Fibromyxoma of the Maxilla – A Case Report

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Abstract
A fibromyxoma is a rare odontogenic mesenchymal tumor of the jaws. It is benign and painless but locally destructive. The mandible is more commonly involved than the maxilla. Its histological and radiological features make a fibromyxoma difficult to differentiate from other odontogenic tumors and it may occasionally be misinterpreted as a malignant lesion. In this paper, we present an aggressive case of a fibromyxoma of the maxilla with radiological consideration, postoperative follow-up, and a brief review of the literature.

Keywords:
fibromyxoma, odontogenic tumor, maxilla, spindle cells

Introduction
A fibromyxoma is an uncommon lesion, accounting for only 1% to 3% of all cysts and tumors of the jaws. Fibromyxomas are benign but aggressive mesenchymal tumors that are slow-growing, expansive, and locally destructive (1). They occur commonly in women with a mean age of 31 years. We report a case of a painful swelling in the left maxillary posterior region of one-month duration. A provisional diagnosis of ameloblastoma was made due to the aggressive nature of the lesion as well as ameloblastoma being a common odontogenic tumor; a malignant tumor was also suspected because of the lesion’s rapid onset. Radiographs showed diffuse radiopaque and radiolucent areas with ill-defined borders in the left maxillary posterior region with displacement of the maxillary left third molar into the infratemporal fossa.

Case Report
A 23-year-old man reported to the Department of Oral Medicine and Radiology with painful swelling in the upper left back teeth region of one-month duration. Pain was pricking, continuous, and radiating toward the left ear and neck region. Swelling was small (5 mm) initially and enlarged to attain the present size (4 cm). No history of epistaxis, difficulty in breathing, anesthesia, or paresthesia was reported. Right and left submandibular lymph nodes were palpable, tender, soft to firm in consistency, and mobile. No relevant medical, surgical, or family history information was reported. A complete physical examination did not contribute useful information.

Examination of the Lesion
Extraoral: A diffuse swelling in the left middle third of the face was seen. The swelling was approximately 4×2 cm and roughly oval in shape, with ill-defined borders and a smooth surface. It extended anterio-posteriorly from 2 cm short of the ala of the nose to 1 cm short of the tragus of the ear and superio-inferiorly from the inferior orbital margin to 3 cm short of the lower border of the mandible. On palpation, a local increase in temperature and tenderness were found; all visual findings were confirmed, the swelling was soft to firm in consistency, and motor and sensory nerve functions were intact (Fig. 1).

Intraoral: Diffuse swelling was seen in relation to teeth #25–28, extending from the distal aspect of tooth #25 to the mesial aspect of tooth #28 on both the buccal and palatal aspects, completely obliterating the buccal sulcus; on the palatal aspect, swelling was 2 cm short of the midline of the palate. The swelling was approximately 3×2 cm on either side of the maxillary left posterior quadrant, oval, and covered with diffuse erythematous areas of mucosa. The anterior border was well-defined, extending from the
region and carcinoma of the alveolus was considered because of the swelling’s rapid onset. Other differential diagnoses such as central giant cell granuloma, central giant cell lesions of hyperparathyroidism, aneurysmal bone cyst, metastatic tumor of the jaw, and sarcoma were also considered.

**Investigations**

A vitality test (heat) performed on teeth #25 and #26 was positive. A routine blood picture was normal. Intra-oral periapical radiographs of the left upper posterior quadrant showed ill-defined radiolucency of the alveolar bone, altered trabecular pattern, and missing tooth #27; resorption of the mesiobuccal and distobuccal roots of tooth #26 was present (Fig. 3). An orthopantomograph showed diffuse radiolucency of the maxillary left posterior quadrant. Clinically, we thought tooth #27 was missing, but the orthopantomograph showed that tooth #27 was distally displaced and tooth #28 was seen in front of the infratemporal fossa (Fig. 4). PNS (Para–Nasal–Sinus) view showed radiopacity involving the left maxillary sinus. A lateral skull radiograph showed displaced tooth #28 in front of the infratemporal fossa (Fig. 5).

A computed tomography scan of the maxilla and paranasal sinus showed an expansile soft tissue density lesion of 64 HU enhancing to 80 HU, in which there was expansion of the buccal cortical plate of distal aspect of tooth #25; the posterior border was ill-defined. Teeth #27 and #28 were clinically missing and tooth #26 was slightly extruded from the socket. The swelling was tender, soft to firm in consistency, all visual findings were confirmed, and tooth #26 exhibited grade III mobility (Fig. 2).

A provisional diagnosis of ameloblastoma was given due to the lesion’s aggressive nature and ameloblastoma being a common odontogenic tumor of the jaw. A differential diagnosis of dentigerous cyst was given due to missing teeth in the same
the maxilla involving the left maxillary sinus with expansion of the sinus wall and a thinned cortical border. The posterior-lateral wall of the left maxillary sinus showed a cortical break with minimum extension. No intraorbital or nasal cavity extension was observed. Thickening of the right maxillary sinus mucosal wall was seen. All of these features were suggestive of an aggressive lesion (Fig. 6a & 6b).

Fine needle aspiration of the lesion was non-diagnostic. Incisional biopsy was performed and the histopathology features showed loosely arranged stellate, spindle-shaped and round cells in an abundant loose myxoid stroma with collagen fibers; these features confirmed the lesion to be a fibromyxoma (Fig. 7).
missing teeth (2, 3). There is controversy regarding the origin of the tumor. Some believe it to be of odontogenic ectomesenchymal origin, because it bears a resemblance to the stellate reticulum, is usually associated with missing or unerupted teeth, and it has the occasional presence of odontogenic epithelium (2). Others believe it to be an odontogenic fibroma that has undergone myxomatous changes, suggesting that the connective tissue cells are actively secreting an abundance of intercellular myxoid substance containing hyaluronic acid, chondroitin sulphate, and varying amounts of collagen fibers (2, 4). Goldblatt (5) did an ultrastructural study of myxomas and concluded that myxoma cells show many characteristics of fibroblasts of the odontogenic apparatus; thus, tumor origin from non-odontogenic mesenchyme cannot be ruled out by existing ultrastructural studies.

Abiose et al. (2) reported that fibromyxomas constituted 3.73% of all benign and malignant oral tumors and 20% of tumors of dental origin, second in incidence to ameloblastoma. Most cases arise in the second and third decades of life. However, James et al. (1) reported the case of a maxillary myxoma in a child of 11 months. In their retrospective study, Keszler et al. (10) found that myxoma in childhood represented 12.5% of the 80 cases of this tumor and stated that fibromyxoma must be taken into account in the differential diagnosis of intraosseous radiolucencies in young patients.

In their review of fibromyxomas, Farman et al. (6) stated that three mandibular cases were found for every two maxillary cases and that fibromyxomas were more common in the premolar and molar regions. Cases have been reported of fibromyxomas in the ascending ramus and condyle without extension to the dental regions; this makes sense give that in early development, odontogenic tissues exist in close proximity to the condyle. Adekeye et al. (7) reported on 18 cases of myxoma, with 11 in the mandible and 7 in the maxilla; 4 of the mandibular tumors and 2 of the maxillary ones were anterior and crossed the midline. Abiose et al. (2) reviewed 10 cases of fibromyxomas, with 6 in the mandible and 4

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**Fig. 8.** Postoperative photograph

**Fig. 9.** Postoperative photograph showing the obturator

**Treatment**

A course of analgesics and antibiotics was given to reduce the pain and likelihood of superadded infection. The patient was treated with subtotal maxillectomy, left coronoidectomy to prevent fibrosis, and orbital floor reconstruction (Fig. 8). After 1 month, an obturator was placed to cover the defect. After 6 months of follow-up, the patient was comfortable but he reported loosening of the obturator with no recurrence of the lesion (Fig. 9).

**Discussion**

A fibromyxoma is a relatively rare benign neoplasm occurring in the jaws, which seems to occur more frequently in the second and third decades of life. However, some literature refers to a myxoma as very common in the mandible and associated with
in the maxilla; these were more commonly situated in the posterior region. Keszler et al. (10) analyzed 10 cases of myxoma in children and noted that most of the lesions were located in the premolar–molar region, predominantly on the right side.

Most myxomas are asymptomatic, although some patients present with progressive pain in lesions involving the maxilla and maxillary sinuses, with eventual neurologic disturbances. Fibromyxoma of the mandible tends to produce extensive bone destruction, invasion into the surrounding structures, and a relatively high recurrence rate; however, metastasis is rare. Fibromyxoma of the maxilla is less frequent but behaves more aggressively than that of the mandible as it spreads through the maxillary sinus, as presented in our case (2–4).

Adekeye et al. (7) in their study of 18 cases of myxomas described this tumor as slowly growing, resulting in gross expansion of the bone, loosening and displacement of teeth, interference with speech and mastication, and facial deformity without significant pain. Keszler et al. (10) in their analysis of 10 cases of myxoma in children found deformation, moderate pain, tooth loss, and malocclusion. Cohen et al. (9) reported a case of myxofibroma of the maxilla that was not painful and had not caused any discomfort.

Allen (8) reported a case with radiographic consideration, in which orthopantomographs disclosed a large honeycombed radiolucent area extending from the mandibular left premolar to the left coronoid and condylar processes. Adekeye et al. (7) in their 18 cases of myxoma showed two different types of radiographic appearance: one radiographic type showed a radiolucent image with diffuse margins and a soap bubble or honeycomb appearance; the other radiographic type showed a well-defined radiolucency with a faint sclerotic margin.

Cohen et al. (9) reported a case with computed tomography findings that revealed a large, honeycombed, expanding lesion involving the entire maxillary sinus. Abiose et al. (2) in their 10 cases of fibromyxomas noted multilocular or honeycombed radiolucencies with varying degrees of root resorption of the involved teeth. Keszler et al. (10) reported that the tumor usually appears as a unilocular or multilocular radiolucency with multiple interlaced and interrupted osseous trabeculae in the central region.

Allen (8) reported the histological study of a fibromyxoma that was composed of loosely arranged and variable numbers of round or stellate cells with ovoid and hyperchromatic nuclei, with odontogenic epithelium and mature collagen fibers found in the intercellular matrix. Gandra et al. (11) reported the microscopic study of a myxoma that was composed of mesenchymal cells, an extremely loose stroma, amorphous ground substances with a mucinous appearances, and some elongated, weakly stained cells. Takahashi et al. (12) performed an immunohistochemical investigation of odontogenic myxomas using a panel of polyclonal and monoclonal antibodies and confirmed that an odontogenic myxoma has a dual fibroblastic histiocytic origin.

Adamo et al. (3) reported a case of myxoma of the mandible that was treated by peripheral ostectomy and immediate reconstruction. Adekeye et al. (7) and Abiose et al. (2) treated their cases with enucleation or surgical resections with maxillectomy or mandibulectomy.

**Conclusion**

A case of aggressive fibromyxoma involving the entire maxillary left posterior quadrant is presented with clinical, radiological, and histological findings. It was a diagnostic challenge to examiners because it mimicked other lesions of a similar nature at different steps. It is necessary to include fibromyxoma in the differential diagnosis of tumors involving the jaws that are associated with unerupted or missing teeth.

**References**


