

Central ossifying fibroma, periapical cemento-osseous dysplasia and complex odontoma occurring in the same jaw

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Abstract

Central ossifying fibroma is a rare, benign fibro-osseous lesion that arises from the periodontal ligament. Periapical cemento-osseous dysplasia is another variant of fibro-osseous lesion which occurs in the anterior region of the mandible of females. Odontoma is a benign odontogenic tumor that contains enamel, dentine cement and pulp tissue. A 46-year-old woman was referred to the Department of Oral Medicine, School of Dentistry, Tehran University of Medical Sciences, with two non-painful swellings on both sides of the mandible, which had been slowly growing over a period of one year. Our differential diagnosis was florid cemento-osseous dysplasia, focal cemento-osseous dysplasia for the right side, complex odontoma for the left side and periapical cemento-osseous dysplasia for the anterior side. The historical feature revealed ossifying fibroma, complex odontoma and periapical cemento-osseous dysplasia. The occurrence of these three lesions in the same jaw has been rarely reported in the literature. The relationship between the occurrence of these three lesions is not obvious it could be coincidental. It seems that more case reports are needed to establish the relationship between them.

Introduction

Fibro-osseous lesions (FOL) of the jaws constitute a group of conditions which are remarkable for their clinico-pathological similarities.¹ They differ, with the exception of fibrous dysplasia, from those found in the rest of the skeleton. FOLs of the face and jaws include ossifying fibroma, fibrous dysplasia, cemento-ossifying fibroma, benign cemento-blastoma and periapical cemental dysplasia (also known as periapical fibrous dysplasia). They are characterized by the replacement of normal bone by connective tissue, with varying degrees of mineralization in the form of woven bone or cementum-like round acellular basophilic structures.² The designation fibro-osseous lesion is not a specific diagnosis and describes

only a process. Radiology defines their diagnosis because the pathology features on a biopsy specimen may be very similar to lesions of diverse cause, but they differ widely in their behavior and prognosis, from dysplasia, hamartoma, to benign neoplasia. Although these processes have been classified under the encompassing heading of benign fibro-osseous lesions, a more specific diagnosis often is critical because the treatment of these pathoses varies, from review and follow-up to surgical recontouring to complete removal. Many examples can be diagnosed from the clinical and radiographic features, but some require knowledge of the histopathologic, clinical and radiographic features for an appropriate diagnosis.³

The Central ossifying fibroma is considered a rare, benign fibro-osseous lesion and true neoplasm with a significant growth potential. It has been suggested that the origin of these tumors is odontologic, or from periodontal ligament, but similar neoplasm has also been reported in the other cranio-facial bones, making these prior theories questionable. Today, many authorities prefer to designate the cementum-like material present in ossifying fibromas as a variation of bone. It appears ossifying fibromas occur across a wide age range, with the greatest number of cases encountered during the third and fourth decades of life.³ Females are more commonly affected, and multiple lesions are more often seen in black patients. Premolar and molar regions of the mandible are involved far more often than the maxilla. The lesion reveals a painless course, very often only discovered during routine radiological examination. Some lesions produce painless bone deformity and facial asymmetry which on occasion reaches grotesque size.^{1,4} Periapical Cemento Osseous Dysplasia (PCOD) is an asymptomatic condition that occurs in the periapical region of the anterior mandible. Both radiologically and histologically, the lesion is indistinguishable from the cementifying fibroma.⁵ Very often multiple teeth are involved, there appears to be a female predominance (ranging from 10:1 to 14:1), and 70% of cases affect blacks. Most patients are diagnosed initially between the ages 30-50 with the diagnosis almost never made in people younger than age 20. Teeth associated with the lesions were almost vital.³ The lesions are completely asymptomatic and seldom exceed 1 centimeter in diameter. It is unusual for a PCOD to become large enough to produce a detectable expansion of the cortical plate.⁵

Odontoma is the most common benign tumor of odontogenic origin, and is more likely to be hamartomatous than neoplastic.⁵⁻⁷ According to the latest classification from the World Health Organization (WHO, 2005), two types of odontomas can be found - complex odontoma and compound odontoma, the latter

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being twice as common as the former. Complex odontoma is found in the posterior mandibular section, over impacted teeth, and can reach several centimeters in size.⁷ It is usually diagnosed by the radiographic identification of a radiopaque mass and consists of enamel, dentin, cementum and pulp that do not morphologically resemble a tooth. The compound odontoma occurs most commonly in the anterior maxilla and consists in a collection of numerous small teeth. Despite its designation as a hamartoma, the compound odontoma is considered the most common of the odontologic tumors. It is usually diagnosed by the radiographic identification of a multiple small tooth structure. The majority of lesions occur in females in their second and third decades. The lesions are non-aggressive; occasionally one reaches a larger size and causes obvious asymmetry of the jaw. Histological examination of the tissue is generally needed to establish the diagnosis. Treatment of the odontoma requires surgical excision. These lesions are not expected to recur.⁵ Multiple bone lesions with different histological appearance are extremely rare. The purpose of this paper is to report a case in which the patient presented two types of fibro-osseous lesions and odontoma complex simultaneously in the same jaw.

Case Report

A 46-year-old woman presented at the Department of Oral Medicine, School of Dentistry, Tehran University of Medical Sciences, with two slowly growing (over a period on one year) non-painful swellings of both sides of the mandible. There were no extra-oral findings including asymmetry, but on oral examination two symmetric rounded swellings presented on the buccal aspect of the mandible in the first molar region (Figure 1). The overlying mucosa was pinkish in color and firm in consistency, and the texture was smooth. No pain or prasthesia was achieved on palpation. Panoramic and periapical views were ordered. Additionally, vitality tests were done for the incisor periapical lesions to rule out periapical endodontic-based granulomas. All the anterior teeth were vital. The radiographic views revealed: i) a well-defined complete opaque lesion extending from the distal of left second premolar posteriorly to the region of second molar; ii) well-defined pure radiolucencies in the periapical regions of right first incisor; iii) a well-defined mixed radio-opaque and radiolucent lesion extending from the distal of right premolar posteriorly to the mesial of the second molar (Figures 2, 3, 4). Our differential diagnoses were: florid cemento-osseous dysplasia, focal cemento-osseous dysplasia for the

right side and complex odontoma for the left side and periapical cement-osseous dysplasia for the anterior side. According to our surgeons, additional radiographic views such as computed tomographic of mandibular bone were not needed. Thus, surgical excision was performed under local anesthesia.

Microscopic examination of prepared sections revealed: i) matured bony trabeculae with prominent osteoblastic rim, and oval to round cementum-like structures in a cellular connective tissue; ii) entin structures with basophilic border similar to cementum containing connective tissue and bony trabeculae arranged in the round; iii) cellular and collagenous stroma containing extravasated erythrocytes and immature bony trabeculae (Figures 5, 6, 7). Excisional biopsy of intrabony lesion of right mandibular body was ossifying fibroma, the left mandibular was complex odontoma, and periapical lesion was PCOD.

Discussion

FOL of the jaws constitute a group of conditions which are remarkable for their clinico-pathological similarities.¹ They differ, with the exception of fibrous dysplasia, from those found in the rest of the skeleton. The designation fibro-osseous lesion is not a specific diag-

nosis and describes only a process. FOLs of the face and jaws include ossifying fibroma, fibrous dysplasia, cemento-ossifying fibroma, benign cemento-blastoma and periapical cemental dysplasia (also known as periapical fibros dysplasia). They are characterized by the replacement of bone by the connective tissue with varying degrees of mineralization in the form of woven bone or cementum-like round acellular basophilic structures. Radiology defines their diagnosis because the pathology for all FOL is identical, although they differ widely in their behavior, from dysplasia, hamartoma, to benign neoplasia. Furthermore, once diagnosed the management of each is different, ranging from review and follow-up, to surgery.²

Central ossifying fibroma is a rare, benign fibro-osseous lesion that occurs in the premolar and molar regions of the mandible in patients in the third and fourth decades.



Figure 1. Two symmetric rounded swellings presented on the buccal aspect of the mandible in the first molar regions.



Figure 3. Periapical view of periapical cemento osseous dysplasia.



Figure 2. Panoramic view of central ossifying fibroma, periapical cemento osseous dysplasia and complex odontoma.



Figure 4. Periapical view of ossifying fibroma.

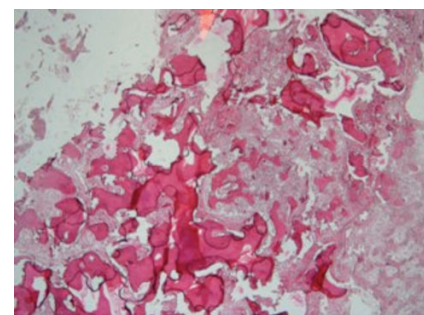


Figure 5. Fibro-osseous lesion. H/E – 40x.

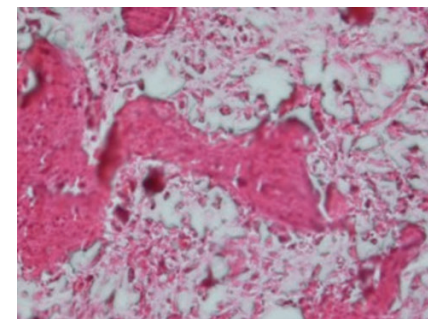


Figure 6. Fibro-osseous lesion. H/E – 400x.

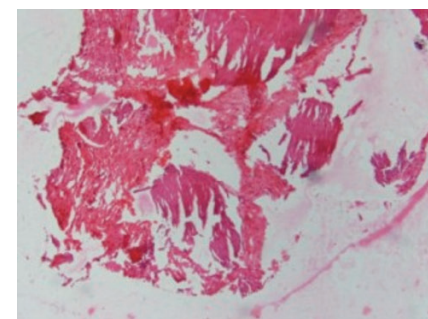


Figure 7. Complex Odontoma. H/E- 40x.

Females are more commonly affected and multiple lesions are often seen in black patients. The lesion reveals a painless course, very often only producing painless bone deformity.^{1,2,4} Pindborg *et al.* defined the central ossifying fibroma as an encapsulated neoplasm containing fibrous tissue, metaplastic bone and mineralized masses with rounded borders and few entrapped cells.⁸ Radiologically, it is typically well defined and unilocular. Some examples show a sclerotic border. Depending on the amount of calcified material produced in the tumor, it may appear completely radiolucent; more often, varying degrees of radiopacity is noted as a mixed radiolucent and radio opaque lesion. It has a spherical shape and can expand cortical bone without causing perforation and may cause tooth divergency or root resorption of teeth associated with the tumor.^{3,4} One important diagnostic feature for this lesion is invasion of the inferior border of the mandible when its size reaches a limit that often demonstrates a characteristic downward bowing of the inferior cortex of the mandible.³ It has a centrifugal growth pattern rather than a linear one, revealing equal expansion in all directions as a round tumor mass.⁴

PCOD occurs in the anterior region of the mandible. Both radiologically and histologically, the lesion has a similar structure to cemento-ossifying fibroma.⁹ Very often multiple teeth are involved, and it appears to be predominant in females. The lesions are completely asymptomatic and seldom exceed 1 cm in diameter. It is unusual for a PCOD to become large enough to produce a detectable expansion of the cortical plate.⁵

It is mainly fibroblastic in the early stages, gradually increasing the amount of bony trabeculae and cementum like tissues.⁹

Odontoma is the most common benign

odontogenic tumor. It is usually asymptomatic and can be found in routine radiologic studies in the second and third decades of life.⁷ Complex odontoma is found in the posterior mandibular section, over impacted teeth, and can reach several centimeters in size.⁵ It is usually diagnosed by the radiographic identification of a radiopaque mass and consists of enamel, dentin, cementum and pulp that do not morphologically resemble a tooth. The compound odontoma occurs most commonly in the anterior maxilla and consists of a collection of numerous small teeth.⁴ Although the lesions are non-aggressive, occasionally they can cause obvious asymmetry of the jaw.⁵

Multiple bone lesions with different histological appearance are extremely rare. To the best knowledge of the authors, this paper reports the first occurrence of these three lesions in the same jaw. However, some comparable literature has been reported.¹⁰⁻¹²

In the present case, the clinical features like age, gender, location of lesions and radiographic features are nearly similar to typical forms. The relationship between the occurrences of these three lesions is not obvious - it could be coincidental. More case reports are needed to establish the relationship between them.

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