International Journal of Oral & Maxillofacial Pathology. 2012;3(4):60-63
Available online at http://www.journalgateway.com or www.ijomp.org
ISSN 2231 – 2250

Case Report

An Unusual Case of Odontogenic Fibromyxoma of Anterior Maxilla

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Abstract

Odontogenic fibromyxoma is a rare locally invasive, benign neoplasm found in the jaws. It commonly occurs in the second and third decade, and mandible is involved more commonly than the maxilla. The lesion often grows without symptoms and presents as a swelling. As the radiographic features are variable diagnosis is difficult in most cases. It poses a diagnostic and therapeutic challenge due to its unusual morphological and biological behavior. This article aims to elucidate rare case Odontogenic fibromyxoma of maxilla in a 27 years old female patient and also an attempt has been made to discuss this relatively rare entity in light of current information from the literature.

Keywords: Myxoma; Myxofibroma; Fibroma; Fibrous Dysplasia; Desmoplastic; Dental follicle.

Mounesh kumar CD, Suresh KV, Pramod RC, Rajendra Baad, Anand SR, Raghavendra M. An Unusual Case of Odontogenic Fibromyxoma of Anterior Maxilla. International Journal of Oral & Maxillofacial Pathology; 2012:3(4):60-63.

Received on: 16/10/2012 Accepted on: 26/02/2013

Introduction

Fibromyxomas of head and neck are rare non capsulated benign neoplasms. The WHO defines the myxomas as a locally invasive neoplasm consisting of rounded and angular cells in the mucoid stroma. The term "odontogenic myxoma"(OM) is often applied when the tumor occurs in the jaws to reflect its odontogenic origin.[1] OM are considered to be the second common odontogenic tumor in many countries, however only 0.5 - 17.7% of them have been reported in Asia, Europe and America. It comprises around 3 - 6% of all odontogenic tumors.[2] The exact pathogenesis of a myxoma is unclear, an origin from the dental follicle seems to be the most reasonable explanation. There are only few cases of OFM of maxillary anterior were recorded in the literature. The aim of this paper is to report a case of fibromyxoma of the maxilla and to discuss its pathogenesis.

Case report

A 27 year old female patient reported to the department, complaining of an asymptomatic unilateral swelling in the right maxillary anterior region since one year. The swelling was initially small and gradually increased to its present size of approximately 5 x 3cm. Extraoral examination revealed a diffuse, bony hard, swelling on the right side of face, obliterating the naso-labial fold. Buccal and palatal cortices were expanded, and no paresthesia was reported. Intraorally swelling was extending from distal part of central incisor to mesial aspect of second molar with obliteration of buccal vestibule. Lateral incisor, canine and second molars were palatally displaced. First premolar was found missing. (Fig 1a) Associated teeth showed Grade II mobility. Skin over the swelling showed a scarring and her right lower eyelid was pushed upwards.

The panoramic radiograph (OPG) was taken which showed an ill-defined radiopaque mass involving right maxillary antrum. Teeth in the affected region showed displacement. First premolar was found horizontally impacted. (Fig 1b) The Computed Tomography image showed an expansile mass in the right maxilla, which completely obliterated the maxillary sinus.

Incisional biopsy was performed, and on histopathological examination, a loose myxoid stroma with numerous stellate to plump spindle shaped cells was seen. Dense diffuse collection of fibrils and fibroblasts and small inconspicuous strands of odontogenic epithelium were also noted. (Fig 1c) Thus a diagnosis of odontogenic fibromyxoma, (OFM) was given. The lesion was surgically excised with en bloc resection of the maxilla and obturator was fabricated to cover the surgical defect. The course has been uneventful after the surgical removal of the tumor. The patient was advised to visit regularly for examination. After 6 months of follow-up, no recurrence was noted.

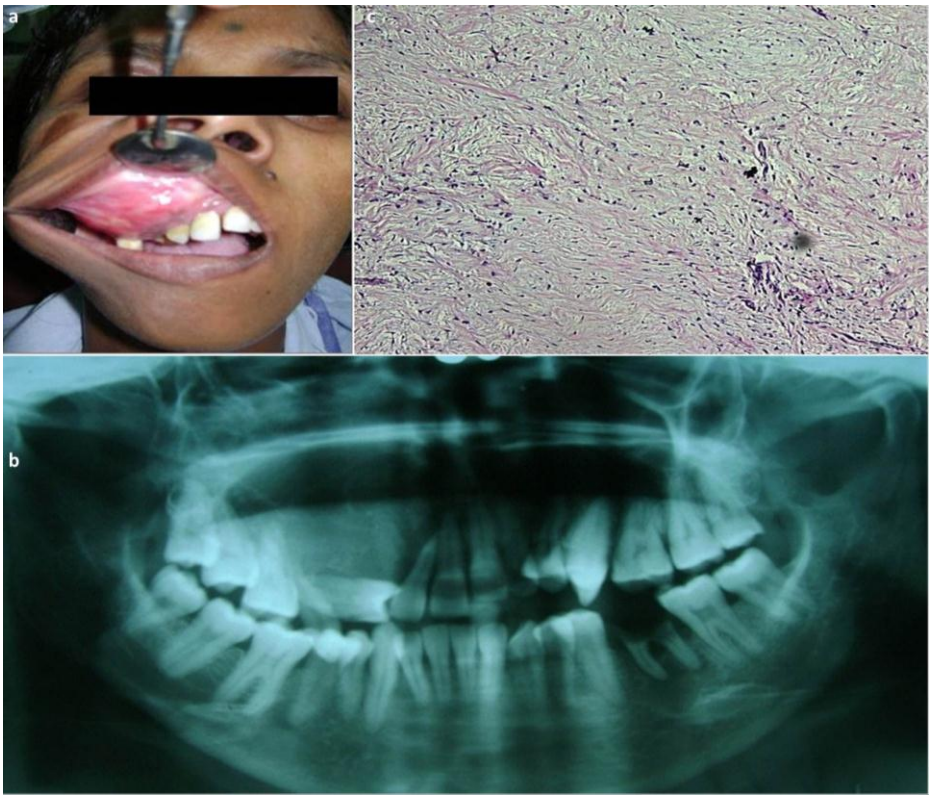

Figure 1: The Clinical photograph showing ill-defined swelling in right anterior maxilla (a) and the orthopantomograph showing ill-defined radiopaque mass (b). The photomicrograph of hematoxylin and eosin stained tissue sections showing loose myxoid stroma with dense collagen bundles and numerous stellate to plump spindle shaped cells in the stroma (c).

Discussion

Fibromyxomas are rare odontogenic neoplasms appear to originate from connective tissues of dental papillae, follicle or periodontal ligament. Fibromyxoma is classified as a specific type of myxoma with a higher fibrous/myxoid tissue ratio than myxoma.[2,3] World Health Organization classifies OM as a benign tumor of ectomesenchymal origin with or without odontogenic epithelium.[4,5,6] According to Dutz and Stout, the term myxoma was first used by Virchow in 1863, but the term fibromyxoma was described by Marcove et al. in 1964.[7,8] In 1947, Thoma and Goldman first described myxomas of the jaws.[9]

Earlier theories suggest that the OFM originates from the neural sheath or the result of degeneration of fibromas, lipomas and so, due to the chronic irritation and the degenerative processes following tissue anoxemia. Recent studies reveal that myxomas/fibromyxomas arise from the mesenchymatous tissue of the dental follicle, thus being described as odontogenic with fibroblasts playing the major role in cell dispersal. This explanation fails to describe soft tissue myxomas. They probably arise from supportive structures of the teeth like the gingiva and the periodontal ligament.[10]

The odontogenic myxoma occurs across an age group that varies from 22.7 to 36.9 years. It is rarely seen in patients younger than 10 years of age or older than 50.[11–13] Our case presented at the age of 27 years, which was in accordance with that reported in literature. The posterior mandible appears to be more frequently affected than the maxilla. The maxilla and anterior region of the mandible are rarely affected. In our case anterior region of maxilla was involved. The majority of myxomas are asymptomatic, although some patients present with progressive pain involving maxilla and maxillary sinus. OM of the maxilla behaves more aggressively than that of the mandible, as it spreads through the maxillary sinus.[12,14] Displacement and mobility of teeth are relatively common. It may be associated with unerupted teeth. Cortical expansion can occur and large lesions can cause perforation.[5,7,13] All these features were in accordance with the present case.

Radiographically, the tumor presents as a unilocular or multilocular radiolucent lesion with well-defined borders with fine, bony trabeculae expressing 'honeycomb,' 'soap bubble,' 'tennis racket', wispy or 'spider web' appearance. Unilocular appearance may be seen more commonly in children and in the anterior part of the jaws.[6,8] Displacement of teeth is a relatively common finding, root resorption is rarely seen.[4,5,7] In the present case OPG showed an ill defined radiopacity with displacement of associated teeth and horizontally impacted first premolar.

The OM exhibits abundant extracellular ground substance and thin fibrils of spindle shaped cells. The undifferentiated mesenchymal cells are capable of fibroblastic differentiation.[7-9] Depending upon the pattern of differentiation, the histological nature of the tumor varies. It may have complete myxomatous tissue or varying proportions of myxomatous and fibrous tissue.[11,12] The differential diagnosis includes ameloblastoma, central haemangioma, fibrous dysplasia, ossifying fibromas, aneurysmal cysts, metastatic neoplasms, well-differentiated liposarcoma, and other rare entities like desmoplastic fibroma.[10]

Radical resection including a margin of 1.5 – 2 cm of healthy bone is the treatment of choice.[2,13] The lack of a capsule and infiltrative growth pattern is responsible for high rate of recurrence when conservative enucleation and curettage are performed. The clinical features of present case suggested an infiltrative tumor of the maxilla. Hence, surgical excision with segmental maxillectomy was performed. Prognosis of myxomas of the jaw is generally good. Recurrence typically occurs during the first 2 years after removal although recurrence has been described over 30 years after original surgery.[14,15]

Myxomas/fibromyxomas show a recurrence rate between 25% and 43%. This is strongly related to the nature of the lesion, presenting without a capsule, thus making the complete removal difficult. The frequency of recurrence of a fibromyxoma of the jaws is higher than that of any other bone thus having a poorer prognosis.[10]

Present case was different in many ways. First of all the location of swelling was unusual and clinical presentation was simulating ossifying fibromas. Secondly, our case was reported in 27 year old female patient and radiographically the epicenter was showing complete radiopacity with horizontally impacted premolar. Although OFM cases has been reported in the previous literature, OFM presenting in maxillary anterior region showing ill defined radiopacity has not been reported elsewhere

Conclusion

Myxomas are less frequent tumors of head and neck region. According to the literature, the maxilla is a uncommon location for fibromyxoma and to best of our knowledge, only 30 cases of a fibromyxoma of the maxilla are reported. OFM poses a diagnostic and therapeutic challenge hence correlation of clinical, radiological and histopathological features are essential when trying to diagnose lesions which lack the characteristic appearance. A complete surgical excision along with proper long term follow up is essential keeping in mind the high recurrence rate, for the successful management of the myxomatous tumor.

Acknowledgement

We would to thank all the staff members of Department of Oral Medicine, Oral Surgery & Oral Pathology for their constant support.

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Source of Support: Nil, Conflict of Interest: None Declared.