Case Report

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Ossifying Fibroma: A Rare Lesion with Rarest Character

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ABSTRACT

Background: A heterogeneous group of benign lesions of unknown aetiology affecting the jaws and other craniofacial bones are called Fibro-osseous lesions. A fibro-osseous lesion of benign and proliferative nature, non-odontogenic in the origin of the jaw is commonly known as Ossifying fibroma. Ossifying fibroma has been reported to occur in long-range of age groups, from children to adults of old age. Can affect both maxilla and mandible but is more frequently affects mandible (70-90%), with a predilection for female patients. It grows slowly and enlarges in size progressively to cause destruction and deformation of the surrounding bone. It is generally accepted that the histological subclassifications of fibro-osseous lesions are of academic interest only since differential diagnosis is often arbitrary and their biological behaviour seems to be identical. Hereby, reporting a case of 20year old male patient who reported to the dental hospital with a complaint of swelling on right side of the face and was diagnosed clinically, radiographically and histologically as a non-aggressive form of an immature ossifying fibroma. Although many cases of Ossifying fibroma have been reported in the published literature, a massive expansile lesions measuring approximately 10 cm, like this case is rare.

Keywords: Fibro-osseous lesions, mandible, ossifying fibroma, mixed radiopaque radiolucent lesion of jaw.

INTRODUCTION

Ossifying fibroma (OF), also known as cemento-ossifying fibroma or cementifying fibroma, is classified under the mesenchymal odontogenic tumour group according to the classification given by WHO in 2005. It is a benign bone neoplasm, a type of fibro-osseous lesions (FOL)[(1)] .These FOL represents a diverse group of lesions characterized by a nearly similar histological appearance[(2)]. This group has very much same clinical presentation, radiographic appearance, and histological criteria. It is therefore, a difficult task for diagnosis, classification, and management by the clinician[(3)]. Such a benign type of FOL can arise in any part of the skull and facial skeleton with > 70 % of cases arising in the upper or lower jaw [(25)]. Previously cemento-ossifying fibroma (COF) term was used to represent two entities depending on the internal content. In one entity, predominant calcified material as bone, for which term ossifying fibroma was used while in second entity cementum-like material was dominant, and the term cementifying fibroma was used[(5,6)]. In 2017 WHO classify ossifying fibromas were into 3 subtypes. The first is designated as cemento-ossifying fibroma and is odontogenic in origin. Others are psammomatoid and trabecular juvenile types[(7)]. Ossifying fibroma demonstrates the proliferation of cellular fibrous tissue with varying amounts of osseous products

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including bone, cementum or a mixture of both. Consisting of thin, isolated trabeculae of bones, trabeculae may show osteoblastic rimming and spherical foci of calcified material, which are relatively acellular resembling cementum[5]. The central variant of ossifying fibroma is relatively rare and is common in females than males with a preference for mandible more than maxilla[7]. From the radiological perspective, the lesion manifests often as unilocular and sometimes multilocular, well-demarcated, mixed radiolucent-radiopaque lesion based on the presence of cementum or bone[4]. The degree of radiopacity depends on the maturity of the lesion. The lesion may show a range of radiolucency or radiopacity, varying from immature lesions which present as a complete radiolucent lesion whereas, the entirely radiopaque representing the mature lesion. However, intermediate lesions can present with varying degrees of radiopacities[4,5]. The lesion is generally encapsulated radiographically.

CASE REPORT

A young male patient aged 18yrs reported to the Department of Oral medicine and radiology on 26 July 2019 with a chief complaint of pain and swelling on the left side of the face for the last 10 days. Swelling is progressive in nature for the last 10 days, with mild continuous pain, difficulty during mastication and foul smell from mouth. Past history suggests that the patient had mild swelling for 7 years in this region, for which he had undergone tooth extraction which was uneventful. The swelling persisted even after extraction but was approximately asymptomatic. He doesn't have any history of tissue abusive habits. His medical and familial histories were non-contributory to the present complaint and his vital signs were within the normal range.

An extraoral examination (Figure. 1) revealed massive swelling over the left lower half of face causing asymmetry with the tense overlying skin, restricted and painful mouth opening. On palpation, the swelling is warm , non-tender, bony hard in nature. Submandibular lymph nodes were palpable, enlarged, mobile, firm, non-tender on left side

An intraoral examination (Figure. 2) shows swelling extending from 34 regions to left retromolar area and causing obliteration of left buccal and lingual vestibule.

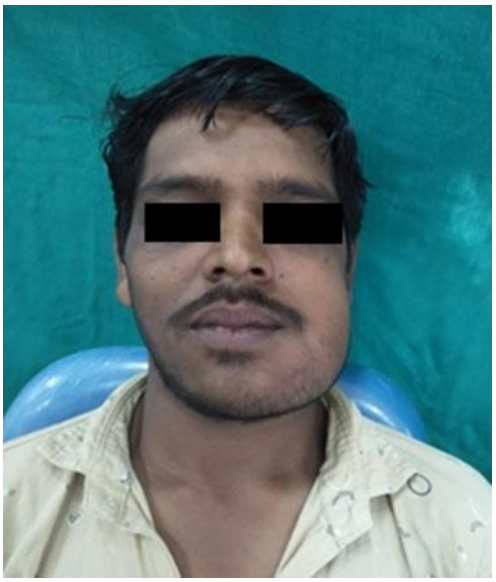

Fig 1: Pre-operative photograph.

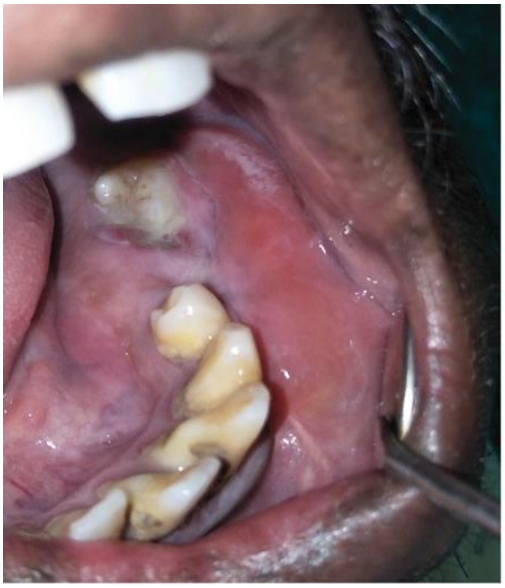

Fig 2: Intraoral view.

Swelling is tender to palpate. There was no associated tooth mobility; however, drifting and displacement were noticed. The related teeth were also vital but displaced. Oral hygiene was poor. The mucosa appeared normal, with no evidence of ulceration or bleeding. Diagnostic guidelines including complete hemogram, serum calcium was suggestive of no abnormalities. Differential diagnosis includes central giant cell granuloma, dentigerous cyst, ameloblastoma, fibrous dysplasia, calcifying epithelial odontogenic tumor.

On radiographic evaluation, mandibular true occlusal view (Figure. 3) shows mixed radiopaque radiolucent lesion with a displacement of adjacent teeth, scalloped well-defined margins and expansion of buccal and lingual cortical plates.

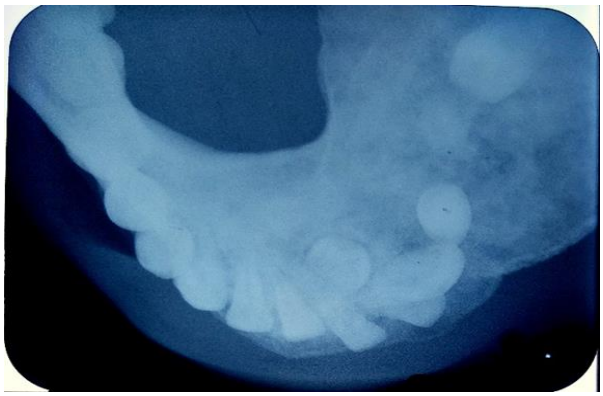

Fig 3: Showing mixed radiopaque radiolucent lesion with the displacement of adjacent teeth, scalloped well-defined margins and expansion of buccal and lingual cortical plates.

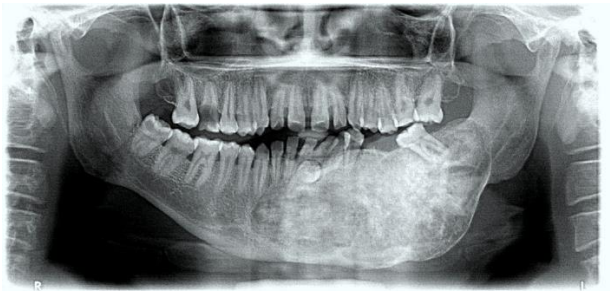

Fig 4: Othopantomograph (Figure. 4) revealed an expansile mixed radiopaque-radiolucent lesion in the lower-left third molar region.

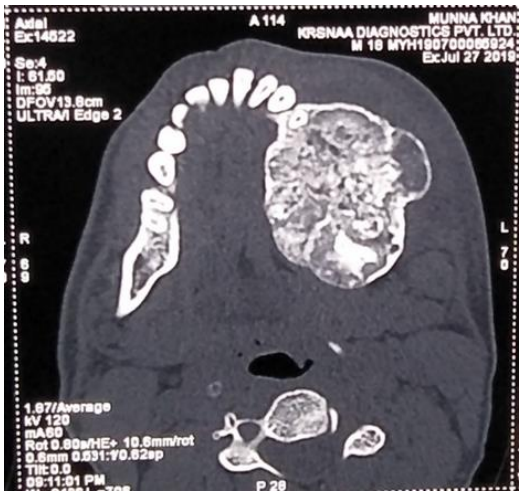

Fig 5: Plain computed tomography imaging of the lesion with 5mm and 2mm thin section at 5mm.

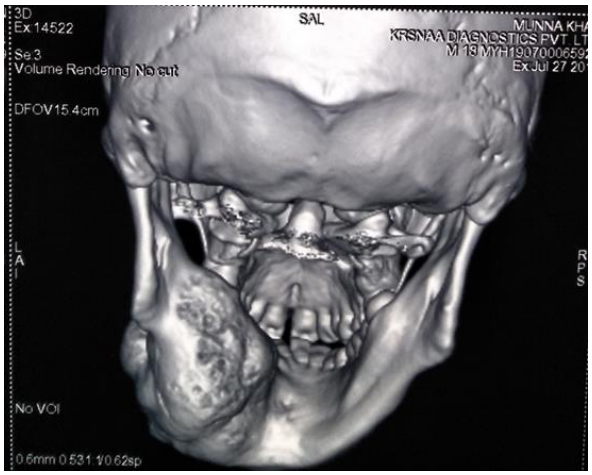

Fig 6: Showing multilocular with ill-defined septa with dense radiopaque masses.

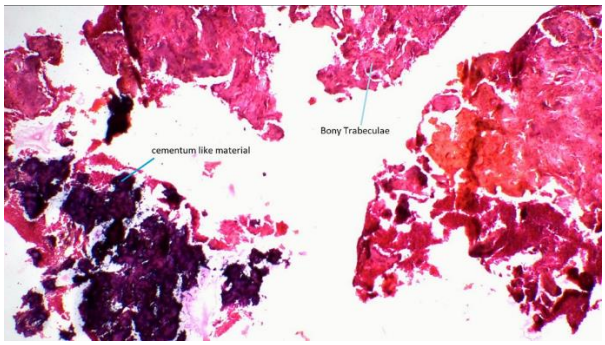

Fig 7: Histological slide shows, moderately dense cellular fibrous connective tissue stroma with irregularly shaped trabeculae of immature bone.

Othopantomograph (Figure. 4) revealed an expansile mixed radiopaque-radiolucent lesion extending from the lower-left third molar region to the contralateral canine crossing the midline without obvious cortical destruction. Superiorly, the lesion extended to the alveolar crest, causing displacement of adjacent teeth. Inferiorly, it spread to the inferior border, causing expansion. Lesion contain an impacted premolar apical to 31. The borders of the lesion were corticated distally and non-corticated in symphyseal region, irregular but well defined and showed some amount of scalloping. The inferior alveolar canal on the left side appeared to be displaced inferiorly.

Plain computed tomography imaging (Figure. 5) of the lesion with 5mm and 2mm thin section at 5mm interval in coronal plane reveals revealed a lesion measuring ~ 5.8 X 4.3 X 4.6 cm in size. Images shows massive buccal and lingual expansion with thinning of the cortical plates without any marked

perforation. The internal structure appears multilocular with ill-defined septa with dense radiopaque masses. 3D CT also supports the findings. (Figure. 6) Differential diagnosis includes Calcifying epithelial odontogenic tumour, Fibrous dysplasia, Ossifying fibroma, Adenomatoid odontogenic tumour.

Multiple soft tissue and bone fragments were taken by open bone biopsy, via the intraoral approach and submitted in 10% formalin for histological evaluation. Histological slide (Figure. 7) shows moderately dense cellular fibrous connective tissue stroma with irregularly shaped trabeculae of immature bone. Fibrous connective tissue shows proliferating fibroblasts with spindle-shaped nuclei. Few areas show coarse woven bone and few areas have extravasated RBCs.

DISCUSSION

Fibro-osseous lesions are lesions of the heterogeneous group of benign nature with a broad spectrum of clinical, histologic and radiographic features [24]. Since 1927, the term ''ossifying fibroma'' has been used, and since 1968 tumours containing-cementum have been grouped together [8]. Ossifying fibroma (OF), has been represented by the MeSH (Medical Search Heading) as ''fibroma, ossifying'' since 1994. MeSH defines OF as ''A benign central bone tumour, usually of the jaws (especially the mandible), composed of fibrous connective tissue within which bone is formed'' [9]. The OF affecting the jaws exhibits a variable behaviour ranging from slow growth to occasionally aggressive local destruction; some cases recur after surgery [10]. Etiologic factors include the existence of previous trauma in the area [11]. Some examples also suggest chromosomal translocations as the etiological agent [3]. In earlier literature, the origin of this tumour is recommended to be either odontogenic or from the periodontal ligament. However, identical neoplasms with the cementum-like material have been identified in the sphenoid, orbital, ethmoid, and frontal bones. This negates the previous theories [5]. OF proliferates following infection, dental extraction, surgery and trauma [1]. Frequency of occurrence is more in females than in males, with a female-to-male ratio of 5:1[14]The age of ossifying fibroma tends to be variable, but predominates in the third or fourth decades of life[4,15,16].The bone most commonly affected is the mandible and most frequent site is the premolar-molar area superior to the inferior alveolar canal. The lesion is generally found to be asymptomatic although the pain was reported in some cases. The growth of the lesion produces a noticeable swelling with mild deformity[4,17]. Although OF has principally been found in the jaws, it has also been reported in the frontal, ethmoid, sphenoid and temporal bones, and in orbit and the anterior cranial fossa [18]. Displacement of teeth is commonly seen but resorption of the root has a rare occurrence. In children, OF has an aggressive form, exhibit rapid growth and occurs in the first two decades of life; they are generally called as ''juvenile ossifying fibroma''[19].

Our case is also consistent in having the above features in that the patient is suffering from facial deformity and gives a history of a slowly growing mass over a period of 7 years. The patient also reported mild pain. The lesion is causing displacement of the tooth without affecting their vitality, and the overlying mucosa remained intact without any ulcer or bleeding.

The affecting the jaws is histologically confirmed not as an OF but only as a fibro-osseous lesion (FOL) [20]. In addition to OF, this histopathological group of injuries includes fibrous dysplasia (FD), florid osseous dysplasia (FOD) and focal skeletal dysplasia (FocOD). The late Charles Waldron wrote ''In the absence of good clinical and radiologic information a pathologist can only state that a given biopsy is consistent with a FOL. With adequate clinical and radiologic information, most lesions can be assigned with reasonable certainty into one of several categories''[20].

Often. OFs are well demarcated and rarely infiltrate the surrounding bone; hence it is easier to remove surgically in comparison to other non-odontogenic or odontogenic tumours [13].

OF has well-defined margins in the jaws [21]. According to Slootweg and Muller, a lesion with a zone of transition of less than 1 mm can be considered to be well defined [22]. This can be quickly and cheaply appreciated on plain film radiographs. FD and OF can be distinguished by radiology; the former has a poorly defined margin, whereas the latter has a well-defined margin [23]. The conventional radiography augmented by

computed tomography is adequate in distinguishing fibrous dysplasia of the jaws from ossifying fibroma in most of the cases.

For cases in which histopathological differentiation between FD and OF is unclear, operative findings should be used because OF is often found to be well-encapsulated and easily enucleated. If the lesion is well-encapsulated and even after excision recurrence occurs, the lesion should be clinically suspected to be OF. Careful observation after the surgery for OF is essential because OF often recurs [2].

Talking about differential diagnosis, it includes other lesions, such as calcifying epithelial odontogenic tumour (CEOT), fibrous dysplasia, calcifying odontogenic cystic (Gorlin cyst), central giant cell granuloma, and adenomatoid odontogenic tumour. Well-defined border of the ossifying fibroma helps to differentiate it from the aggressive mixed lesions as metastatic malignant lesions, osteosarcoma and chondrosarcoma [4].

CONCLUSION

FOLs, though have similar histopathology, have very different clinical and radiological presentations, behaviour, and treatment outcomes. Fibrous dysplasia of the jaw, which though becoming inactive, does not involute and therefore requires aesthetic correction and life-long review. Ossifying fibroma should be removed entirely surgically as it has a probability to recur [12]. COF lesions may grow slow with symptoms often gradual and long-standing in some cases. Follow-up is required for these cases [24].

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

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