

Case Report

Maxillary cemento-ossifying fibroma – A rare occurrence

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Received : 28 December 2022

Accepted : 27 January 2023

Published : 20 February 2023

DOI

10.25259/GJHSR_20_2022

Quick Response Code:



ABSTRACT

Cemento-ossifying fibromas are benign fibro-osseous neoplasm that involves the jaws. These tumors are a rare group of mesodermal odontogenic tumors occurring in the maxilla. Most of the lesions are centrally within the jaw as a slow and progressively growing lesion. If left untreated, they achieve vast size with consequent malformation. Radiographically, they are presented as well-defined unilocular or multilocular intraosseous masses. They are less sensitive to radiotherapy and recurrences are unusual. A rare occurrence of cemento-ossifying fibroma of the anterior maxilla is noted in a 42-year-old male patient. Clinically, they manifested as a well-demarcated swelling in the right anterior region of the maxilla. Radiographic characteristics, histological, and surgical findings are enlisted and the several differential diagnoses are mentioned.

Keywords: Cementifying fibroma, Fibro-osseous lesion, Jaw tumor

INTRODUCTION

Cemento-ossifying fibroma is categorized as a form of distinct benign fibro-osseous (FO) tumor, occurring mostly in the facial region. It occurs more commonly in the mandible in 60–80% of individuals, 22% in maxillary posterior region, and uncommon in the anterior region of maxilla. The common site of occurrence is the premolar region. Females are affected more frequently than males, which are between the third and fourth decades of life. It is not commonly seen in the maxilla.^[1-3] Cemento-ossifying fibroma was classified as a FO neoplasm by the World Health Organization (WHO).^[4] Their resemblance to cemento-osseous dysplasia and ossifying fibroma is evident that they have an odontogenic origin. They develop from the mesenchymal blast cells of the periodontal ligament and can develop into bone, cement, fibrous tissue, or a combination of these things.

The usual bone architecture is replaced by collagen fibers and fibroblasts that contain various levels of mineralized materials. These structures might have a bony or cement-like appearance, and they are known as FO lesions.^[2] This calcification is present in many lesions. Fibrous dysplasia, periapical cemento-osseous dysplasia, localized cemento-osseous dysplasia, florid cemento-osseous dysplasia, and cemento-ossifying fibroma are among the FO lesions.

Three radiographic stages are used to categorize FO lesions: initial or early (radiolucent), mixed (radiolucent and radiopaque), and mature (radiopaque). The radiographic pattern can range from ground glass appearance or being radiolucent to having a more well-defined cyst-like lesion with varied quantities of radiopaque material.^[2,4] This lesion is unusual in that it has a centrifugal development pattern rather than a linear one. As a result, it tends to spread evenly in all directions, causing the bone structure to enlarge, and it manifests as a circular tumor mass.

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Chondrosarcoma, Pindborg's tumor, odontogenic cysts, fibrous dysplasia, squamous cell carcinoma, Gorlin's cyst, and osteosarcoma are among other lesions that might have comparable radiographic characteristics. The radiographic differential diagnosis of cemento-ossifying fibroma is comprised of these abnormalities. The aggressive sarcomas and carcinomas, which present with ill-defined and infiltrative margins on radiographs, can be distinguished from cemento-ossifying fibroma by their well-defined borders.^[5]

To successfully manage this tumor, a correct diagnosis and treatment strategy are necessary. Surgical enucleation along with bone curettage is the treatment of choice. It has been shown that radiotherapy is ineffective. Inadequate surgical treatment may lead to recurrence of the lesions. We present a case of cemento-ossifying fibroma involving the maxillary region, a rare site for its occurrence.

CASE REPORT

A 42-year-old male reported with a complaint of painless swelling in the right upper jaw for the past 6 months [Figure 1]. When the swelling first began, it was small in size. Over time, it slowly increased in size which caused an asymmetry in the face.

Extraoral exploration had shown the presence of swelling on the right quadrant of the maxilla extending from ala of the nose to the angle of mouth [Figure 1]. It measured about 2.0 cm × 1.5 cm in size, hard in consistency, and non-tender on palpation. Lymphadenopathy was not visible on either side of the neck. On intraoral inspection, the swelling extends from the right lateral incisors up to the right first premolar tooth. The mucosa over the swelling appears intact with mild obliteration of buccal vestibule. There was a significant expansion of buccal and palatal cortex with no



Figure 1: Intraoral photographs showing expansion of buccal cortex over the right incisors and premolar region.

evidence of pus discharge at the site of swelling [Figure 2]. Teeth associated with the lesion appears to be mobile and pathologically migrated [Figure 2].

Radiographic examination revealed moderate amount of bone loss in relation to 11, 12, 13, and 14 in orthopantomograph radiograph [Figure 3]. Based on clinical findings, a provisional diagnosis of FO lesion of the maxilla was made.

Routine hemogram and serum chemistry (serum alkaline phosphatase and serum acid phosphatase) values were within normal range. Following oral prophylaxis, an excisional biopsy was planned based on clinical and radiographic investigations. General anesthesia was administered after intraoral and extraoral antisepsis. The procedure includes surgical enucleation of the lesion and bone curettage of the affected area [Figure 3]. Excised fragments were sent for histopathological examination. After a week, the patient was called back for dressing removal and a check-up.

Histopathological examination had shown woven and lamellar bone matrix with osteoblastic rimming. Highly cellular fields with calcified areas are prominent. Cellular element like fibroblasts is arranged in different patterns. Calcified areas appeared to be composed of cementum like



Figure 2: Panoramic radiograph reveals a well-defined unilocular radiolucent lesion in the anterior maxillary region with expansion of cortical plate.



Figure 3: Surgical enucleation with curettage of the lesion.

calcification [Figures 4 and 5]. On correlating the clinical features, radiographic, and histological findings, the final diagnosis was cemento-ossifying fibroma. The patient was reviewed and the follow-up revealed that the size of the swelling had decreased and healing had taken place.

DISCUSSION

Cemento-ossifying fibroma is a benign FO tumor.^[5-10] According to the WHO, it is a delimited or infrequently encapsulated neoplasm made up of fibrous tissue with various levels of mineralized material such as bone and cementum. It affects people of all ages, with the majority of instances occurring in the third and fourth decades of life with a distinct female predilection. It arises from the multipotent cells of the periodontal membrane which are able of producing lamellar bone, cementum, and fibrous tissue.

Radiographically, it is categorized by three stages: initial or early, mixed, and mature stages. In the early stages, the cemento-ossifying fibroma appears as a radiolucent lesion due to proliferation of fibrous tissue occurs.^[8,9] As the tumor advances, flecks of calcification appear making the lesion a mixture of radiolucency and opacity. The lesion appears

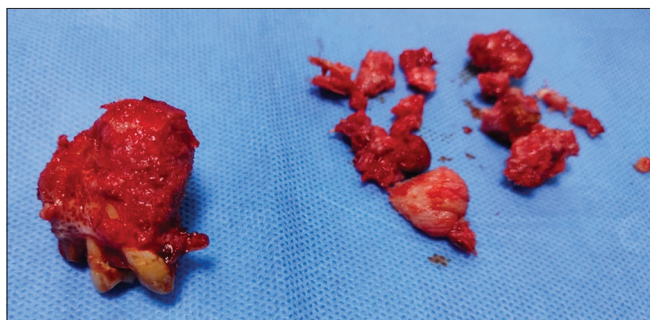


Figure 4: Gross specimen after surgical enucleation.

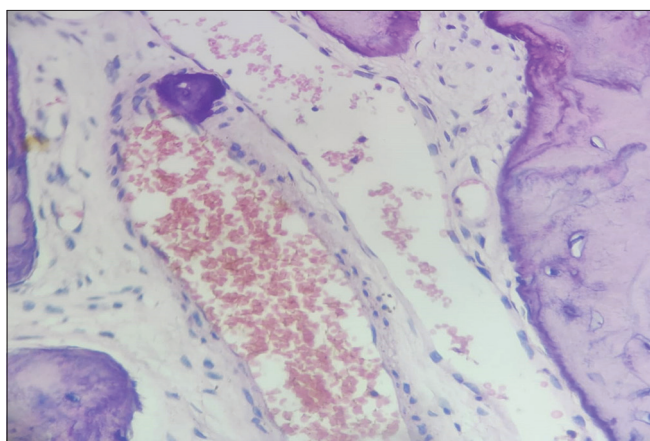


Figure 5: Photomicrograph showing fibrocellular stroma with osseous trabeculae and cementoid tissue.

as a complete radio-opaque mass due to the advanced calcification of bone and cementum.^[3] However, there is controversy over such an origin, since tumors of similar histology have been reported in the bone lacking periodontal ligament and they are not located in the maxillary region, such as ethmoid bone, frontal bone, or even long bones of the body.^[10] This emphasizes the need for a histopathological examination of the biopsy specimen for an accurate diagnosis due to the difficulty in diagnosing cemento-ossifying fibroma based only on clinical observations. Microscopically, cemento-ossifying fibroma reveals many delicate interlacing collagen fibers, seldom arranged in discrete bundles, interspersed with large numbers of active, proliferating fibroblasts, and cementoblasts. Although mitotic figures may be present in small numbers, there is remarkable cellular pleomorphism. As the lesion matures, the islands of cementum increase in number, enlarge, and ultimately coalesce, while one-half of all cases being asymptomatic, the growth of the tumor over time may lead to facial asymmetry, the mass causing discomfort, or maxillary expansion.^[4]

Displacement of teeth may be an early clinical feature.^[7] Most of the lesions typically show slow and often expansile growth, centrally within the jaws, and characteristically behave in a benign form, but occasionally they may present as an aggressive gigantiform lesion.^[5] The majority of instances in the literature have been discovered to have a history of trauma in the area of the lesion, despite the fact that the precise underlying etiology is still unknown. Our patient claimed to have experienced trauma in the affected location, which is consistent with the information from the literature. As a result, this suggests that trauma may have been the triggering factor for the lesion, suggesting that it is a connective tissue reaction rather than a benign tumor. Thus, this points to trauma as a possible triggering factor, postulating the lesion to be a connective tissue reaction rather than a benign neoplasm.^[6] Due to the good delimitation of the tumor, surgical removal and curettage are the treatment of choice. The prognosis is usually good, since recurrences are <25%.

CONCLUSION

The examination of cemento-ossifying fibroma is an extremely complex process, as it requires a clinical, radiological, and histopathological assessment to exclude other FO lesions and odontogenic tumors. Since the tumor is well-delimited, surgical excision and curettage are the preferred course of treatment. When there are really massive lesions with significant tissue involved replacing the damaged tissue presents a challenge after ablation. The prognosis is often favorable, because recurrences are uncommon. This was validated in our situation, because the affected area's repair was found to be accurate a year later.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Geetha R, Pasupathy S, Ragupathy K, Ganesh R. Maxillary cemento-ossifying fibroma – A rare occurrence. *Glob J Health Sci Res* 2023;1:54-7.