CEMENTO-OSSIFYING FIBROMA OF THE MANDIBLE: REPORT OF A CASE

Damla Torul[1], Kadircan Kahveci[1], Mehmet Melih Omezli[1], Ferhat Ayranci[1], Zerrin Unal Erzurumlu[2]*

1. *Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ordu University, Ordu, Turkey.*
2. *Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Ordu University, Ordu, Turkey.*

ABSTRACT

Background and Aim: Cemento-ossifying fibroma is a rare benign neoplasm, affecting the jaws. Because it shares many features with other lesions of the jaws, definitive diagnosis of the cemento-ossifying fibroma is a challenge for the clinician and requires the integration of histopathological, radiological, and clinical examinations. In this article, the diagnosis and surgical management of a cemento-ossifying fibroma, localized in the premolar area of the mandible are presented.

Case Report: A 25-year old male patient, admitted with a complaint of intraoral swelling located on the right side of the mandible. On panoramic radiography, a well-demarcated, unilocular radiolucency was observed between the right mandibular premolars. After radiological and clinical examinations, surgical excision via intraoral approach under local anesthesia was used to totally enucleate the lesion. There were no signs of recurrence.

Conclusion: COF which may be encountered in various radiological and clinical features should be differentiated from other pathologies by considering histopathological, radiological, and clinical features together.

Key words: Cemento-ossifying fibroma, fibro-osseous lesion, mandible, neoplasm, enucleation.

Introduction

Cemento-ossifying fibroma (COF) is an almost rare benign fibro-osseous lesion, which originates from mesenchymal cells of the periodontal ligament [1-3]. The COF can be seen in any part of the facial skeleton such as paranasal sinuses, nasopharynx, and orbitofrontal bone; however, the lesion is detected in the jawbones much more common than other facial region parts [1, 4, 5]. Tooth bearing premolar and molar areas of the mandible are predominantly preferred regions of the lesion, with an incidence of 70 %. COF generally occurs in young adults and middle-aged people and has a marked female predilection with a ratio of 2:1 [1]. Because of the multipotential periodontal ligament cells, that are capable to form bone, cementum, and fibrous tissue, COF microscopically consists of cementoid or calcified material islands within the predominant fibrous tissue matrix [6, 7].

The COF is a well-demarcated and occasionally capsulated lesion, which does not exhibit aggressive behavior [1, 4, 5, 8]. It has a slow growth pattern and remains asymptomatic for a long time unless swelling occurs [2, 8, 9]. In some cases, it may cause symptoms like expansion, and paresthesia or anesthesia in the adjacent nerves due to the increase in the size of the lesion [10]. Radiologically, the lesion may have different appearances according to the stage of development, either mature or immature. In the immature stage, the lesion is mainly seen radiolucent because of the low calcified content, however, radiopaque or mixed appearance can be observed in the mature stage [4, 8].

Because the COF shares similar features with other fibro-osseous lesions like ossifying fibroma, focal cemeto-osseous dysplasia, and fibrous dysplasia in different maturation stages, differentiation of COF from other fibro-osseous lesions causes confusions and requires rigorous histopathological, radiological, and clinical examinations together [5, 11]. Surgical resection or enucleation may be considered as the treatment modalities in the management of this lesion [9, 11]. Although the recurrence of the COF is uncommon, recurrence has been reported in some published cases, treated with enucleation or curettage [12, 13].

In clinical terms, differential diagnosis of the COF, which may have various radiological appearances in different maturation stages, still poses a challenge for the clinician like other fibro-osseous lesions. In addition, the different clinical behaviors of COF ranging from slow growth to aggressive local destruction cause confusions regarding the treatment of the tumor. Thus, the differential diagnosis and treatment approach is very important in terms of the prognosis of the COF. In this article, the diagnosis and surgical management of a cemento-ossifying fibroma, localized in the premolar area of the mandible are presented.

Case Report

A 25-year old male patient admitted to Oral and Maxillofacial Surgery Services of Ordu University with intraoral swelling located on the right side of the mandible. The medical history of the patient was unremarkable except a childhood trauma. The patient stated that swelling had been for a long time but has increased in the last six months. During the extraoral examination of the patient swelling, lymphadenopathy, redness or trismus were not observed. However, the intraoral examination showed an expansion in the buccal region of the mandible between the right premolars (Figure 1). Fistula formation or paresthesia were not observed and all the teeth adjacent to the lesion were responded positively to vitality test. On panoramic radiography, a well-demarcated, unilocular radiolucency

was observed between the right mandibular premolars (Figure 2). Slight displacement was detected in the roots of the right mandibular premolars but resorption or mobility was not present.

After radiological and clinical examinations, a surgical excision via intraoral approach was planned under local anesthesia. Following anesthesia induction with 2% articaine, a mucoperiosteal flap was raised with releasing vertical incisions (Figure 3). The lesion was enucleated totally after reaching the lesion with the help of the burs. Then, a saline solution was used to irrigate the cavity and the primary closure was performed with silk sutures (Figure 4). The specimen was sent to the histopathological examination, and the histopathology report revealed the diagnosis as cemento-ossifying fibroma (Figure 5-6). Periodic follow-up of the patient who was under control for 6 months was ongoing and no recurrence was detected in this period (Figure 7).

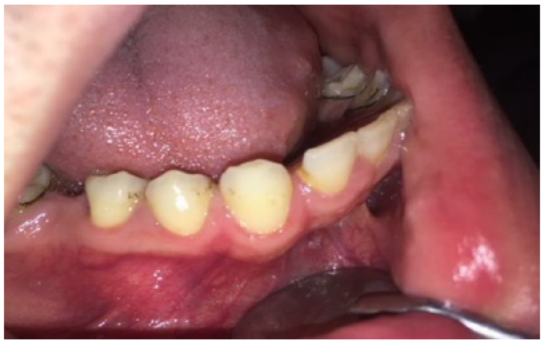

Figure 1: The lesion's clinical appearance before surgery.

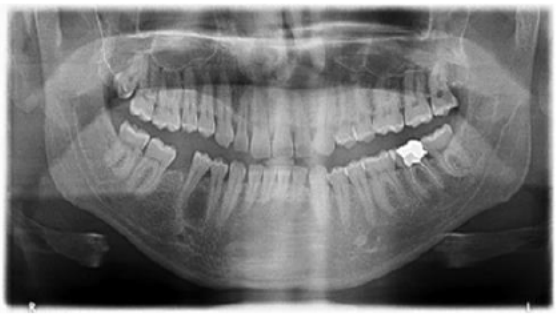

Figure 2: An orthopantomograph showing an apparent radiolucency with sclerotic margins between mandibular right premolars.

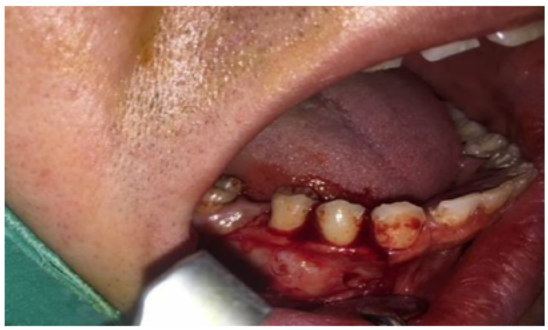

Figure 3: Intra-operative view showing the perforated buccal cortical plate.

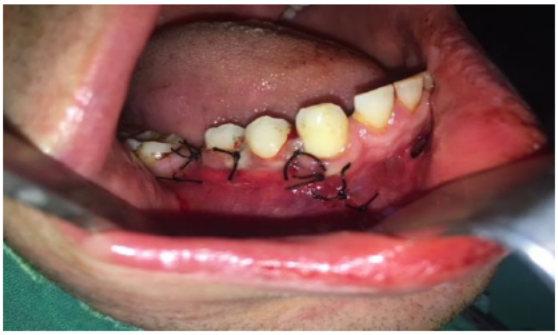

Figure 4: The primary closure was performed with silk sutures.

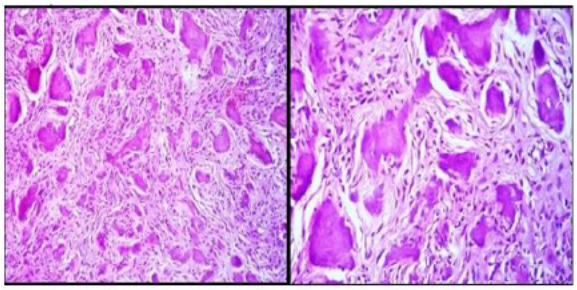

Figure 5: High power histopathological picture showing woven, lamellar bone with osteoblastic rimming deposits and cementum-like calcifications distributed throughout the lesion.

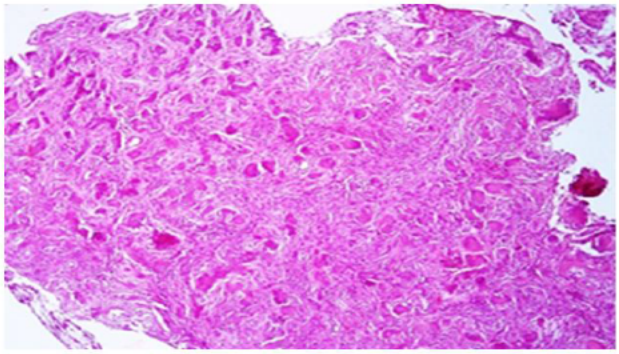

Figure 6: The photomicrography of the COF. Low power histopathological picture showing mineralized, pink amorph droplets within the fibro-collagenous stroma.

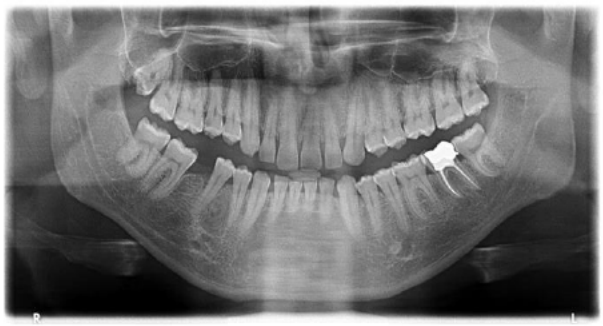

Figure 7: The postoperative 6-month control shows the new bone formation.

Discussion

COF is a benign neoplasm, in which bone is replaced by fibrous tissue, admixed with different amounts of bone or cementum-like calcifications [14]. Although; this benign pathology arises from the mesenchymal cells of the periodontal ligament, the exact mechanism, contributing to the development of this lesion is still under debate. Various theories have been speculated previously. Bernier and Thompson [15] claimed that the infection, which results in inflammation and fibrosis in the periapical area may stimulate the periodontal ligament. Also, it is hypothesized that trauma especially caused by tooth extraction affects the periodontal tissue in the alveolus and triggers the occurrence of the COF [3, 10]. However, these theories failed to explain the occurrence of the COF in other facial regions such as the maxillary antrum, temporal, sphenoid, and frontal bones [5]. The presence of COF in the craniofacial bones outside the jawbones is explained by the ectopic periodontal membrane or the capability of the primitive mesenchymal cells, situated in these bones to differentiate for the generation of COF [3]. In our case history, infection or extraction was not present. We think that the history of childhood trauma, reported by the patient, may be the possible etiologic factor, contributing to the tumor occurrence.

The cemento-ossifying fibroma is usually painless and causes gradually increasing the expansion of the jaws [5]. Loss of vitality or root resorption in adjacent teeth are rarely seen features, thus the lesion is frequently overlooked until a noticeable facial asymmetry occurs [9]. In a systematic review, conducted by MacDonald-Jankowski, it was reported that 84% of the COFs cause buccolingual expansion [16]. Moreover, a clinically noticeable buccal expansion was seen in our case without the presence of any other symptoms. The lesion usually involves the craniofacial bones and is rarely seen in the long bones [17]. Majority of the COF's were detected in the mandible with an incidence of 70%, whereas the lesion also can be detected in the maxilla, orbital, and ethmoidal regions and in petrous bone with the prevalence of about 22% [8]. COF mainly has a marked female predilection and is frequently detected between 2-4 Decades of age [5, 9]. However, different age and gender predilections have also been reported. In a review of 75 patients with COF, conducted by Su et al. it was reported that the patients were between 10-59 years old and the COF showed an equal sex predilection [9, 10]. Also, Dalghous and Alkhabuli [11] reported a patient with COF located in the mandible of a 70-year old female. In our case, COF, involving the right mandible was detected in a 25-year old male patient in concordance with the age-gender predilection, was reported in the article.

The COF has a specific centrifugal pattern of growth that represents the growth of the tumor in all directions, equally [1, 3]. Thus, a typical round tumor mass occurs with a well-defined unilocular or multilocular appearance. Depending on the maturation degree, the lesion radiographically shows an extremely variable opacification [9]. Moshy et al. [18] suggested that these variabilities in the appearance cause confusions regarding the differentiation of COF. Thus, the differential diagnosis of COF from other well-defined radiolucent lesions, containing radiopacities can only be performed by means of histopathological, radiological, and clinical examinations together. Also, the fibro-osseous lesions associated with hereditary hyperparathyroidism-jaw tumor syndrome should be kept in mind for the differential diagnosis in the patients presenting with cemento ossifying fibroma [19]. The COF differs from the malignant lesions with the well-defined border, observed during radiological examination. The lesion differentiates from the fibrosis dysplasia, which is another fibro-osseous lesion, with the absence of ground glass like appearance that is characteristic for fibrosis dysplasia [20]. Unlike the COF, ossifying fibroma contains woven bone and a fibrous tissue rich in osteoid cells [5]. However, the COF contains collagen bundles with lots of fibroblasts and cementoblasts. The cementum-like calcifications determine the characteristic of the lesion [1]. The cementum-like calcifications around the woven and lamellar bone, that are observed during the histopathological examination help to reach a definitive diagnosis of COF.

Treatment methods may vary depending on the size of the case. Lesions that have not reached a large size and have caused minor expansion, can be treated with enucleation or curettage which is the most common technique [12, 21]. The resection and reconstruction may be the preferred treatment option in the cases of recurrence or aggressive lesions that cause serious facial asymmetry and functional problems [9, 22]. In the literature, the rate of recurrence was higher in the cases, treated specially with curettage. Eversole et al. [12] reported the recurrence rate %28 in patients who were treated with curettage or enucleation. Cecchetti et al. [2] treated a patient with curettage and they observed recurrence after 5 years follow-up. Also, recurrence was reported in fibro-osseous lesions that were treated with resection [17]. Titinchi et al. [23] stated that there is not any definitive treatment method that can be applied for all cases and therefore a case-specific treatment should be chosen to achieve good clinical results and minimize the recurrence rates. Also, the periodic follow-up of the patient poses a crucial role in the prevention of recurrences. In our case, by considering the lesion location and size and also the patient's age, we preferred enucleation as the treatment modality.

Conclusion

As a conclusion, COF which may be encountered in various radiological and clinical features should be differentiated from other pathologies by considering histopathological, radiological, and clinical features together. When considering the various clinical behaviors of the COF, determination of the most appropriate treatment option and a long-term follow-up is mandatory

because recurrences can develop for up to 10 years after treatment.

Acknowledgment

The authors would like to thank Dr. Büşra Erşan Erdem, Department of Pathology, Faculty of Medicine, Ordu University for her valuable contribution during the preparation of the pathologic images.

Conflict of Interest

There was no conflict of interest related to this case report.

References

1. Sarwar HG, Jindal MK, Ahmad S. A case report of cemento-ossifying fibroma. Journal of maxillofacial and oral surgery. 2010;9(2):178-81.
2. Cecchetti F, Luciani F, Bramanti E, Bartuli FN, Ottria L, Arcuri C. Cemento-ossyfying fibroma juvenile of the oral cavity. ORAL & implantology. 2010;3(1):33.
3. Bala TK, Soni S, Dayal P, Ghosh I. Cemento-ossifying fibroma of the mandible: A clinicopathological report. Saudi Medical Journal. 2017;38(5):541.
4. Mohapatra M, Banushree CS, Nagarajan K, Pati D. Cemento-ossifying fibroma of mandible: An unusual case report and review of literature. Journal of oral and maxillofacial pathology: JOMFP. 2015;19(3):405.
5. Ong AH, Siar CH. Cemento-ossifying fibroma with mandibular fracture. Case report in a young patient. Australian dental journal. 1998;43(4):229-33.
6. Bertrand B, Eloy PH, Cornelis JP, Gosseye S, Clotuche J, Gilliard CL. Juvenile aggressive cemento-ossifying fibroma: Case report and review of the literature. The Laryngoscope. 1993;103(12):1385-9.
7. Hamner JE, Lightbody PM, Ketcham AS, Swerdlow H. Cemento-ossifying fibroma of the maxilla. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology. 1968;26(4):579-87.
8. Khanna M, Buddhavarapu SR, Hussain SA, Amir E. Cemento-ossifying fibroma of paranasal sinus presenting acutely as orbital cellulitis. Journal of radiology case reports. 2009;3(4):18.
9. Katti G, Khan MM, Chaubey SS, Amena M. Cemento-ossifying fibroma of the jaw. BMJ case reports. 2016 May 12;2016:bcr2015214327.
10. Naik RM, Guruprasad Y, Sujatha D, Gurudath S, Pai A, Suresh KV. Giant cemento-ossifying fibroma of the mandible. Journal of Natural Science, Biology, and Medicine. 2014;5(1):190.
11. Dalghous A, Alkhabuli JO. Cemento-ossifying fibroma occurring in an elderly patient. A case report and a review of literature. Libyan journal of Medicine. 2007;2(2):95-8.
12. Eversole LR, Leider AS, Nelson K. Ossifying fibroma: a clinicopathologic study of sixty-four cases. Oral surgery, oral medicine, oral pathology. 1985;60(5):505-11.
13. Mayo K, Scott RF. Persistent cemento-ossifying fibroma of the mandible: report of a case and review of literature. Journal of Oral and Maxillofacial Surgery. 1988;46(1):58-63.
14. Mainville GN, Turgeon DP, Kauzman A. Diagnosis and management of benign fibro-osseous lesions of the jaws: a current review for the dental clinician. Oral diseases. 2017;23(4):440-50.
15. Bernier JL, Thompson HC. The histogenesis of the cementoma: Report of 15 cases. American journal of orthodontics and oral surgery. 1946;32(9):A543-55.
16. MacDonald-Jankowski DS. Ossifying fibroma: a systematic review. Dentomaxillofacial radiology. 2009;38(8):495-513.
17. Liu Y, Wang H, You M, Yang Z, Miao J, Shimizutani K, Koseki T. Ossifying fibromas of the jaw bone: 20 cases. Dentomaxillofacial Radiology. 2010;39(1):57-63.
18. Moshy J, Dimba E, Ocholla T, Chindia M. Characteristic radiological and histological patterns of fibrous dysplasia and ossifying fibroma of the jaws at University of Nairobi Dental Teaching Hospital. Surgical Science. 2012;3(04):189.
19. Fanibunda K, Reed M. Cemento-ossifying fibroma of the mandible. Dentomaxillofacial Radiology. 1997;26(4):246-8.
20. Kuta AJ, Worley CM, Kaugars GE. Central cementoossifying fibroma of the maxillary sinus: a review of six cases. American Journal of Neuroradiology. 1995;16(6):1282-6.
21. Mishra AK, Maru R, Dhodapkar SV, Jaiswal G, Kumar R, Punjabi H. Peripheral cemento-ossifying fibroma: A case report with review of literature. World Journal of Clinical Cases: WJCC. 2013;1(3):128.
22. Verma P, Rathore PK, Mrig S, Pal M, Sial A. Cemento-ossifying fibroma of the maxilla: a case report. Indian Journal of Otolaryngology and Head & Neck Surgery. 2011;63(1):38-40.
23. Titinchi F, Morkel J. Ossifying fibroma: analysis of treatment methods and recurrence patterns. Journal of Oral and Maxillofacial Surgery. 2016;74(12):2409-19.

Corresponding Author

Kadircan Kahveci
Department of Oral and Maxillofacial Surgery,
Faculty of Dentistry, Ordu University,
Ordu, Turkey.
Postal Code: 52200
Phone: 0 452 212 12 83.
Email Id: kahveci__kadircan@hotmail.com