Case report: Ossifying fibroma

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Abstract

Ossifying fibroma is a benign fibro-osseous tumor, also known as central fibro-osteoma, cemento-ossifying fibroma, cementing fibroma. This neoplasm arises from the periodontal ligament and shows slow growth and proliferation of cellular fibrous tissue with varying amounts of osseous products such as bone, cementum or a mixture of both. Ossifying fibroma is a localized, asymptomatic swelling with cortical expansion in the mandible rather than the maxilla in mandible in the premolar and molar region in the third to fourth decade of life with female predilection. Radiologically, the lesion shows a well-defined, mixed radiolucent-radiopaque internal structure. Ossifying fibromas can grow to large sizes resulting in aesthetic and functional deformities. Hence, we present a classic case of an 18-year-old male patient with ossifying fibroma on the left body of the mandible, with all clinical, radiological and histopathological features, along with a brief discussion.

Keywords: ossifying fibroma, benign fibro-osseous tumor, central fibro-osteoma, cemento-ossifying fibroma, cementifying fibroma.

Introduction

Ossifying fibroma is a benign fibro-osseous tumor. It is a neoplasm consisting of fibrous tissue containing various amounts of mineralized material. The tumors are thought to arise from the periodontal ligament and are composed of varying
amounts of cementum, bone, fibrous tissue. It is most commonly seen in the mandibular molar, premolar region and occurs with female predilection in the third and fourth decades of life.

**Case Report**

A 18 years old male patient reported to the Department Oral Medicine and Radiology with a chief complaint of swelling in lower left back region of the jaw since 1 yr.

The patient stated that the swelling had been gradually increasing to present size in 1 yr. The past medical, dental family histories were non-contributory. Also, h/o pain, trauma, toothache, fever, cough, weight loss, difficulty in swallowing, restricted mouth opening were non-contributory.

On general examination, he had normal gait and posture and was well oriented, conscious and ectomorphic in built. No evidence of pallor, icterus, cyanosis and clubbing was present.

On extraoral examination, face was symmetrical, no lymphadenopathy was detected.

On intraoral examination, on inspection, [Fig No:1]

A solitary, localized, oval, well defined, reddish pink swelling present on the body of mandible w.r.t 35,36,37 with cortical expansion which was extending anteroposteriorly from distal aspect w.r.t 34 to distal aspect w.r.t. 37on both buccal & lingual aspect and superioinferiorly from 1cm below to occlusal level upto buccal vestibule with no obliteration on buccal aspect and occlusal level upto lingual sulcus with no obliteration on lingual aspect which is approxi. 4x3cm in dimensions, adjacent mucosa is normal.

On palpation, swelling was nontender and hard in consistency. There was no pus discharge, periodontal pocket, mobility, displacement of teeth in the affected area.

Based on clinical findings provisional diagnosis benign lesion was suspected, probably ameloblastoma or fibrous dysplasia was made.

In radiographic evaluation, Intraloral periapical view [Fig No.2] showed mixed radiolucency – radiopacity in the apical portion with 34 to 37.

Mandibular occlusal view [Fig No.3] showed cortical expansion on both buccal and lingual aspect with 34, 35,36.

Orthopantomograph (OPG) [Fig No.4] revealed an irregular, well defined border with mixed radiolucency-radiopacity which was approximately 4 × 3 cm in size extending anteroposteriorly from 34 to 37 region and superiorly from apical portion of 34 and 37 to inferiorly upto the 1cm of inferior border of mandible.
CBCT scan [Fig No.5,6] revealed heterodense lesion with expansion of buccal and lingual cortical plates and with central foci of calcification.

Incisional biopsy was taken and histopathological investigation [Fig No.7] revealed, a well vasularized, fibrocellular tissue with immature bony trabeculae, some areas in the connective tissue shows presence of cementoid and calcification. The inflammatory response is mild.

Thus, the final diagnosis ossifying fibroma w.r.t 34,35,36,37 is made. Lesion was surgically excised, teeth also extracted due to lesion involving bone w.r.t. 34, 35, 36,37. [Fig No.8]

2 months follow-up visit revealed wound healing without signs of recurrence.[FIG NO.9]

Discussion
Fibro-osseous lesions are defined as lesions characterized by normal bone migration but tissue composed of collagen fibers and fibroblasts containing varying amounts of mineralized...
material, which may be bone or cementum like material. [Waldron 1985].

**Synonyms:** Central fibrotheoma, cemento-ossifying fibroma, cementifying fibroma.

**Etiology**
The etiology of ossifying fibromas is not known but traumatic, odontogenic, and developmental origins have been suggested, and periodontal ligaments are thought to be of origin because of their ability to produce cementum and osteoid material.[2] Local trauma-induced stimuli, tooth extraction, periodontal disease [3,4,5,6] and poor bone maturation of congenital origin have also been considered among the possible precipitating factors for the occurrence of ossifying fibroma.[7]

**Mechanism of Disease**
Although the pathogenesis of ossifying fibroma is not specified, several theories are advanced. Ossifying fibroma develops from pluripotent mesenchymal cells capable of producing cementum, bone and fibrous tissue. Two possible origins have been suggested: excessive proliferation of periodontal ligament cells and a metastatic process occurring in the connective tissue fibers (non-periodontal) of the root. The jaw is commonly affected, possibly due to extensive mesenchymal cellular induction in bone and cementum during odontogenesis.[8] Trauma, previous extractions, periodontitis and possible genetic defects are being investigated, particularly mutations of the hyperparathyroidism 2 (HRPT2) gene have been suggested as possible causative agents.[9,10]

These bone tumors consist of highly cellular, fibrous tissue containing varying amounts of abnormal bone. Previously, the acellular amorphous calcified material contained in some lesions was defined as cementum or cementum-like; However, it now appears that this represents an unusual shapeless bone. Therefore, there is no proper rationale for using the term cementum or cementum-like tissue. Other internal parts of this tumor may contain irregular trabeculae of bone. The resulting internal pattern may be very similar or indistinguishable from that of fibrous dysplasia. A characteristic feature that may be present is a soft tissue capsule at the periphery that is not seen in fibrous dysplasia.[11]

**Clinical features:**
Age: Third and fourth decades of life
Sex: A definite female predominance with female to male ratio—5:1
Site: Mandible is the most common site—premolar molar area.
Maxilla— common site— posterior maxilla.
The lesion is generally asymptomatic. The growth produces a noticeable swelling and mild deformity; displacement of teeth may be an early clinical feature. It is a relatively slow growing tumor and may be present for some years. Because of the slow growth, the cortical plates of bone and overlying mucosa or skin are almost invariably intact.[2]

Ossifying fibroma is a benign osseous tumor, most frequent in children and most common in the maxillary sinus and the mandible (75-89%). Sciubba et al.[12] and Yih et al.[13] recognized the first case of ossifying fibroma to Menzel in 1872 as a rare benign primary craniofacial skeleton bone tumor that generally affects the jaws.[8] The term ossifying fibroma was first used in 1927 by Montgomery[14]. The 4th Edition of the World Health Organization Classification of Head and Neck tumors described odontogenic and maxillofacial bone tumors classify OF under fibro and chondro-osseous lesions along with familial gigantiform cementoma, fibrous dysplasia, cemento-osseous dysplasia and osteochondroma.[15]

Three variants of ossifying fibroma are known: One is cemento-ossifying fibroma and another two are form of juvenile ossifying fibroma i.e. juvenile trabecular ossifying fibroma (JTOF) and juvenile psammommatoid ossifying fibroma (JPOF). WHO in 1972 classified it in two types as
ossifying fibroma and cementifying fibromas; but in 1992 WHO considered it in one heading as cement-ossifying fibroma. Further, the term “cementoossifying fibroma” was replaced by “ossifying fibroma” in 2005 under the new WHO classification.[16,17] cemento-ossifying fibroma has an odontogenic origin and is slow-growing with a predilection for the posterior mandible and rarely the maxilla with female predilection[10]. Juvenile ossifying fibroma is a very aggressive form of ossifying fibroma that occurs in the first 2 decades of life. Although the histopathologic definition of this entity is controversial, the radiologic appearance has similarities to that of ossifying fibroma but may be much more expansile.[11] JPOF is rare, with a mean age range of 16-33 years, and occurs mainly in the extragnathic regions of the craniofacial bones with predominance in the orbit and ethmoids.[18] JPOF, more aggressive, presents with a well-delineated periphery with mixed radiopacity and radiolucency. Seventy percent of ossifying fibroma involve the mandible, with 22% found in the molar region of the maxilla, the ethmoids, orbital regions, and rarely the petrous bone.[19]

**Radiological Features**

**Location:** Ossifying fibroma appears almost exclusively in the facial bones and most commonly in the mandible, typically inferior to the premolars and molars and superior to the inferior alveolar canal. In the maxilla, it occurs most often in the canine fossa and zygomatic arch area.[11]

**Periphery:** The borders are usually well defined. A thin, radiolucent line, representing a fibrous capsule, may separate it from surrounding bone.[Fig No.10] Sometimes the bone next to the lesion develops a sclerotic border.[11]

**Internal Structure:** The internal structure is a mixed radiolucent-radiopaque density with a pattern that depends on the amount and form of the manufactured calcified material. In some instances, the internal structure may appear almost totally radiolucent with just a hint of calcified material. In the type that contains mainly abnormal trabeculae of bone, the pattern may be similar to that seen in fibrous dysplasia or a wispy (similar to stretched tufts of cotton) or flocculent (similar to large, heavy snowflakes) pattern may be seen. Lesions that produce more amorphous bone may contain solid, homogeneously radiopaque region that do not have any intrinsic pattern.


**Effects on Surrounding Structures:** Ossifying fibroma can be distinguished from the bone dysplasia by its tumor-like behavior; this is reflected in the growth of the lesion, which tends to be concentric within the medullary part of the bone with outward expansion approximately equal in all directions. This growth can result in displacement of teeth or the inferior alveolar canal and expansion of the outer cortical plates of bone. A significant point is that the outer cortical plate, although displaced and thinned, remains intact. Ossifying fibroma can grow and occupy the entire maxillary sinus expanding its walls.
outward; however, a bony partition always exists between the internal aspect of the remaining sinus and the tumor. [Fig No.11] The lamina dura of involved teeth usually is missing and resorption of teeth may occur.\textsuperscript{[11]}

**Differential Diagnosis**

The differential diagnosis of bone fibroma includes lesions with radiolucent and translucent internal structures. Differentiation from fibrous dysplasia can be very difficult. The borders of bony fibroids are usually more defined, and these lesions may have a soft tissue capsule and cortex, whereas fibrous dysplasias usually blend with the surrounding bone. The internal structure of fibrous dysplastic lesions of the maxilla may be more homogeneous and less variable. Both types of lesions can cause displacement of teeth, but bone fibroids move from a specific point or center. In fibrous dysplasia, teeth are rarely resorbed. Jaw enlargement associated with osteomyelitis is more concentrated around a well-defined central point, whereas in fibrous dysplasia the bone is enlarged but with minimal distortion of overall shape. In other words, the expanded bone still resembles its natural morphology. If the lesion extends to the maxillary antrum, it may be difficult to diagnose bony fibroma and fibrous dysplasia. Fibrous dysplasia typically moves the lateral wall of the maxilla into the maxillary sinus and preserves the contours of the wall, whereas bony fibroma takes on a more convex shape that extends into the maxillary sinus. Fibrous dysplasia can also change the bone around the tooth without displacing the tooth from the apparent center of the concentrically growing benign tumor. The importance of this distinction in treatment is that it is removed in case of ossification of the fibroma, and observation is necessary in the case of fibrous dysplasia. Differential diagnosis of osteofibroma types that produce predominantly homogeneous amorphous bone from periapical osteodystrophy (POD) can be difficult, especially with large solitary lesions of POD. However, POD is often multifocal, whereas osteomyelitis is not. The presence of simple bone cysts is characteristic of ossified osteodystrophy, or POD, whereas bone fibroids exhibit more tumor-like behavior, including tooth movement and concentric enlargement. Wide sclerotic borders are more characteristic of cemento-osseous dysplasia with slow growth and wave expansion. Other lesions to consider include those with internal calcification similar to the pattern seen in bone fibroids. These include giant cell granulomas, calcifying odonto-geic cysts, calcifying epithelial odontogenic tumors (Pienburg), and adenomatoid odontogenic tumors.\textsuperscript{[11]}

![Fig No.11](image)

**Histological Features**

The lesion consists essentially of many interwoven collagen fibers, rarely arranged in discrete bundles, interspersed with numerous proliferating fibroblasts. A small number of mitotic figures may be present, but there is little significant cellular pleomorphism. This connective tissue characteristically shows numerous small foci of irregular trabeculae that may have similarities to the peculiar kanji shape of trabeculae in fibrous dysplasia of bone. As the lesion matures, the number of ossified islands increases, expands and finally fuses together. This is probably the reason for the increased radiopacity of the lesion on radiographs, along with the increased degree of calcification.\textsuperscript{[2]}

**Management**

The lesion is clearly distinguishable from the surrounding bone and can be relatively easily
separated into one or several large pieces. Intraoral method with extraction method is preferable. Adjacent teeth, neurovascular bundles and bone should be preserved as much as possible.[20] Perform complete surgical resection with curettage, surgical resection, or en bloc resection, depending on the size of the lesion.[21] Radical surgery is required to reduce the tendency of recurrence and the possibility of malignant transformation. The fibrous capsule facilitates surgical resection and resection. The duration of recurrence is unpredictable but varies from 6 months to 7 years after surgery. Therefore, it is recommended that the follow-up period be up to 10 years.[22, 23]

**Conclusion**

Accurate diagnosis of bone fibroma may be obtained by clinicoradiological and histopathological examinations. Although extensive lesions require radical surgical excision, surgical excision is sufficient in most cases of ossifying fibroma. To reduce the risk of recurrence, for extensive lesions, complete resection of the block is necessary. Although the recurrence rate is not very high, regular long-term follow-up is required.

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