Clinics in Surgery

Case Report
Published: 04 Oct, 2018

Two Different Odontogenic Tumor in the Mandible: A Case Report

Akyol UK[1], Akyol A[2] and Bedir R[3]*

[1]Department of Maxillofacial Surgery, R.T.E University, Turkey

[2]Department of Maxillofacial Surgery, Private Dental Clinic, Turkey

[3]Department of Pathology, R.T.E University, Turkey

Abstract

Cemento-Ossifying Fibroma (COF) is a benign lesion containing fibrous tissue and calcified material resembling bone and/or cementum, and may be encapsulated or unencapsulated. They occur most frequently in the third and fourth decades, in females, and in the premolar/molar areas of the mandible. They are generally slow-growing and may cause functional deformities in patients at advanced stages.

Another lesion which may arise in the mandible is the keratocystic Odontogenic Tumor (KCOT). These are developmental epithelial cysts which remain asymptomatic until they cause bone expansion, infection, pain, edema, or perforation. KCOT arises about twice as frequently in the mandible as in the maxilla. Like COF, they are typically diagnosed in the third and fourth decades, and have a high recurrence rate.

In this case report, we present the radiographic findings and surgical treatment of a patient who developed both COF and KCOT in different parts of the mandible 26 years after being treated for a KCOT that involved the entire mandible.

Case Presentation

A 64-year-old man with a 1-month history of fistula was referred to our clinic for further investigation and treatment. On oral examination, a fistula was noted on the retromolar region of the left mandible. Aspiration of fluid from the fistula site or mandibular bone expansion was not observed. Paresthesia of the inferior alveolar nerve was not clinically evident. The patient had no history of systemic disease. His medical history included an operation of the mandible 26 years earlier [1-4].

On evaluation of his previous medical records and radiographs, an extensively large radiolucent cystic lesion completely filling the mandibular corpus was observed on the panoramic film. Twenty-six years earlier, following a histopathologic diagnosis of extensive KCOT from biopsy material, the patient was treated with marsupialization and enucleation surgery (performed by A.A., DDS). An iodoform gauze drain was used for decompression of the cyst. The patient was followed with panoramic control films for 5 years following the surgery. It was noted that the final radiograph demonstrated complete resolution of the lesion with new formation of normal bone. The patient stated that he has discontinued the follow-up visits since he has no complaint of pain, extra-and/or intraoral swelling, drainage, or neurologic impairment of his mandible during the 21-year period since the last follow-up examination [5,6].

Panoramic radiographs taken at the patient's recent presentation to our clinic revealed two large, unconnected lesions on the left and right sides of the mandible. The lesion on the left side of the mandible extended from the molar area to the ramus and appeared multiocular, and was larger than the lesion on the right side. The left lesion was observed to be displacing the mandibular alveolar ridge downward. The other lesion appeared uniocular. Incisional biopsy of the lesions showed that they were not histomorphopathologically similar [7,8].

For the left lesion, hematoxylin and eosin–stained sections showed cementum-like material (black arrow) within fibrous stroma. It consisted of a mineralized component with woven bone (black arrow) and fibrous stroma. The lesion on the right mandible showed extensive desquamation of keratinized epithelium into the lumen. It extended into the adjacent soft tissues and presented

OPEN ACCESS

Correspondence:
Utkan Kamil Akyol, Department of Maxillofacial Surgery, R.T.E University, Rize 53020, Turkey, E-mail: utkankamilakyol@yahoo.com

Received Date: *28 Aug 2018*
Accepted Date: *01 Oct 2018*
Published Date: *04 Oct 2018*

Citation:
Akyol UK, Akyol A, Bedir R. Two Different Odontogenic Tumor in the Mandible: A Case Report. Clin Surg. 2018; 3: 2134.

a characteristic lining with parakeratinized epithelial surface (black arrow). Based on the clinical imaging and histopathologic features, a diagnosis of COF (left) and KCOT (right) was established.

The patient's serum Calcium (Ca), Phosphorus (P), and Parathyroid Hormone (PTH) levels and blood values were tested to determine whether the lesions were a result of hyperparathyroidism. All values were within normal limits.

Under general anesthesia, the mandibular cysts were enucleated and Cornoy's solution was applied to right lesion cavity (by U.K.A). The postoperative course was favorable, and four months later the patient reported no discomfort in the area. The patient is still under periodic clinical and radiographic follow-up.

Discussion

COF forms from mesenchymal blast cells of the periodontal ligament, is nonodontogenic, and is classified as a fibro-osseous neoplasma [9,10]. Due to its relatively slow growth, the cortical bone layer surrounding the lesion remains intact, and diagnosis may be delayed [8,11]. We estimate that in the present case, this process lasted approximately 21 years, or until the fistula formed in the left mandible. Pathologic examination results in the patient's records reported that the lesion enucleated after marsupialization 26 years earlier was a KCOT. It is possible that COF developed from periodontal ligament remnants in the left mandible after the KCOT healed. There are different opinions regarding the development of COF. The tumor is also seen in bones that do not contain periodontal ligaments such as the ethmoid bone, frontal bone, and the long bones. On the other hand, some authors claim that trauma to the COF area, tooth extraction, and periodontitis are also triggering factors. Furthermore, COF may also develop in hormonal disorders such as hyperparathyroidism with hypercalcemia [12-14] Hyperparathyroidism-Jaw Tumor Syndrome (HPT-JT) can lead to recurrent or multiple COFs in the jaws. Our patient exhibited normal serum Ca, P, and PTH levels. He did not have teeth in the left mandible, nor did he have a history of trauma. The data in the literature regarding the clinical characteristics and origin of COFs are scarce and controversial. Previous studies have focused more on lesions that develop after COF treatment than the lesions that cause COF. Aneurysmal bone cyst and central giant cell granuloma are such lesions [15,16]. These lesions have been attributed to the differentiation of mesenchymal cells to osteoclast giant cells mediated by yet unidentified triggers as a result of reactions causing stromal changes in COF [16,17].

Like the COF in our patient's left mandible, the KCOT in his right mandible was a clinically silent lesion until the development of bone expansion or infection [6]. KCOT is an epithelial developmental cyst that commonly arises in the mandible and has a characteristic parakeratinized squamous epithelium lining [8]. In the current case, KCOT recurrence was diagnosed in the patient's right mandible 21 years after his last follow-up. It is purported that KCOT originates from the residual dental lamina epithelium and oral epithelium basal cells [19]. They are known to frequently arise in the posterior ramus area of the mandible [20]. Initial treatment of the patient 26 years earlier was marsupialization to the entire mandibular corpus, followed by enucleation. According to the literature, rates of KCOT recurrence range from 2.5% to 62.5%, and recurrence occurs at least 5-7 years or later after treatment [21,22]. Our case is consistent with these literature data. Active mural growth and epithelial proliferation are factors known to induce KCOT recurrence [6].

There are case reports in the literature of KCOT co-occurring with odontogenic cyst or calcified dentigerous cyst, and aneurysmal bone cyst with ossifying fibroma in different locations of the same patient [23-26]. There is only one reported case of ossifying fibroma and KCOT occurring simultaneously on different parts of the jaw in the same patient [27].

Our patient fully recovered after treatment 26 years earlier of a KCOT that completely covered his mandible, but 21 years after his last follow-up radiography, recurrence was observed as unconnected COF and KCOT in the mandible. Although COF and KCOT are relatively common lesions, we found no other cases in the literature of recurrence manifesting as COF in one part of the mandible and KCOT in another 26 years after surgical treatment of a full mandibular KCOT. We recommend long-term clinical and radiographic follow-up for all KCOT patients.

References

1. Buchet C, Baralle MM, Gosset P, Lecomte-Houcke M, Donazzan M. Maxillary ossifying fibroma: Apropos of 3 cases. Rev Stomatol Chir Maxillofac 1994;95(2):95-7.
2. Hemalatha VT, Austın RD, Mathew P. Juvenile Ossifying Fibroma Masquerading as Maxillary Sinus Malignancy: Case Report. Turkiye Klinikleri J Dental Sci. 2012;16(3):305-10.
3. Smith SF, Newman L, Walker DM, Papadopoulos H. Juvenile aggressive psammomatoid ossifying fibroma: an interesting, challenging, and unusual case report and review of the literature. J Oral Maxillofac Surg. 2009;67(1):200-6.
4. Speight PM, Carlos R. Maxillofacial fibro-osseous lesions. Current Diag Pathol. 2006;12(1):1-10.
5. Shear M. Developmental odontogenic cysts. An update. J Oral Pathol Med. 1994;23:1-11.
6. Cawson RA, Odell EW. Essentials of Oral Pathology and Oral Medicine; Churchill Livingstone, 6th Edition. Edinburgh. Cysts of the Jaws. 1998:106-8.
7. Partridge M, Towers JF. The primordial cyst (odontogenic keratocyst): Its tumour-like characteristics and behaviour. Br J Oral Maxillofac Surg. 1987;25(4):271-9.
8. Sapp JP, Eversole LR, Wysocki GP. Bone lesions. In: Sapp JP, Eversole LR, Wysocki GP, editors. Contemporary oral and maxillofacial pathology. 2nd ed. Mosby Elsevier Science; 2004;116-7.
9. Liu Y, Wang H, You M, Yang Z, Miao J, Shimizutani K, et al. Ossifying fibromas of the jaw bone: 20 cases. Dentomaxillofac Radiol. 2010;39(1):57-63.
10. Vegas-Bustamante E, Gargallo-Albiol J, Berini-Aytés L, Gay Escoda C. Benign fibro-osseous lesions of the maxillas: Analysis of 11 cases. Med Oral Patol Oral Cir Bucal. 2008;13(10):E653-6.
11. Gondivkar SM, Gadbail AR, Chole R, Parikh RV, Balsaraf S. Ossifying fibroma of the jaws: Report of two cases and literature review. Oral Oncol. 2011;47(9):804-9.
12. Koury ME, Regezi JA, Perrott DH, Kaban LB. Atypical fibro-osseous lesions: Diagnostic challenges and treatment concepts. Int J Oral Maxillofac Surg. 1995;24:162-9.
13. Martín-Granizo R, Sánchez-Cuellar A, Falahat F. Cemento ossifying fibroma of the upper gingivae. Otolaryngol Head Neck Surg. 2000;122(5):775.
14. Yamashita Y, Akiyama T, Mizusawa N, Yoshimoto K, Goto M. A case of hyperparathyroidism-jaw tumour syndrome found in the treatment of an ossifying fibroma in the maxillary bone. Int J Oral Maxillofac Surg. 2007;36(4):365-9.

15. Prado Ribeiro AC, Carlos R, Díaz KP, Gouvêa AF, Vargas PA. Bilateral central ossifying fibroma affecting the mandible: report of an uncommon case and critical review of the literatüre. Ana Carolina Prado Ribeiro. 2011;111(2):e21-6.

16. El Deeb M, Sedano HO, Waite DE. Aneurysmal bone cyst of the jaws. Report of a case associated with fibrous dysplasia and review of the literature. Int J Oral Surg. 1980;9(4):301-11.

17. Kaplan I, Manor I, Yahalom R, Hirshberg A. Central giant cell granuloma associated with central ossifying fibroma of the jaws: A clinicopathologic study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007;103(4):35-41.

18. Barnes L, Eveson JW, Reichart P, Sidransky D. Pathology and genetics of head and neck tumours. Lyon: IARC. 2005.

19. Hyun HK, Hong SD, Kim JW. Recurrent keratocystic odontogenic tumor in the mandible: A case report and literature review. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2009;108(2):7-10.

20. Ahlfors E, Larsson A, Sjogren S. The odontogenic keratocyst: A benign cystic tumor? J Oral Maxillofac Surg. 1984;42(2):10-9.

21. Voorsmit RA, Stoelinga PJ, van Haelst UJ. The management of keratocysts. J Maxillofac Surg. 1981;9(4):228-36.

22. Forssel K, Forssel H, Kahnberg KE. Recurrence of keratocysts in a long-term follow-up study. Int J Oral Maxillofac Surg. 1988;17(1):25-8.

23. Basile JR, Klene C, Lin YL. Calcifying odontogenic cyst with odontogenic keratocyst: a case report and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010;109(4):e40-5.

24. Zhang LL, Yang R, Zhang L, Li W, MacDonald-Jankowski D, Poh CF. Dentigerous cyst: A retrospective clinicopathological analysis of 2082 dentigerous cysts in British Columbia, Canada. Int J Oral Maxillofac Surg. 2010;39(9):878-82.

25. Ribeiro AC, Carlos R, Díaz KP, Gouvêa AF, Vargas PA. Bilateral central ossifying fibroma affecting the mandible: report of an uncommon case and critical review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111(2):e21-6.

26. Kaplan I, Manor I, Yahalom R, Hirshberg A. Central giant cell granuloma associated with central ossifying fibroma of the jaws: A clinicopathologic study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007;103(4):e35-41.

27. Shimamoto H, Kishino M, Okura M, Chindasombatjaroen J, Kakimoto N, Murakami S, et al. Radiographic features of a patient with both cemento-ossifying fibroma and keratocystic odontogenic tumor in the mandible: A case report and review of literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2011;112(6):798-802.