palate inferiorly. Laterally, it extended up to preauricular region. Medial extension of the tumor pushed the lateral wall and septum, blocking the nasal cavities. The nose was deviated to the opposite side, and the mouth opening was normal but patient had slight difficulty in mastication and speech. The patient also had diplopia because of superolateral deviation of the orbit.

A computed tomography (CT)-scan revealed a well-circumscribed...
solid-cystic mass of size 11.6 T × 11.4 AP × 12.9 CC cm [Figure 2]. There was marked thinning of the sinus wall and superolateral displacement of right eyeball with no obvious infiltration of intra-/extra-conal fat. Posteriorly, it occupied infratemporal fossa with obliteration of pterygomaxillary fissure, pterygopalatine fossa, vidian canal, and inferior orbital fissure. It was inseparable from pterygoid plates and reached up to the nasopharynx. It was just touching the floor of middle cranial fossa with no skull base erosion. Inferiorly, it involved the alveolar process with gross mal-alignment and displacement of teeth. Medially, it involved the right ethmoidal cells, obliterating the frontoethmoidal recess with gross deviation of lateral nasal wall and septum. The bony and soft tissue window of CT-scan showed an expansile, well-circumscribed, multilocular, solid-cystic mass with centric, and eccentric foci of mineralization with marked bony remodeling causing thinning and expansion of the bony cortex [Figures 3 and 4]. These findings suggested probable diagnosis of either a giant cell tumor or giant cell reparative granuloma. Fine-needle aspiration cytology smear showed a cluster of spindle cells in a myxoid background suggesting fibromyxoid tumor.

The large tumor was excised completely using Weber-Fergusson incision [Figures 5 and 6]. It was a multilocular swelling with well-defined bony margins [Figure 7]. The bony wall of orbital floor, the lower margin of alveolus and floor of maxillary sinus were intact. The lateral nasal wall defect on the right side was closed using the available mucosa. Horizontal plate of expanded palate was osteotomized parasagittally to elevate the palate. More osteotomies were done all around the alveolus to reposition it vertically forming part of medial maxilla. The alveolus thus formed was stabilized with 3 cm × 7 cm titanium mesh superiorly to the nasal bone and zygomatic arch and inferiorly to the new formed lower margin.

The mass was sent for histopathology. Grossly it was a solid cystic mass measuring 11 cm × 11 cm × 13 cm [Figure 7]. The cut surface showed multiple cysts of varying sizes, filled with gelatinous material. The sections showed a tumor composed of fibrous tissue with cystic spaces containing dentine like material with scattered areas of calcification, suggesting the diagnosis of odontogenic fibroma [Figure 8].
Once the tension from the removal of the tumor mass was released, the orbit slowly moved to a medial position. Six months later when the eye was in a more neutral position, the patient was taken for another surgery. Titanium mesh was removed, and zygoma implant (Biopore) was fixed. Two years later when the remodeling of palate took place, the patient was fitted with a partial denture. Three years follow-up of the patient showed no sign of recurrence of disease. The patient has a cosmetically acceptable face with normal mastication and binocular vision [Figure 9].

**DISCUSSION**

Odontogenic fibroma of maxilla is a rare tumor. Wesley et al. described odontogenic fibroma as a slow progressive, painless cortical expansion of the jaw, which is seen as multilocular radiolucent lesion with or without unerupted/displaced teeth in radiography and dense collagen with fibroblasts and odontogenic epithelium histopathologically.[1] Most of the studies in literature observed that the lesions were smaller with higher prevalence in females in the second to fourth decade of life and mandible was involved more than maxilla.[6,7] Our case is a male patient of 20 years with giant lesion involving right maxilla. Earlier cases of odontogenic fibroma in the maxilla, although rare, have been reported but such giant odontogenic fibroma has not been reported as per our search in the available literature.

Radiographic studies demonstrate a soft tissue mass with some areas of calcification and do not involve underlying bone.[8] The CT-scan in our case showed a solid-cystic mass with marked bony...
thinning of sinus as well as adjacent bony walls. It was labeled as a benign swelling with marked displacement of adjoining structures.

The challenge was complete excision as well as repair of the defect. Weber-Ferguson approach was taken for complete removal, and immediate reconstruction was done using titanium mesh. The recurrence of this tumor is uncommon with no evidence of malignant transformation.

Odontogenic fibroma is histologically characterized by fibroblastic proliferation with scattered amount of calcified material and a variable amount of odontogenic epithelium. Similar histological features have been observed in other lesions like hyperplastic dental follicles. The histological features in our case were consistent with findings of odontogenic fibroma. The patient is followed for 3 years and no signs of recurrence were seen.

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