

Ossifying Fibroma of the Mandible – Case Report and Literature Review

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Abstract: ***Background:** Ossifying fibroma (OF) is a rare neoplasm of the craniofacial skeleton. It can affect all ages, although it is discovered mostly in the second to fourth decade of life. It occurs predominantly in females and most common site is the mandible (molar and premolar region). OF is a benign tumor with usually slow and asymptomatic development. In some cases described as “juvenile” more rapid and aggressive behavior is observed. **Materials and Methods:** We report a case of 26-year-old woman with OF of the mandible that was presented to our department. The patient was referred to us for clinical evaluation, diagnosis and treatment of an asymptomatic, mixed radiolucent/radiopaque lesion from second left mandibular incisor to the first left mandibular premolar. A decision for excisional biopsy was taken. **Results and Conclusion:** The patient underwent operation under local anesthesia – enucleation of the tumor and peripheral osteotomy, the histopathological findings were conclusive with OF. No recurrence was seen after 2 years follow up.*

Keywords: Ossifying fibroma, Maxillofacial, Mandible

1. Introduction

Ossifying fibroma (OF) is a rare benign tumor of the jawbones consisting of fibrous connective tissue with sites of calcifications resembling bone and/or cement(12,26). It is agreed that the terms ossifying fibroma, cemento-ossifying fibroma and cementifying fibroma describe the same pathologic process(13,15). According to WHO, OF belongs to fibro-osseous lesions along with fibrous dysplasia (FD), florid osseous dysplasia and focal osseous dysplasia(13,15).

OF is further divided into conventional and juvenile (JOF) subtypes. The histological characteristics of both fibrous tissue and bony material may vary in wide range. Woven and/or lamellar bone and/or cementoid could be observed in COF and the structure of the stroma may vary from highly cellular to prominently vascular(13,15). On the other hand the JOF has two well defined subtypes based on the histological findings – trabecular (JTOF) and psammomatoid (JPOF). The most distinctive feature of the JPOF is the formation of “psammomatoid bodies” spherical calcified ossicles containing osteocytes, with basophilic center distributed in dense cellular fibrous stroma.(13) The JTOF forms immature irregular trabeculae with osteoid seams.

Although the data differ most authors report significant female predilection of OF. It can be found in any age from 3 to 84 (15), but most often it occurs in second to fourth decade of life. COF has highest incidence in the mandible (88% according to Kaplan et al. (10) with a definite predilection to the premolars and molars region. JPOF is most frequently found in the sinonasal and orbital bones and the JTOF predominantly affects the jaws with a preference to the maxilla(10). There are occasional reports for OF found in other bones, such as the calvaria (Sze Yin Lam et al reports a case of an occipital OF), frontal, sphenoid, temporal bones and the base of the skull (6,7,12,17).

The pathogenesis of this lesion is still uncertain. There are authors that describe only some of those lesions as neoplastic and other as reactive in nature (15). Most researchers adopt the theory that the COF arises from the

periodontal ligament which contains pluripotential cells capable of forming bone, cement and fibrous tissue, whereas the etiology of JOF is suggested to be from overproduction of the myxofibrous cellular stroma normally involved in the growth of the septa in the paranasal sinuses as they enlarge and pneumatize (13,15).

The radiographic presentation of OF varies from radiolucent (small lesions) through mixed radiolucent/radiopaque to completely radiopaque with thin radiolucent rim in more mature lesions. It usually appears as round lesion with well-demarcated borders often with osteosclerosis. Big lesions may represent as multilocular and in some cases of JOF the borders may be more difficult to determine, which correlates with the more aggressive behavior of those tumors. The lesion could dislocate the roots of the adjacent teeth or in some cases to cause root resorption.

OF usually has slow, asymptomatic growth. The most commonly observed clinical symptom is painless enlargement of the affected bone, which leads to facial asymmetry. Despite the slow growth some lesions could reach huge dimensions. Big lesions could perforate the cortical plate and in such cases crepitation may be presented. Pain is also a common symptom. Inflammation and/or hemorrhage are rare and secondary clinical presentations of this tumor.

Being a benign tumor COF could be treated successfully with enucleation and only larger lesions enforce resection with a subsequent reconstruction usually with iliac or fibular auto transplant, whereas JOF, because of its aggressive growth, shows high recurrence rate after enucleation, which imposes en-bloc resection in healthy borders to be the treatment in choice for those lesions.(8,15,26)

In rare cases OF could occur in association with other bone lesions such as Aneurysmal Bone Cyst (ABC), Central Giant Cell Granuloma (CGCG) and Complex odontoma or it could develop as part of the hyperparathyroidism-jawtumour (HPT-JT) syndrome. Triantafillidou et al presented a case of OF associated with a recurrent ABC previously treated by curettage. CGCG of the jaws is a rare benign lesion

containing osteoclast-like giant cells scattered in a cellular fibrovascular stroma (15,26). Kaplan et al reported three cases of combined CGCG-COF lesions. Matsuo et al observed a case of OF that occurred in the place of a removed complex odontoma. Very rarely synchronous development of multiple OFs not associated with HPT-JT syndrome could be observed (Akcam et al report such case).

Differential Diagnosis of OF is not always easy and should be made based on the thorough clinical and radiographic examination and histopathological findings. Osteoblastoma, fibrous dysplasia, central giant cell granuloma, ameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma, adenomatoid odontogenic tumor, aneurismal bone cyst and calcifying odontogenic cyst could resemble OF in their clinical and radiographic representation and should be considered in the differential diagnosis (1,2,9,11,14,18). Chang et al. (4) observed two cases of OFs misdiagnosed as radicular cyst and adenomatoid odontogenic tumor in which the first diagnosis was based only on the radiographs. Some authors suggest that peritrabecular cleaving may be an important additional microscopic diagnostic criterion to diagnose and differentiate FD from OF. (15, 20, 25, 26)

2. Case Report

We present a 26 years old female with OF of the mandible. The patient was referred to us from her general dentist for clinical evaluation, diagnosis and treatment of an asymptomatic, mixed radiolucent/radiopaque lesion from second left mandibular incisor to the first left mandibular premolar which was revealed on a routine OPG. On clinical examination the patient was without subjective complaints. There was no bone deformation or pain and crepitation on palpation. Radiographic imaging showed radiolucent/radiopaque lesion on left side of lower jaw, with well-defined borders and no resorption of roots of teeth 32,33, unilocular in appearance (Fig.1). OF and calcifying odontogenic cyst were considered as possible, based on the clinical examination and the radiographic findings. Decision for excisional biopsy was taken and the patient was operated under local anaesthesia. The tumour mass was removed through an intraoral approach and curettage with peripheral osteotomy with preservation of the continuity of the mandible with at least a 2 mm margin was performed. Although the vitality pulp test showed that teeth 32, 33 are vital we decided to perform endodontic treatment prior the operation. The patient recovered from the operation normally. The histological findings were consistent with OF (Fig 2).

3. Result

At the 2-year clinical and radiological follow up there was no sign of recurrence. The postoperative defect is almost fully reconstructed (Fig. 3).

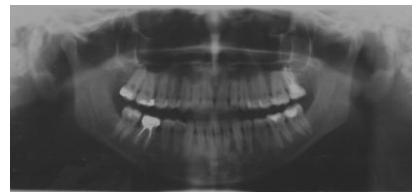


Figure 1: Preoperative OPG

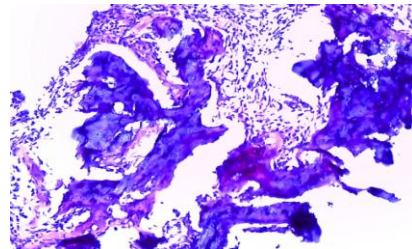


Figure 2: Histological image

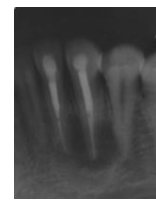


Figure 3: 2 year post operation

4. Discussion

Ossifying fibroma of craniofacial bones is a benign neoplasm, mainly composed of 2 components: fibrous stroma and bone elements that show various degrees of maturation. The stroma consists of fibroblasts and collagenous fibers. Bone elements include mineralized bodies (ossicles), osteoids, fiber bone (woven bone), and mature bone (lamellar bone) (26).

Ossicles connect to form bone trabeculae that usually is surrounded by osteoblasts and occasionally by osteoclasts. Rounded cementum-like masses may be present either alone or together with the trabeculae (3,6,15,17). Because of the variation in the configuration of these calcified deposits, such tumors have been referred to as both ossifying and cementifying fibroma (15,16,22,23,31,32).

Marx and Stern (15) have stated that OF occurs frequently in the jaws, probably because these lesions are related to an extensive mesenchymal cellular induction into bone and cementum, required in odontogenesis. Therefore, when there is an error in the tissue induction process, an OF may be developed in the jaws. It is thought that some fibro-osseous lesions arise from the periodontal ligament, which contains pluripotential cells capable of forming cementum, bone, and fibrous tissue (3,15,16,31). The neoplastic nature of OF is attributed to the fact that large lesions exhibit aggressive behavior, producing significant osseous destruction. Additionally, recurrences, though rare, have been described in some cases of OF. Others categorize this lesion as a localized dysplastic process in which bone metabolism is altered (22,29,31).

By either method of treatment, the recurrence rate for OF is low (15,23). For aggressive tumors that show rapid

enlargement, the recurrence rate is estimated to be 20%-25%. Such tumors will usually require radical resection. It is advisable that the clinician follow these patients with yearly examination. (3,4,5,8,15,31,32) The recurrence rate in the present patients (14%) is in accordance to that estimated in literature.

In any case, the decision on whether to enucleate or resect radically, depends on involvement of the inferior border of the mandible and the spread of the lesion in the soft tissues or in the maxillary sinus and nasal cavity (15,21,28,30). Both surgical methods of treatment for OF (conservative or radical) are acceptable by most authors in the English-language literature during the past 30 years, as reported in their clinical studies (3,4,16,21,24,30,31,32). Conservative surgery was addressed for small lesions, whereas larger lesions required radical surgery. The above surgical protocol was also applied for the patient of the present study.

Differential diagnosis between OF and other fibro-osseous lesions sometimes is difficult, because all of these lesions may exhibit similar clinical, radiographic, and histologic features (23,28,31,32,27). Distinguishing between fibrous dysplasia (FD) and OF is the primary differential diagnostic challenge. The most helpful feature distinguishing FD and OF is the well circumscribed appearance of OF in radiography and the ease with which it can be separated from normal bone in surgery. Histologically, FD is reported to contain only woven bone, without evidence of osteoblastic rimming of bone. The presence of mature lamellar bone in histology is believed to be characteristic of OF (3,15,23,32). If the OF occurs around tooth roots, it may also resemble a cementoblastoma or florid cemento-osseous dysplasia. These rare lesions may be distinguished from an OF by their radiographic appearance. The cementoblastoma is fused to the root of the involved tooth, and florid cemento-osseous dysplasia exhibits not 1 but several sclerotic densities in the alveolar bone of one or both jaws (15, 24)

Conclusion. OF is a benign fibro-osseous tumor, most commonly found in the jaws, especially in the mandible. Buccal or lingual cortical bone swelling or expansion is the most common clinical feature. The radiographic picture of OF is more frequently a well defined mixed lesion (radiolucent/radiopaque). Most OFs can be treated by conservative surgical excision (23). Although the recurrence rate of this tumor seems to be extremely low, the patients must be followed up carefully, particularly because the tumor has proved to be aggressive, recurring sometimes after conservative surgical procedures.

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