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Case Report

Central Ossifying Fibroma of the Maxilla: A Rare Case Report

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Abstract

Maxillofacial fibro-osseous lesions comprise a group of face and jaw disorders characterized by the replacement of normal bone by connective-tissue matrix with varying amounts of mineralized substances. One such lesion is Ossifying Fibroma, which is believed to be derived from the multipotent mesenchymal cells of periodontal ligament origin. Most of the studies showed a female predominance with the most common site being the posterior mandibular region. Here we report a case of central ossifying fibroma occurring in anterior maxilla in a 16 year old male.

Keywords: Anterior Maxilla; Cemento-ossifying Fibroma; Central Ossifying Fibroma; Fibro Osseous Lesions; Jaw Tumors; Mixed Lesions.

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Introduction

Central Ossifying Fibroma (COF) is a rare, destructive benign fibro-osseous neoplasm, characterized by replacement of normal bone by fibrous tissue containing a newly formed mineralized product such as bone or cementum or both.[1,2] It is believed to be derived from the multipotent mesenchymal cells of periodontal ligament.[2,3] Most of the lesions are reported in the mandible in third and fourth decade of life, with female predominance.[4] Herewith a rare case of COF involving anterior maxilla in a male patient is reported.

Case Report

A 16 year old male patient reported to the department of Oral Medicine and Radiology, with a chief complaint of swelling in the upper front tooth region since two years. Patient was asymptomatic two years back, and then noticed a small peanut sized swelling in the upper front tooth region which gradually increased to the present day size and was associated with mild, intermittent pain since one week. Past medical and family histories were non-contributory.

On extraoral examination mild facial asymmetry was observed due to swelling on the left side of the face, measuring about 2×2.5 cm in size, extending anterio-posteriorly, from the left ala of the nose to two centimeter away from it, superio-inferiorly, from the level of left ala of the nose to vermillion border of left upper lip; with normal skin over the swelling, with a smooth surface and no visible pulsations. On palpation, it was tender, firm in consistency, non-compressible, non-reducible (Figure 1a). On intraoral examination, a solitary well-defined swelling in the upper front tooth region measuring about 2×2.5 cm in size and extending anterio-posteriorly from the distal aspect of 22 to mesial aspect of 24 was evident. Surface of the swelling had indentations of the lower teeth with displacement of tooth in relation to 23. On palpation it was hard in consistency, non mobile, non-compressible, non–reducible, fixed to the over lying mucosa and was tender (Figure 1b). Based on the history and clinical findings, a provisional diagnosis of peripheral ossifying fibroma was made with a differential diagnosis of central ossifying fibroma, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor and ameloblastoma.

Routine base-line investigations (complete hemogram) were non-contributory. Radiographical examination of Intraoral periapical radiograph (Figure 1c) revealed a well-defined radiopacity in the region of 23, panoramic view (Figure 1d) and maxillary occlusal view (Figure 1e) revealed a well-defined lesion of mixed radio densities (multiple radiopaque foci) measuring 2×2.5 cm in size extending from the distal aspect to 22 to mesial aspect of 24 beyond the alveolar ridge with ill defined periphery. Labially displaced tooth in relation to 23. Axial and Coronal section (Figure 1f) of computed tomography (CT) showed hyperdense foci in the left anterior maxilla.

After obtaining consent from the patient, complete surgical excision of the lesion was done and the specimen was sent for

histopathological examination, (Figure 2a&b) which revealed hyperplastic pseudo-epitheliomatous stratified squamous epithelium with underlying connective tissue showing bundles of collagen fibers interspersed with loosely arranged fibrous tissue. In addition foci of bony trabeculae were seen without osteoblastic rimming; suggestive of central ossifying fibroma. The overall clinical, radiographically and histopathological features were consistent with the central ossifying fibroma. The post-operative orthopantomogram was taken and patient was under regular follow up since one year, showing no signs of recurrence (Figure 2c).

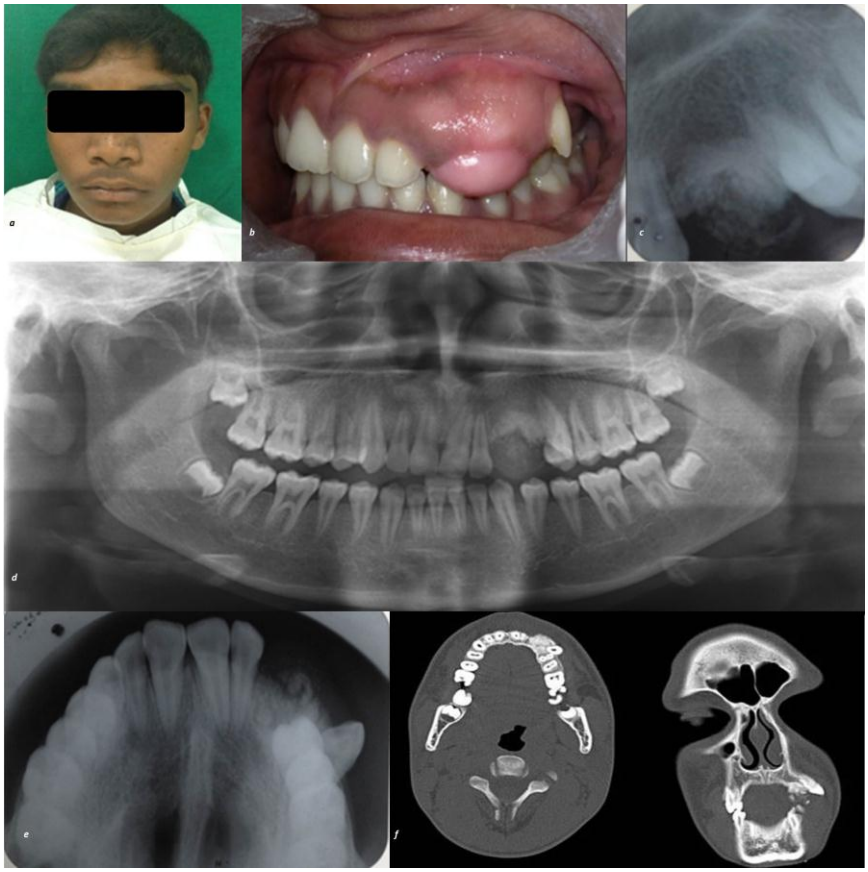

Figure 1: The photograph showing extra oral view (a) and intraoral (b) swelling on left maxillary gingiva. The Intraoral periapical radiograph (c), Orthopantomograph (d) and occlusal (e) radiograph reveals a well-defined mixed radiodensity in relation to 23 region with axial and coronal section of computed tomography (f) showing a hyperdense foci in relation to left maxillary region.

Discussion

According to World Health Organization (WHO) classification in 1992, central ossifying fibroma is a well defined or rarely encapsulated neoplasm consisting of fibrous tissue and varying amount of mineralized material resembling bone or cementum. In 1872, Menzel was the first person, to report ossifying fibroma under the name of cemento-ossifying fibroma, involving mandible in a 35-years old female patient, appointed by Montgomery in 1927.[3-5] Branon and Fowler first used the term 'Ossifying Fibroma' (OF) in place of Cemento Ossifying Fibroma (COF), which was accepted by WHO in 2005 and later replaced the term COF with OF.[6]

Clinically, ossifying fibroma presents as a painless, slow growing mass in the jaw where displacement of teeth may be the only early clinical feature.[2] The lesion is therefore frequently ignored by the patient until the growth produces a noticeable swelling and facial deformity.[7] All the above clinical features were evident in the present case except the patient being symptomatic. OFs are usually seen in the third and fourth decades of life with female predominance in ratio of 5:1.[3,8] The most commonest site of occurrence being the mandible (62% to 89%), in the premolar-molar region (77%),[9] but the case reported here is of a male

patient of second decade occurring in anterior maxilla.

Radiologically, these tumors present with a different pattern depending on their degree of mineralization. Based on the radiographic features MacDonald-Jankowski described OFs into three stages; an initial radiolucent stage, mixed stage and followed by sclerotic stage.[8] There are three different patterns of radiographic borders of cemento-ossifying fibroma which are: defined lesion without sclerotic border (40%); defined lesion with sclerotic border (45%); and lesion with ill-defined border (15%) indicating a rapidly growing tumor.[10] The present case presented as mixed lesion with well defined borders.

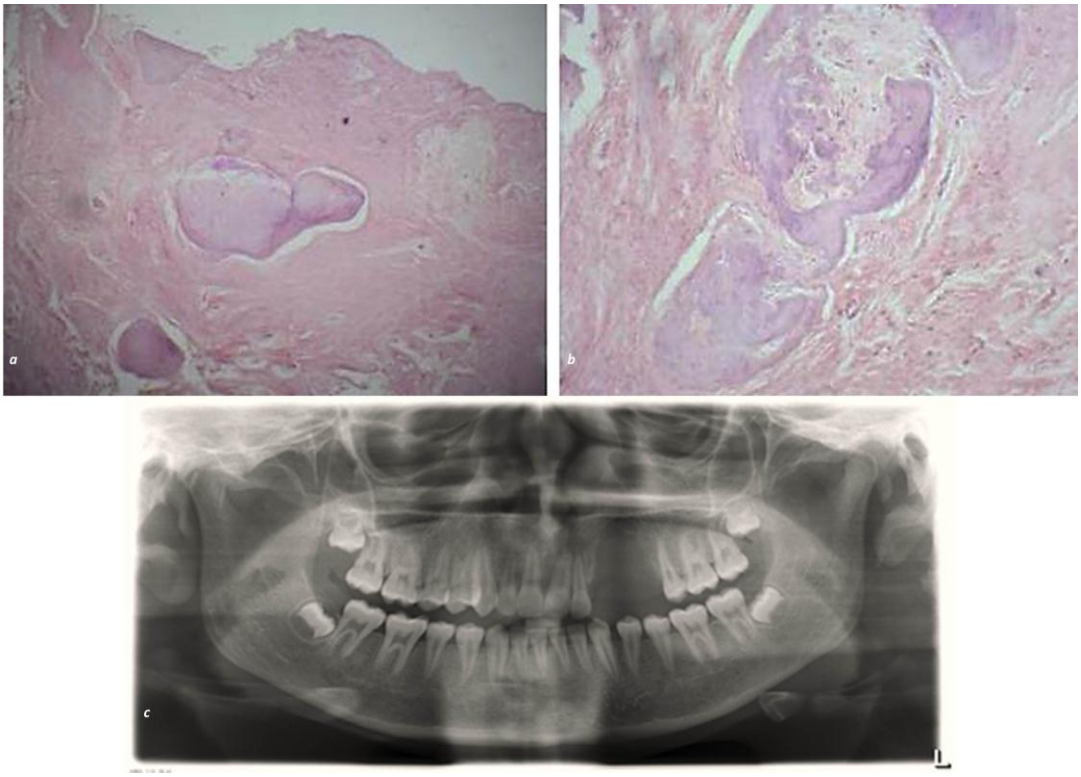

Figure 2: The photomicrograph of hematoxylin and eosin stained section at low (a) & high (b) power view show foci of bony trabeculae without osteoblastic rimming. The Post-operative Orthopantomograph (c) show no signs of recurrence since one year follow up.

The differential diagnosis of COF is based on radiographic features. As a completely radiolucent lesion, COF can be misdiagnosed as a lesion with a radiographically similar appearance, such as focal cemento-osseous dysplasia, odontogenic cyst, periapical granuloma, traumatic bone cyst, unilocular ameloblastoma, and central giant cell granuloma. A mixed radio-opaque and radiolucent lesion show features similar to other fibro-osseous lesions like calcifying odontogenic cyst, adenomatoid odontogenic tumor, or odontogenic fibroma. A fully radiopaque ossifying fibroma may be misdiagnosed as retained root, odontoma, idiopathic osteosclerosis, condensing osteitis, cemento-osseous dysplasia (late stage) or osteoblastoma. The imaging features in the present case were similar to those of most reported cases, showing a circumscribed radiolucent lesion with well-defined margins and intralesional calcification.[1,3,5,9]

Histopathologically, COF shows a well vascularized fibrocellular connective tissue with immature bony trabeculae and cementoid. The present case showed hyperplastic pseudo-epitheliomatous stratified Squamous epithelium with underlying connective tissue showing bundles of collagen fibers interspersed with loosely arranged fibrous tissue. In addition foci of bony trabeculae without osteoblastic rimming were seen. Due to overlapping histologic features, the diagnosis of the individual lesions in the fibro-osseous lesions group poses a difficulty.[2,11]

Treatment of ossifying fibroma generally depends on the size and location of the

individual lesion. Surgical curettage or enucleation with a long term follow-up is the initial treatment of choice for small COFs, whereas radical surgery is indicated for the large lesions. Eversole et al. reported a recurrence rate of 28% following curettage. Hence, a long term follow-up of the patients is recommended.[8,12] In the present case surgical enucleation by curettage was done and one year follow-up revealed normal healing without any sign of recurrence.

Conclusion

Although it is relatively not difficult to establish the diagnosis of central ossifying fibroma from clinical, radiographic, and microscopic features, these tumors may exhibit variation in their neoplastic behavior. It is, therefore, important to take into account the individual tumor behavior when planning a surgical treatment in order to eliminate and avoid tumor recurrence and at the same time improve the patient's cosmetic and functional problems.

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