Cemento-ossifying fibroma of the mandible: a case report

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European Journal of Medical Case Reports

Volume 4(10):339–342 © EJMCR. https://www.ejmcr.com/ Reprints and permissions: https://www.discoverpublish.com/ https://doi.org/10.24911/ejmcr/ 173-1588719344

ABSTRACT

Background: Cemento-ossifying fibroma (COF) is a relatively rare benign nonodontogenic fibro-osseous lesion, which arises from the mesenchymal blast cells of the periodontal ligament and involves immature bone trabeculae and cementoid formations.

Case Presentation: A 45-year-old female patient presented with a unilocular lesion placed in the posterior region of the mandible. Uniform, rounded swelling was present in the buccal vestibule of the mandible. Clinical, radiologic, and histologic features of COF are discussed in this article.

Conclusion: Although the COF etiology is not exact as yet, clinicians may keep in mind that tooth extraction history may accompany COF, as seen in this case.

Keywords: Cementoma, mandible, fibro-osseous, pathology, tumor, case report.

Received: 06 May 2020 Type of Article: CASE REPORT	Accepted: 14 September 2020 Specialty: Oral and Maxillofacial Surgery	Correspondence to: Sinan Yasin Ertem *Department of Oral and Maxillofacial Surgery, School of Dentistry, Ankara Yildirim Beyazit University, Ankara, Turkey. Email: syertem@ybu.edu.tr
Funding: None.		Full list of author information is available at the end of the article.
Declaration of conflicting inter- no conflict of interest regarding	ests: The authors declare that there is the publication of this article.	

Background

Cemento-ossifying fibroma (COF) is a nonodontogenic benign fibro-osseous lesion and the most common benign fibro-osseous neoplasm of the oral and maxillofacial region. It is mostly seen in young and middle-aged patients (third and fourth decades) and predominantly affects female patients [1]. Generally, COF is seen in the mandible premolar-molar region. In clinical progress, the tumor is asymptomatic, slowly growing, and when the growth causes evident swelling, facial asymmetry may occur. In radiographic process, at early stages, COF barely appears as a radiolucent lesion with a distinct boundary, and when the tumor matures, radiopacity rises, and eventually the lesion appears as an extremely radiopaque mass. Well-vascularized fibro cellular tissue with the capacity of immature bone trabeculae and cementoid formations is seen in the histopathological examination. The treatment procedure of COF is surgical resection of the lesion with enucleation and curettage of the bone [2].

Case Presentation

A 45-year-old female patient presented to our department. The patient did not indicate any systemic disease in her anamnesis and complained about painless intraoral swelling in the posterior region of the mandible. Mucosa over the lesion was normal. In the orthopantomography, 25, 35, and 45 were missing and there were no caries, fillings, and restorations detected. Radiography showed a well-defined, mixed radiolucent, and radiopaque lesion near the mesial root of 46, which extended to the distal side of 44

(Figure 1). Then, cone-beam computerized tomography (CBCT) was taken from the right mandibular posterior region. CBCT showed an expansile lesion with patchy sclerosis at the premolar-molar region of the right mandible (Figure 2). Intraoral examination of the patient showed uniform, rounded swelling present in the buccal vestibule, extending from 44 to 46, and measured approximately 2×1 cm (Figure 3).

Excisional biopsy and surgical enucleation with curettage of the lesion were carried out under local anesthesia and sent for histopathological examination (Figure 4). In macroscopic evaluation, the material contained both soft and hard tissue. The irregular brown hard texture was $1 \times 08 \times 0.5$ cm in size. The complete material was acidtracked and followed on three pieces of one cassette. The soft specimen consisted of two soft brown tissues in $2 \times 1.8 \times 1$ cm dimension. In the examination, some of the material was followed on two pieces and two cassettes.



Figure 1. Panoramic radiography showing the lesion boundaries.

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(A)



(B)

Figure 2. (A) 3D CBCT view of the COF. (B) Coronal view of CBCT image of the COF.

The histopathological examination revealed COF. The specimen showed a benign fibro-osseous lesion with mature cellular fibroblasts on thin fibrillary collagen connective tissue and cement-like spherules and occasional osteoid trabeculae cells. The connective tissue cells that make up the bone trabeculae and cementum structures are plump, usually pale nuclei with occasional spindle cells, and differentiation loss was not observed. When the bone tissue specimen was examined, and a large calcified trabecula with cementum lines was observed in the center with more connective tissue at the periphery (Figure 5).

The patient was reviewed at the first, third, and sixth month of surgery. The patient was clinically asymptomatic and during 1-year follow-up period, there was no recurrence.

Discussion

COF is a benign nonodontogenic tumor, which arises from the mesenchymal blast cells of the periodontal ligament (PDL), seen in teeth bearing jaws and facial bones. It can also be seen in orbit and sinuses, such as maxillary,



Figure 3. Intraoperative intraoral view of the COF.



Figure 4. View of the enucleated COF.



Figure 5. Histopathological view of the specimen.

ethmoidal, frontal, and sphenoidal [2]. As the tumor may progress from an ectopic PDL, it occurs in except teeth bearing jaws. In histological characteristics, the tumor consists of fibrous tissue with different quantities of mineralized substances similar to the bone and/or cementum; this is why it is called COF [3].

The tumor can be seen at any age; however, it mostly occurs between the third and fourth decades of life and predominantly affects women, with a ratio of 1:5 male:fe-male [3,4]. If it occurs in childhood, it is called juvenile aggressive COF. Juvenile aggressive COF is a more clinically aggressive pattern and pathological examination shows more vascularity [5].

Different authors have described radiographic views. MacDonald-Jankowski defined three radiographic stages for COF: early radiolucent stage, followed by a mixed stage, and finally a sclerotic stage [6]. In radiographic images initially, a lesion was seen as a radiolucent area with a distinct boundary, as it extends and matures it is seen as mixed radiolucent and radiopaque areas, and eventually becomes completely radiopaque [7,8]. Actually, a centrifugal growth pattern describes equal expansion in all directions, which is an important feature of COF [9]. According to Barberi et al. [10], there are different radiographic patterns; 40% of the cases show that a defined lesion does not include sclerotic border, and 45% are defined lesions with a sclerotic border. In this case, it is a mixed stage because radiolucent and radiopaque lesions with sclerotic boundaries were examined in the radiographic image. While lesions generally give a completely radiolucent appearance at the initial stage, radiopacity increases due to the progression of the lesion because of the stroma mineralization. Complete radiolucent lesions are seen in younger patients and calcification in the lesion increases with age. Therefore, since the patient's age was 45 years, it is expected to be a mixed stage, considering the size of the lesion. Fibrous dysplasia, cemento-osseous dysplasia, odontogenic cysts, keratocystic odontogenic tumor, calcifying odontogenic cysts, and calcifying epithelial odontogenic tumors can be counted as the differential diagnoses of COF. The etiology is not certain, but according to Galdeano et al. [11], trauma may be the predisposing factor. It has been mentioned that the remnant of PDL due to the trauma or teeth extractions may stimulate the cementum deposition. Likewise, the patient mentioned in this case had a tooth extraction history. It is known that root displacement and, less commonly, root resorption may be seen with COF. Titinchi et al. [12] reported that root resorption was noted in 12.7% of the cases and it was mostly with well-defined multilocular lesions at a younger age. Although the patient was not young and the lesion was not multilocular, in this case, root resorption was seen. Surgical excision and curettage are recommended for the COF treatment. Sheikhi et al. [13] reported that the recurrence of COF may reach a higher rate of 28%. Liu et al. [14] mentioned that the recurrence time of COF is not predictable, ranging from post-operatively 6 months to 7 years. Therefore, long-term follow-up is recommended after the surgery.

Conclusion

COF is a rare benign fibro-osseous lesion that may occur in the jaws, predominantly affects the females, and is seen in third and fourth decades. The present case shows a unilocular, mixed radiographic pattern with a sclerotic boundary, and less commonly expected root resorption was seen. Since the etiology was not exact as yet, clinicians may keep in mind that tooth extraction history may accompany COF, as seen in this case.

What is new?

COF is a relatively rare benign nonodontogenic fibro-osseous asymptomatic lesion. In the current literature, there is no consensus about whether the trauma (tooth extraction) may be the predisposing factor of COF. Some authors have stated that tooth extraction may be counted as a risk factor for COF; however, in our clinical report, a patient with the history of surgical extraction was diagnosed with COF.

List of Abbreviations

CBCT Cone-beam computerized tomography COF Cemento-ossifying fibroma

PDL Periodontal ligament

Conflicts of interests

The authors declare that there is no conflict of interest regarding the publication of this article.

Funding

None.

Consent for publication

Informed consent was obtained from the patient.

Ethical approval

Ethical approval is not required at our institution to publish a case report.

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Summary of the case

1	Patient (gender, age)	Female, 45 years old	
2	Final diagnosis	Cemento-ossifying fibroma	
3	Symptoms	Uniform, rounded swelling presented in the buccal vestibule	
4	Medications	Prescribed oral antibiotics for 5 days. Amoxicillin 1,000 mg tablets, with 1 tablet taken three times daily, and Ibuprofen 600 mg tablets, with 1 tablet taken every 6 hours as needed	
5	Clinical procedure Surgical enucleation with curettage		
6	Specialty	Oral and maxillofacial surgery	