Cemento-Ossifying Fibroma in the Mandibular Posterior Region: 2 Case Reports

Merve Hacer DURAN[1], Mehmet Buğra TÜRKCAN[1], Sümeyye COŞGUN BAYBARS[1], Mustafa ÇAĞDAŞ ÖÇAL[1]

ABSTRACT

Aim Cemento-ossifying fibroma (COF) is a mesenchymal, benign odontogenic tumor of the jaws that originates from the mesenchymal blast cells of periodontal ligament and can form osteoid, bone, cement-like tissue, fibrous cellular tissue or a combination of them. In this case report, it is aimed to present clinical, radiological and histopathological examination of two cemento-ossifying fibroma cases.
Case Report A 36-year-old systemically healthy female patient was referred to our faculty due to a lesion detected in the right mandibular posterior region. As a result of the clinical and radiological examination, an asymptomatic tumoral structure with a mixed appearance and regular borders was detected. No expansion was detected in the right mandibular posterior region. The patient was referred to the Department of Oral and Maxillofacial Surgery for biopsy. According to the biopsy report, it was learned that this tumoral structure was a COF. A 38-year-old systemically healthy female patient was admitted to our faculty due to gingival bleeding. As a result of the clinical and radiological examination, an asymptomatic lesion with a radiolucent appearance and sclerotic borders was detected in the right mandibular posterior region. According to the patient's biopsy report, it was discovered that this lesion was a COF.
Discussion COF may exhibit different clinical and radiological behaviors based on its stage. Diagnosis and treatment planning of COF should be made with clinical, radiological and histopathological examination.
Conclusion Two COF cases are reported with their detailed clinical and radiological examinations in this paper.

Keywords Benign, CBCT, Cemento-ossifying fibroma, Mandible, Odontogenic tumor

Introduction

Cemento-ossifying fibroma (COF) is a mesenchymal, benign odontogenic tumor of the jaws that originates from the mesenchymal blast cells of periodontal ligament and can form osteoid, bone, cement-like tissue, fibrous cellular tissue or a combination of them (1). In 1971, COF was first classified under the cementum-contained lesions including cementifying fibroma, fibrous dysplasia and ossifying fibroma by the World Health Organization (WHO) (2,3). In 2017, the term "cemento-ossifying fibroma" was defined as a mesenchymal, benign odontogenic tumor specific to the tooth- beared areas of the jaws (4). Several synonyms for COF such as osteo-fibroma, fibro-osteoma, and benign fibro-osseous lesion of periodontal ligament origin have been used (5). COF is classified as the central type originating from the periodontal ligament adjacent to the root apex and the peripheral type, which occurs only in the soft tissues of the tooth-bearing regions (6). Although the etiology of COF is not known exactly, there are reports of previous trauma to the lesion site (7). It is also known that it may be associated with congenital problems during the maturation of dental hard tissues that can form cementum and bone. Although it occurs especially in patients aged 20-40, it can occur in children and adolescents as well as in older adults. Women are affected more frequently than men at a ratio of 5:1 (8). This tumor, which is usually seen in the mandibular premolar-molar region, may also involve the maxillary region and paranasal sinuses and larger lesions may be encountered in this region as a result of the larger expansion area (9).

Radiological Features of Cemento-ossifying Fibroma

COF reveals well-circumscribed, mostly unilocular or rarely multilocular radiolucency with or without radiopaque foci native to the degree of calcification (1,10). Initially, COF presents as a radiolucent lesion without internal radiopacity. With the developing tumor maturity, radiopaque masses appear, which can merge to form a large radiopaque focus surrounded by a radiolucent border (11). There are three distinct patterns of radiographic borders: a lesion with no sclerotic border (40%), a lesion with a sclerotic border (45%), and a lesion with poorly defined margins (15%) which implies a fast-growing tumor (12,13). The non-linear centrifugal growth pattern is a specific diagnostic radiographic feature for COF and the lesion enlarges uniformly in all directions and presents as a rounded tumor mass in this pattern (11-13). COF may cause mobility, divergence and root resorption in the adjacent tooth (11,13). In addition, lingual and buccal bone expansion without cortical perforation may be encountered (1,14).

Clinical and Histopathological Features of Cemento-ossifying Fibroma

Clinically, COF is usually observed as an asymptomatic and slow-growing intraosseous lesion but when enlarged it may cause facial asymmetry or bone fractures. Although the adjacent teeth usually remain vital; pain or paresthesia may develop when pressure is applied on the adjacent nerve. It is mostly solitary but

Correspondence: Merve Hacer DURAN, dtmerveduran@gmail.com

[1] Fırat University, Faculty of Dentistry, Department of Dentomaxillofacial Radiology, Elazığ, Turkiye

Received: 13.07.2023 / Accepted: 21.08.2023 / Published: 30.04.2024

may rarely present as isolated or multiple lesions as a component of the hyperparathyroidism-jaw tumor syndrome (1,4,10).

Histologically, COF consists of well-vascularized fibrocellular tissue that capable of producing immature bone trabeculae and cementoids (1,7,14). Bone trabeculae are variable in size, often showing a combination of lamellar and woven patterns. Bone usually shows peripheral osteoblastic and osteoid and rimming (Figure 1). Cementoids are basophilic spherical bodies that mixed with adjacent connective tissue and represent the peripheral brush border (1,15).

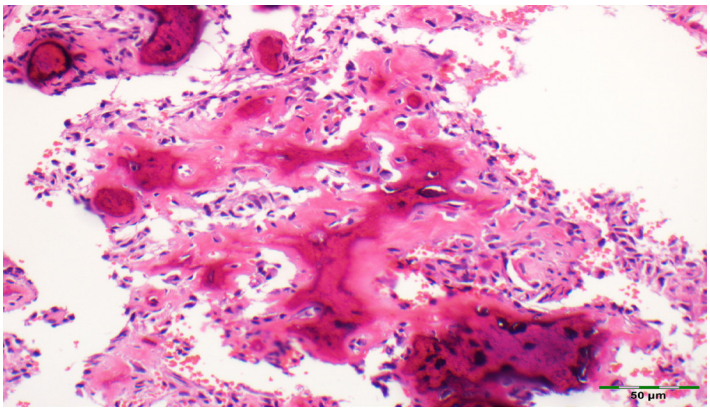

Figure 1: Histopathological section of cemento-ossifying fibroma

The aim of this study is to present two COF cases with clinical, radiological and histopathological findings.

Case Report

Case #1: A 36-year-old systemically healthy female was referred to our faculty from a private dental clinic due to a mandibular lesion. In the radiological examination, a radiolucent lesion with regular borders was detected in the right mandibular posterior region. In the intraoral and extraoral examination, it was observed that the right mandibular first molar was extracted, there was minimal lingual and buccal expansion and the area was asymptomatic. Axial CBCT sections showed thinning of the buccal and lingual cortical bone in the lesion area. Minimal buccal and lingual expansion was observed in coronal CBCT sections. The patient was referred to the surgical department for biopsy with a preliminary diagnosis of residual cyst. Enucleation of the lesion, which diagnosed as COF as a result of incisional biopsy, and right mandibular second molar tooth extraction were performed. In the follow-up radiographs of the third month after the operation, bone formation was observed in the lesion area (Figure 2a-b-c). Written informed consent was obtained from the patient that her medical and dental data could be used in scientific research.

Case #2: A 38-year-old systemically healthy female was admitted to our faculty due to gingival bleeding. As a result of radiological examination, a tumoral structure with mixed appearance and well-defined borders was detected in the right mandibular posterior region. In the intraoral and extraoral examination, it was determined that the right mandibular first molar was extracted, there was no buccal and lingual expansion and the related area was asymptomatic. The patient was referred to the surgical department for biopsy. As a result of excisional biopsy, it was learned that the lesion was COF. Two years later, recurrence was detected in the routine control (Figure 3a-b). Written informed consent was obtained from the patient that her medical and dental data could be used in scientific research.

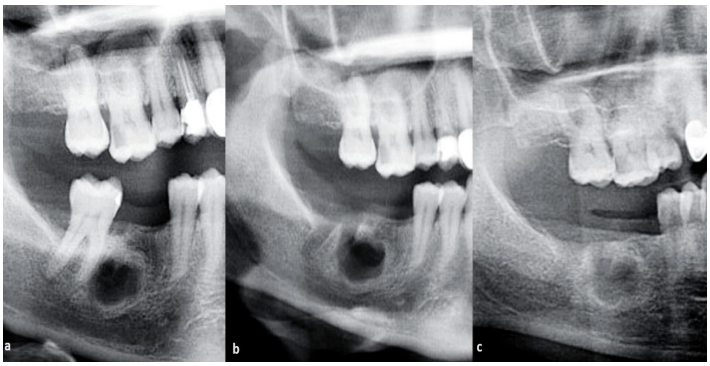

Figure 2: Case #1; a- Before the operation b- Immediately after the operation c- 3 months after the operation

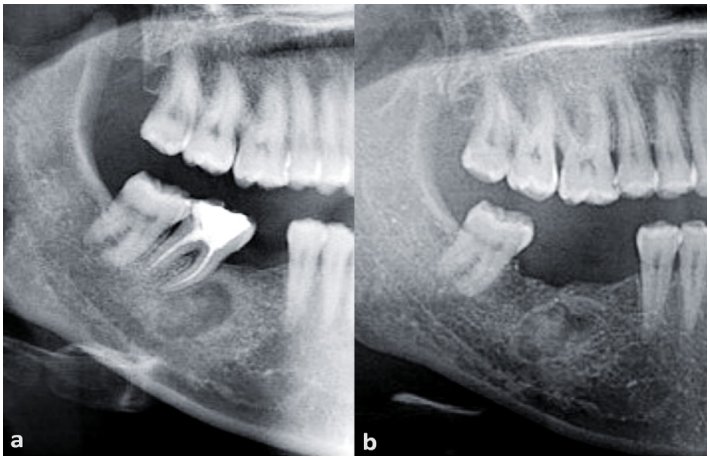

Figure 3: Case #2; a- Before the operation b- 2 years after the operation, recurrence was detected at the previous lesion site

Discussion

COF is a small and usually asymptomatic tumoral formation. However, it has been reported that COF lesions reaching large sizes may cause tooth displacement, nerve compression and paresthesia (16-19). In our cases, there was minimal expansion in the first case and no expansion in the second case. There was no displacement or paresthesia in the teeth. In the radiological examination, COF is radiolucent in the initial stage and it can be observed as mixed and radiopaque in advanced stages due to increased calcification. Radiographically, COF has sclerotic and smooth borders (20). The radiographic pattern of COF has been described by various researchers in the literature. Waldron and Giansanti stated that COF cases were observed as 63% radiolucent lesions with radiopaque foci, 26% radiolucent lesions and 12% diffuse and homogeneous lesions (21).

Titinchi et al. conducted a retrospective study and found that approximately 49.2% of COF lesions were radiopaque, 34.9% mixed radiolucent-radiopaque and 15.9% were radiolucent. Also,

84.1% of the lesions were reported as unilocular and 15.9% of them were multilocular on panoramic radiographs. In addition, multilocular radiolucency was observed in the posterior mandibula in all cases (22). Barberi et al. categorized the radiographic pattern of COF as a prominent lesion without a sclerotic border (40%), a prominent lesion with a sclerotic border (45%) and a lesion with indistinct borders (15%) (23). In our study, the first case was radiolucent with smooth borders and minimally expanded in the buccal and lingual directions, while the second case showed a mixed and regular border.

Differential diagnosis of COF include pathological formations such as central giant cell granuloma, adenomatoid odontogenic tumor, fibrous dysplasia and calcified epithelial odontogenic tumor. Fibrous dysplasia is not well-circumscribed and gives a ground-glass appearance on radiographs. Calcified epithelial odontogenic tumor and adenomatoid odontogenic tumor cannot be exactly differentiated from COF radiologically, but can be distinguished by histological examinations. Central giant cell granuloma is usually seen in younger patients (24). Before a definitive diagnosis can be made; clinical, radiological and histological evaluations should be evaluated as a whole.

Although the surgical approach is controversial in asymptomatic COF cases, enucleation and resection are usually performed (15). Enucleation by curettage is the first choice for small lesions; surgical resection and reconstructive surgery are preferred for larger lesions (1). Radiotherapy has been confirmed to be ineffective and contraindicated due to its inductive effect for malignant transformation (14). In our first case, surgical procedure was performed with the suspicion of cyst. In the second case, surgical procedure was the treatment of choice.

It has been reported that the recurrence rate in COF is 12% and the recurrence rate is higher especially in younger patients. Since the tumor growth is easier in the maxillary region than in the mandible, it has been determined that the recurrence is more common in maxillary cases (18). In our second case, although the patient was middle-aged and the related tumor formation was in the mandible, recurrence was detected two years later.

Conclusion

COF should be suspected in any lesion originating from the mandible or maxilla showing varying amounts of fibrous and osteoid tissue. In the diagnosis and treatment planning of COF, the importance of detecting the lesion at an early stage by performing radiological, clinical and histopathological examinations should be explained to the patients and the necessity of routine periodic controls should be stated to prevent recurrence or future complications after treatment.

Declarations

Author Contributions: *Conception/Design of Study- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Data Acquisition- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Data Analysis/Interpretation- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Drafting Manuscript- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Critical Revision of Manuscript- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Final Approval and Accountability- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Material and Technical Support- M.H.D., M.B.T., S.C.B., M.Ç.Ö.; Supervision- M.H.D., M.B.T., S.C.B., M.Ç.Ö.*

Conflict of Interest: *Authors declared no conflict of interest.*

Financial Disclosure: *Authors declared no financial support.*

REFERENCES

1. *Akkitap M, Gumru B, Idman E, Erdem N, Gumuşer Z, Aksakalli F. Cemento-Ossifying Fibroma: Clinical, Radiological, and Histopathological Findings. Clinical and Experimental Health Sciences. 2020;10(4):468- 472.*
2. *Pindborg JJ, Kramer IRH. Histological typing of odontogenic tumours, jaw cysts and allied lesions. International histological classification of tumours. Geneva: WHO;1971. p.31-34.*
3. *Kramer IR, Pindborg JJ, Shear M. The World Health Organization histological typing of odontogenic tumours; introducing the second edition. Eur J Cancer B Oral Oncol. 1993;29:169-171.*
4. *El-Mofty SK, Nelson B, Toyosawa S. Ossifying fibroma. El- Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, editors. WHO Classification of Head and Neck Tumours. 4th ed. Lyon: IARC Press; 2017. p. 251-252.*
5. *Naik RM, Guruprasad Y, Sujatha D, Gurudath S, Pai A, Suresh K. Giant cemento-ossifying fibroma of the mandible. J Nat Sci Biol Med. 2014;5(1):190-4.*
6. *Bala TK, Soni S, Dayal P, Ghosh I. Cemento-ossifying fibroma of the mandible a clinicopathological report. Saudi Med J 2017;38:541-545.*
7. *Silvestre-Rangil J, Silvestre FJ, Requeni-Bernal J. Cemento-ossifying fibroma of the mandible: Presentation of a case and review of the literature. J Clin Exp Dent. 2011;3(1):66-9.*
8. *Wanzeler AMV, Rohden D, Arús NA, Silveira HLD, Hildebrand LC. Central cemento-ossifying fibroma: clinical-imaging and histopathological diagnosis. Int J Odontostomat 2018;12:233-236.*
9. *Reddy R, Sarkar P, Manuel RA, Saxena D, Hoisala VR. Cementoossifying fibroma: A case report. IJSS Case Reports and Reviews 2016;3:13-15.*
10. *Neville BW, Damm DD, Allen CMA, Chi AC. Oral and Maxillofacial Pathology. 4th ed. St. Louis, Missouri: Elsevier; 2016.*
11. *Mithra R, Baskaran P, Sathyakumar M. Imaging in the diagnosis of cemento-ossifying fibroma: A case series. J Clin Imaging Sci 2012;2:52.*
12. *Rani A, Kalra N, Poswal R, Sharma S. Cemento ossifying fibroma: Report of a case and emphasis on its diagnosis. Indian J Multidiscip Dent 2017;7:140-143.*
13. *Swami A, Kale L, Mishra S, Choudhary S. Central ossifying fibroma of mandible: A case report and review of literature. J Indian Acad Oral Med Radiol 2015;27:131-135.*
14. *Dalghous A, Alkhabuli JO. Cemento-ossifying fibroma occurring in an elderly patient: A case report and a review of literature. Libyan J Med 2007;2:95-98.*
15. *Kharsan V, Madan RS, Rathod P, Balani A, Tiwari S, Sharma S. Large ossifying fibroma of jaw bone: a rare case report. Pan Afr Med J 2018;30:306.*

16. Pandey V, Sharma A, Sudarshan V. Cemento-ossifying fibroma – a rare case report with review of literature. IJCMR 2016;3:2681-2682.
17. Sridevi U, Jain A, Turagam N, Prasad MD. Cemento-ossifying fibroma: A case report. Adv Cancer Prev 2016;1:111.
18. Peravali RK, Bhat HH, Reddy S. Maxillo-Mandibular Cemento-ossifying fibroma: A rare case report. J Maxillofac Oral Surg 2015;14:300-307.
19. Ram R, Singhal A, Singhal P. Cemento-ossifying fibroma. Contemporary Clinical Dentistry. 2012;3(1):83–85.
20. Kaur T, Dhawan A, Bhullar RS, Gupta S. Cemento-ossifying fibroma in maxillofacial region: a series of 16 cases. J Maxillofac Oral Surg. 2021;20:240–245.
21. Waldron CA, Giansanti JS. Benign fibro-osseous lesions of the jaws: a clinical-radiologic-histologic review of sixty-five cases. Oral Surg. 1973;35:340–350.
22. Titinchi F, Morkel J. Ossifying fibroma: analysis of treatment methods and recurrence patterns. J Oral Maxillofac Surg 2016;74(12):2409-19.
23. Barberi A, Cappabianca S, Collela G (2003) Bilateral cementossifying fibroma of the maxillary sinus. Br J Radiol 2003;76:279–280.
24. Trijolet JP, Parmentier J, Sury F, Goga D, Mejean N, Laure B, et al. Cemento-ossifying fibroma of the mandible. Eur Ann Otorhinolaryngol Head Neck Dis. 2011;128:30–3.