Ossifying fibroma of maxilla in a male child: Report of a case and review of the literature

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ABSTRACT

Ossifying fibroma is a rare benign fibro-osseous neoplasm of the jaw characterized by substitution of normal bone by fibrous tissues and newly formed calcified products such as bone, cementum or both. It is a well-demarcated lesion that differentiates it from fibrous dysplasia. This case report describes a rare case of ossifying fibroma arising in the maxilla of an 11-year-old child treated with enucleation. The clinical, radiographical, surgical and histological findings are presented. Controversies regarding the terminology and classification along with the differential diagnosis are discussed and a review is provided of the literature on the subject.

Key words: Bone neoplasm, cemento-ossifying fibroma, fibro-osseous lesionmaxilla, ossifying fibroma

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Introduction

According to the 1992 World Health Organization (WHO) classification, [1] an ossifying fibroma is a "demarcated or rarely encapsulated neoplasm consisting of fibrous tissue containing varying amounts of mineralised material resembling bone and/or cementum". Ossifying fibroma is a part of benign fibro-osseous lesions of the jaw that are characterised by replacement of normal bone by fibrous tissue containing a newly formed mineralized product. Other commonly included among the fibro-osseous lesions of the jaw are fibrous dysplasia, focal cemento-osseous dysplasia and florid cemento-osseous dysplasia. [2]

Ossifying fibromas often occur in patients in the



second to fourth decade of life with a definite female predilection and the mandible is involved far more often than the maxilla, especially the premolar and molar region. [3-6] Su *et al.* [6] reported that 52 (70%) of their 75 cases of ossifying fibromas were located in the mandible, with 43% located in the posterior region including the ramus area, followed by 22% in the posterior maxilla. The clinical presentation of ossifying fibroma is usually a round or ovoid, expansive, painless jaw bone mass that may displace the roots of adjacent teeth and sometimes cause root resorption. Early lesions are small and radiolucent. As they mature, they become mixed radiolucent and radiopaque lesion and finally a radiopaque lesion. [4]

It is believed to derive from the multipotential mesenchymal cells of the periodontal ligament which are able to form cementum, bone and fibrous tissue^[5,7-12] However, microscopically identical neoplasms with cementum-like differentiation have also been reported in the orbital, frontal, ethmoid, sphenoid and temporal bones as well as nasopharynx and paranasal sinuses, leaving these prior theories of origin open to question.^[12-15] About 70% of these benign fibro-osseous lesion arise in the head and neck region^[13,14]. Although the precise pathogenesis is still unknown, Wenig *et*

al.^[16] has suggested that trauma-induced stimulation may play a role.

The present paper describes a rare case of ossifying fibroma of the maxilla in an 11-year-old male child, with a review of the literature on the subject.

CASE REPORT

An 11-year-old male child reported to the Department of Pedodontics and Preventive Dentistry, Saraswati Dental College, with the chief complaint of swelling in right back region of upper jaw. His past medical and family histories were non-contributory. The swelling had been first noticed 18 months back which gradually increased to the present size. Initially swelling was asymptomatic but had become painful on closure of jaw due to the contact pressure from lower molars. On extra-oral examination, swelling was evident on right side of the face leading to slight facial asymmetry [Figure 1]. Patient presented with adequate mouth opening and intraoral examination revealed swelling of size approximately 2 × 2 cm distal to 16 with buccal as well as lingual expansion and indentation imprint of lower molar [Figure 2]. Swelling was accompanied by marked buccal displacement of 16 and missing/unerupted 17. The overlying oral mucosa was intact and normal in appearance. Swelling was non-tender and bony hard in consistency with welldefined margin. There were no palpable cervical or submandibular lymph nodes.

Panoramic radiographical examination [Figure 3] revealed a well-defined homogenous radiopaque mass distal to the tooth 16 and coronal to un-erupted tooth 17 with radiolucent margin. There was distal and superior displacement of tooth 17 with its root dilacerations. Tooth follicle of 18 was also present. CT scan [Figure 4 a-c] showed a well-circumscribed and sharply defined lesion consists of a mixture of radiolucency and radiodensity with sclerotic border.

Excisional biopsy of the mass was planned under local anesthesia. After achieving adequate anesthesia, incision was given directly over the alveolar ridge swelling that extended anteriorly in the crevice of maxillary first molar to its mesial aspect and from here a vertical releasing incision was given. A full-thickness mucoperiosteal flap was reflected buccally and palatally. On exploration, the mass was well-demarcated from its surrounding bone and easily separated from the bony bed. The lesion was enucleated and was taken out in toto and sent for histopathological examination. Tooth 17 was also extracted during the procedure because of its extreme buccal malposition [Figure 5] and to



Figure 1: Facial swelling on right side of face leading to slight facial asymmetry



 $\textbf{Figure 2:} \ \textbf{Enlargement of the posterior maxilla caused by the lesion}$



Figure 3: Panoramic radiograph showing a large well-defined homogenous radiopaque mass distal to tooth 16 and coronal to 17 with radiolucent margin

facilitate the complete enucleation of the lesion. The extracted 17 was presented with root dilacerations with its palatal root curved sharply in the palatal direction while mesiobuccal and distobuccal roots curved distally [Figure 6 a and b]. The wound was closed in single layer with 3-0 black silk suture. Healing was uneventful on subsequent postoperative follow-up.



Figure 4: (a) Axial, (b) coronal, (c) saggital CT scan view showing a mixture of radiolucency and radiopacity along with the sclerotic border

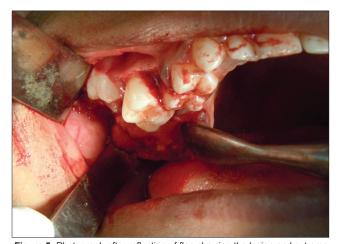


Figure 5: Photograph after reflection of flap showing the lesion and extreme buccal as well as superior displacement of tooth 17

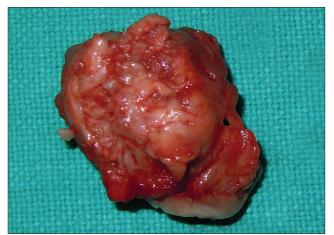


Figure 7: Excised specimen showing a well-circumscribed tumor that shelled out in in one piece

Grossly, the specimen was a well-circumscribed mass measuring about 2.5×2.2×1.6 cm [Figure 7] with areas of hardness. Radiograph of the excised specimen revealed radiolucency in the peripheral region and mixed radiopacity and radiolucency in the central region [Figure 8]. On cutting, the mass

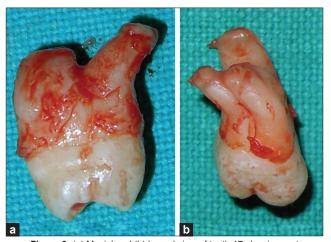


Figure 6: (a) Mesial and (b) buccal view of tooth 17 showing root dilacerations

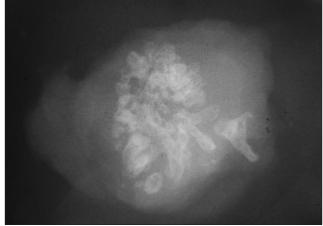


Figure 8: Radiograph of specimen showing mixture of radiopacity and radiolucency in the central region

was hard and gritty in consistency. Microscopical examination of decalcified sections revealed a well-circumscribed fibrous connective tissue mass showing areas of mineralization. The hard tissue was present in the form of trabeculae of mature lamellar bone, immature woven bone and areas of unmineralized

osteoid [Figure 9a]. The connective tissue adjacent to the bone was delicately fibrillar composed of spindle-shaped cells. The trabeculae of woven bone and osteoid showed brush border at the periphery and were lined by plump osteoblasts [Figure 9b]. Focal areas of osteoclastic activity in form of multinucleated osteoclasts and prominent reversal lines were also obvious. No cellular atypia or increased mitosis was seen. Based on the above histopathological features and correlating it with the clinical and radiological findings, a final diagnosis of central ossifying fibroma was made.

DISCUSSION

Yih et al.[17] and Sciubba et al.[5] attributed the first description of this disorder to Menzel, in 1872. Montgomery^[18] appears to have been the first to designate jaw lesions of this type as ossifying fibromas, by which the lesion is currently known. Lack of standardized terminology and classification of central or intraosseous cemento-osseous lesions of the jaws have long posed a dilemma for histopathologists and clinicians. Until 1948 it was believed that fibrous dysplasia and ossifying fibroma were either the same entity or variant of one same lesion.[19] That year, Sherman and Sternberg^[20] published a detailed description of the clinical, radiological and histological characteristics of ossifying fibroma, and since then most researchers coincide in considering the two lesions to be different clinical entities. [3,19-21] Jaffe [22] originally believed these lesions to be monostotic manifestation of fibrous dysplasia, although 5 years later^[23] he considered the ossifying fibroma, which he called fibrocementoma, as a separate entity from fibrous dysplasia.

Various terms have been used to describe these benign fibro-osseous neoplasms. When bone predominates in a lesion, it is called an ossifying fibroma; while the

term cementifying fibroma has been assigned when curved/linear trabeculae or spheroidal (psammomalike) calcifications are encounterd. When both bone and cementum-like material are observed, the lesions are then referred to as cemento-ossifying fibromas.[1] Earlier, many investigators classified cementifying fibromas separately from ossifying fibromas because the former were considered to be of odontogenic origin and the latter to be osteogenic. It is now agreed that both types fall under the same classification as osteogenic neoplasms. On the basis of an analysis of 64 cases classified as ossifying and/or cementifying fibroma, Eversole et al.[3] concluded that a distinction between these two variants would be academic, as no behavioural or histological differences exist. They suggested that nomenclature could be simplified by referring to all lesions in this group as ossifying fibroma.

Since 1968 cementum-containing tumors have been grouped together.[24] The WHO classification of 1971[25] used the unifying concept of cementomas to group together lesions containing cementum-like tissue, thus forming a complex group of lesions with ill defined characteristics in which both neoplastic (benign cementoblastomas, cemento-ossifying fibromas) and non-neoplastic lesions (periapical cemental dysplasias, gigantiform cementomas) formed one group. According to the second WHO classification of 1992[1] benign fibroosseous lesions in the oral and maxillofacial regions were divided in to two categories, osteogenic neoplasm and non-neoplastic bone lesions thus clearly separated neoplastic from non-neoplastic lesions containing cementum-like tissue. Cementifying ossifying fibroma belonged to the former category. During the 2003 Consensus Conference, [26] held in conjunction with the preparation of new WHO volume Tumors of the Head and Neck, some changes were made. Osseous neoplasm and non-neoplastic lesions were categorized under the section "Neoplasms and Other Lesions

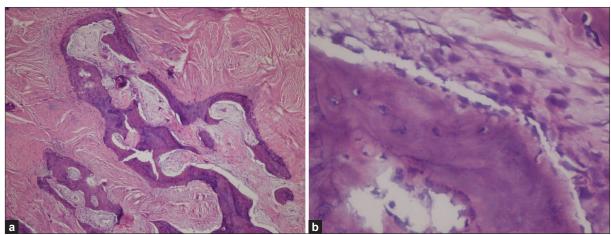


Figure 9: (a) Photomicrograph showing trabeculae of woven bone in a background made up of dense mature collagen fibers (H and E, x4), (b) High-power view showing woven bone rimmed by plump osteoblasts (H and E, x40).

Occurring in the Maxillofacial Skeleton." The section on osseous neoplasms includes ossifying fibroma (formerly cemento-ossifying fibroma) and the section on non-neoplastic lesions comprises fibrous dysplasia, osseous dysplasias, central giant cell lesion/granuloma, cherubism, aneurismal bone cyst and simple bone cyst.

Ossifying fibroma of the jaws generally manifests in second to fourth decades of life^[3,5]; in our case, patient was 11-year old. The juvenile (aggressive) ossifying fibroma (JOF) mainly affects individuals younger than 15 years of age^[1] but behaves in an aggressive fashion when compared to ossifying fibroma. Bone swelling or expansion at the buccal and/or lingual cortical plates is the most frequent clinical sign of ossifying fibroma. [3,4,5,27] They are slow-growing lesions and because of the slow growth, the cortical plates of the bone and the overlying mucosa or skin are invariably intact. They are generally asymptomatic until the growth produces a noticeable swelling and deformity. Small lesions are often discovered incidentally. Ossifying fibromas are usually solitary, but bilateral as well as multiple familial ossifying fibromas have also been reported.[17,28-30] Root divergence, displacement of teeth in the tooth-bearing region or root resorption may be associated with the tumor. [2,30,31] In the present case, maxillary right permanent first molar was erupted but displaced buccally while the maxillary right permanent second molar was still unerupted and was displaced buccally as well as superiorly. The maxillary right permanent second molar was also presented with root dilacerations with its palatal root curved sharply in the palatal direction and mesiobuccal and distobuccal roots curved distally. Large ossifying fibromas of the mandible often demonstrate a characteristic downward bowing of the inferior cortex of the mandible.^[2] The size of the lesion can range from 0.2 to 15 cm^[6]; in our case the excised fibroma measured 2.5×2.2×1.6 cm.

The most important radiographical feature of this lesion is well-circumscribed and sharply defined border. [32] The presence of well-defined margin was held by Sciubba and Younai^[5] to be consistent and reliable radiological marker for ossifying fibroma. Eversole et al[3] examined 64 cases of cemento-ossifying fibroma and found that were all well-defined unilocular, round or oval structures. Larger tumors may have a multilocular radiographical appearance. MacDonald-Jankowski^[33] described three stages in the radiographic appearance. Initially the lesion is radiolucent (osteolytic image), which then becomes progressively radiopaque as the stroma mineralizes thus transforming in to mixed lesion. Eventually, the individual radiopacities coalesce to the extent that the mature lesion may appear sclerotic or radiopaque lesion. He also presented a summary of radiological features in 177 reported cases of cementoossifying fibromas from the literature and his own files^[5,6,21,33] demonstrating that 42% were radiolucent, 24% were radiopaque and 34% had mixed appearance. Three different patterns of radiographical borders were reported by Su *et al*^[6]: A defined lesion without a sclerotic border (40%), a well-defined lesion with a sclerotic border (45%), and a lesion with an ill-defined border (15%).

At surgical exploration, the lesion is well demarcated from the surrounding bone, thus permitting relatively easy separation of tumor from its bony bed. Some ossifying fibromas show, grossly and microscopically, a fibrous capsule surrounding the tumor. Most are not encapsulated but are well demarcated grossly and microscopically from the surrounding bone. Ossifying fibromas consist of fibrous tissue that exhibits varying degrees of cellularity and contains mineralized material. The hard tissue portions consist of trabeculae of osteoid and bone or basophilic and poorly cellular spherules that bear a resemblance to cementum. The bony trabeculae vary in size and often demonstrate a mixture of woven and lamellar patterns. Peripheral osteoid and osteoblastic rimming are usually present. The spherules of cementum-like material often demonstrate peripheral brush borders that blend in to the adjacent connective tissue. Significant intralesional hemorrhage is unusual. Variation in the types of mineralized material produced may be helpful in distinguishing ossifying fibroma from fibrous dysplasia, which has a more uniform pattern of osseous differentiation.[2] It is important to stress again that the ossifying fibroma is a sharply demarcated lesion the hard tissue of tumor do not fuse with the surrounding bone, except occasionally in limited areas.[1] This is a significant feature in distinguishing an ossifying fibroma from a fibrous dysplasia, in which it is common to find that the metaplastic bone of the lesion fuses directly to the bordering cortical bone. [26]

Distinguishing between ossifying fibroma and fibrous dysplasia is the primary diagnostic challenge. Both lesions may exhibit similar clinical, radiographical and microscopic features. The most helpful feature in distinguishing the two is the well-circumscribed radiographical appearance of ossifying fibroma and the ease with which it can be separated from the normal bone. In most cases the well-defined appearance of ossifying fibroma is evident radiographically. Historically, differentiating the two lesions was based primarily on histological criteria. Fibrous dysplasia was reported to contain only woven bone, without evidence of osteoblastic rimming of bone. The presence of more mature lamellae bone was believed to be characteristic of ossifying fibroma. Most authorities now acknowledge that these criteria are unreliable, because both types of bone and cellular features may be found in either lesion. Other differential considerations are osteoblastoma, focal cemento-osseous dysplasia and focal osteomyelitis. Osteoblastoma is evident in slight younger age group and is often characterised by pain. The osseous trabeculae in these lesions are rimmed by abundant plump osteoblasts, but the supporting stroma is loosely fibrovascular with dilated channels and focal hemorrhagic areas. Periapical cemento-osseous dysplasia in posterior teeth may appear radiographically similar and require a biopsy to separate it from ossifying fibroma. Focal osteomyelitis is associated with a source of inflammation and is possibly accompanied by pain and swelling. [32]

Differential diagnosis of ossifying fibroma depends on the radiographical features of the lesion. [27] Ossifying fibroma with a completely radiolucent lesion may be misdiagnosed as cemento-osseous dysplasia (early stage), odontogenic cyst, periapical granuloma, traumatic bone, ameloblastoma or central giant cell granuloma. Differential diagnosis for mixed radiographical feature may include a non-specific diagnosis of fibro-osseous lesion, calcifying odontogenic cyst (Gorlin cyst), adenomatoid odontogenic tumor, rarefying and condensing osteitis, cemento-osseous dysplasia, calcifying epithelial odontogenic tumor (Pindborg tumor), odontogenic fibroma or ameloblastic fibro-odontoma. Furthermore, ossifying fibroma with completely radiopaque radiographical features may be misdiagnosed as retained root, odontoma, idiopathic osteosclerosis, condensing osteitis, cemento-osseous dysplasia (late stage) or osteoblastoma. In addition, ossifying fibroma with a very large size may be misdiagnosed as osteogenic sarcoma.

The term JOF is used for an active growing lesion that mainly affects individuals younger than 15 years of age.[1] This lesion behaves in an aggressive fashion, reaching massive proportions with extensive cortical expansion. Although the patient in our case was just 11-year old, the diagnosis of JOF was ruled out because the lesion did not show any aggressive growth (it was a very slow-growing tumor) and histopathological examination favor the diagnosis of ossifying fibroma. JOF is a welldefined clinical and histological entity that has recently been separated from other central fibro-osseous lesions, including the ossifying fibroma.^[34] It is described in WHO classification^[1] as "an actively growing lesion consisting of a cell-rich fibrous stroma, containing bands of cellular osteoid without osteoblastic rimming together with trabeculae of more typical woven bone. Small foci of giant cells may also be present, and in some parts there may be abundant osteoclasts related to the woven bone. Usually no fibrous capsule can be demonstrated, but like the ossifying fibroma (and unlike fibrous dysplasia), the JOF is well demarcated from the surrounding bone."

Surgical curettage or enucleations are the initial treatment of choice for most small ossifying fibromas.^[26] The circumscribed nature of the ossifying fibroma generally permits enucleation of the tumor with relative ease. Larger lesions that have destroyed considerable bone may necessitate surgical resection and bone grafting. The prognosis is very good, and recurrence after removal of the tumor is rarely encountered. [2,32] However, Eversole et al. in a study of 64 cases of cemento-ossifying fibromas, reported a recurrence rate of as high as 28% following surgical curettage with a mean follow-up period of 38 months. Liu et al. [35] observed that the time of recurrence was always unpredictable, ranging from 6 months to 7 years after the operation. Therefore, there must be a long enough follow-up period of at least 10 years.[35] There is no evidence to suggest that ossifying fibroma ever undergo malignant change. [2] Our patient showed no clinical or radiological evidence of recurrence after 5 months of post-enucleation follow-up. Since the time elapsed from surgery is still short, continued clinical and radiological monitoring is required.

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