Cumhuriyet Dental Journal

Volume 17 Supplement 1

doi:10.7126/cdj.58140.1008002107

available at http://dergipark.ulakbim.gov.tr/cumudj/

CASE REPORT

Calcifying epithelial odontogenic tumor: a case report

M. Selim Yavuz, DDS, PhD[a], M. Cemil Büyükkurt, DDS, PhD[a], M. Melih Ömezli, DDS, PhD[b], Sare Şipal, DDS, PhD[c]

[a]*Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Sifa University, İzmir, Turkey*
[b]*Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ordu University, Ordu, Turkey*
[c]*Department of Pathology, Faculty of Medicine, Atatürk University, Erzurum, Turkey*

ARTICLE INFO

Article history:
Received 15 April 2013
Accepted 28 Mayl 2013

Keywords:
Calcifying epithelial odontogenic tumor,
Pindborg tumor,
Mandible

ABSTRACT

The calcifying epithelial odontogenic tumor (CEOT) is a rare benign odontogenic tumor. It was firstly described by Pindborg, and thus, is also referred to as the "Pindborg tumor." CEOT accounts for approximately 0.4-3 % of all the odontogenic tumors. Pindborg tumor is a benign but locally aggressive. It usually presents as a hard painless mass, generally affecting the mandible. Epithelial calcified odontogenic tumor usually found in the posterior mandibular bone, and it shares many features with the ameloblastoma. In this case report, clinical, radiological findings and surgical treatment of the patient with Pindborg tumor were presented.

INTRODUCTION

The calcifying epithelial odontogenic tumor (CEOT) also known as Pindborg tumor is an uncommon odontogenic neoplasm with a variable biologic behavior ranging from very mild to moderate invasiveness.[1-3] Pindborg tumor is located either intraosseously or extraosseously in sequence with 94% and 6%.[4,5] The age range of all patients with CEOT varies between 8 and 92 years at the time of diagnosis with a mean of 40 years.[4-6] Although CEOT shares many features with the ameloblastoma, microscopically, there is no resemblance to the ameloblastoma.[1,2] It is accepted that it derives from the epithelial elements of the enamel organ, but there are controversies about which part it derives exactly from, being able to originate from the external epithelium or from the stratum intermedium of the enamel organ.[2,7] In approximately 50% of the cases the

Corresponding author at: Yrd. Doç. Dr. Mehmet Melih ÖMEZLİ, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ordu University, 52100, Ordu, Turkey, Business Tel: +90.452.2121286, Fax: +90.452.2121289. E-mail: mmelihomezli@hotmail.com

*This case report was presented at the International Oral and Maxillofacial Surgery Society Congress.

tumor is associated with impacted teeth and is twice as prevalent in the mandible than the maxilla.[8] The lesion appears to occupy a position between hamartoma and aggressive neoplasm; however there is no question that complete excision the CEOT is required.[3] Surgical procedure may involve conservative enucleation, marginal (segmental) resection or partial resection in larger and more aggressive tumors, or very rarely, composite resection in cases showing malignant transformation and invasion.[9]

CASE REPORT

We report a case of 53 year-old male patient with Pindborg tumor in mandibular molar area. The patient appealed to the Ataturk University, Faculty of Dentistry for prosthetic treatment, and before prosthetic treatment, a panoramic radiograph was taken for routine radiographic examination. In the panoramic radiograph, a multilobulated radiolucent lesion was seen in the right mandibular premolar-molar area. (Figure 1) Therefore, the patient was referred to our clinic. He has just anterior teeth in the maxilla and mandible. The patient has no complain from this lesion, although there is a slight swelling on his face. No clinical signs or symptoms were associated with this lesion. He was healthy, the medical history and physical exam were unremarkable, and there was no family history of any similar problems or any developmental abnormalities. Intraoral appearance of the patient was also unremarkable. (Figure 2) CT scans of the maxillofacial region showed a multilocular radiolucent lesion about 40 x 20 mm in diameter and caused bone expansion. (Figure 3) After clinical and radiological examination, a biopsy was done with ameloblastoma, odontogenic mixoma or CEOT prediagnosis. The histopathologic examination showed epithelial polyhedral cell islands with pleomorphism and prominent intercellular bridges. (Figure 4a) Concentric lamellar calcifications which are characteristic for this lesion were seen among epithelial polyhedral cell islands. (Figure 4b) The patient was operated under local anesthesia. (Ultracain D-S Forte, Aventis, Turkey) The lesion was curetted aggressively with a clear margin. (Figure 5) Mandibular body contours could

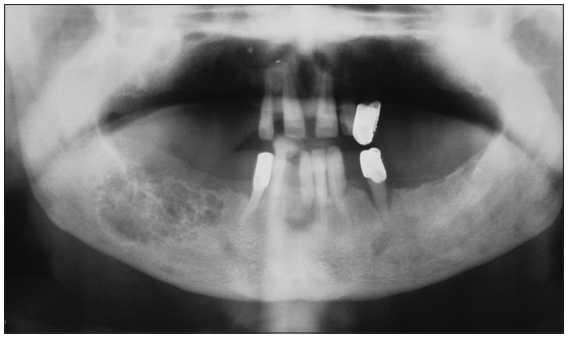

Figure 1. Preoperative panoramic radiograph.

Figure 2. Intraoral appearance of the mass.

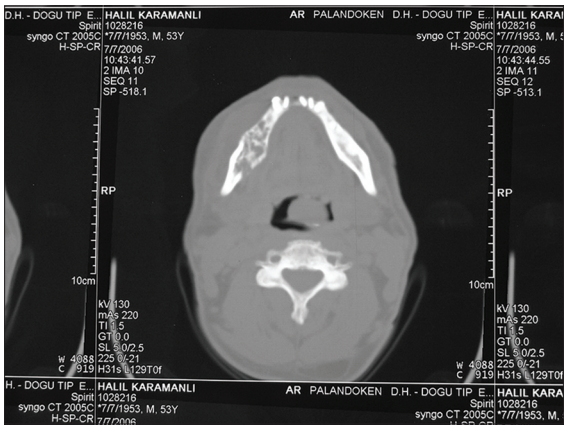

Figure 3. CT scans of the maxillofacial region.

be saved with this treatment modality. Postoperative healing was uneventful, and panoramic radiograph showed satisfactory bone healing at sixth month. (Figure 6) The patient still has been followed up, and no recurrence was observed.

DISCUSSION

CEOT is a rare benign odontogenic tumor, described in 1955 by Pindborg and also known as Pindborg tumor. This lesion represents about 0.4-3 % of all odontogenic tumors.[4] The intraosseous CEOT most often presents as a painless mass with slow growth. The most frequent presenting symptom of CEOT is swelling or expansion (72%); 22% of cases are asymptomatic and 13% complained of pain or discomfort.[8] In our case, slight swelling was the only finding of the tumor. There was no complaint of pain in our case.

Most of the authors agree that intraosseous CEOT is usually located in the premolar and molar regions, with a mandibular to maxillary ratio of 2:1 or 3:1.[1-5] The majority of CEOTs are associated with impacted or unerupted teeth.[5] The age of the patients affected by this tumor ranges from 8 to 92 years, with a mean age of 40.[4-6] In this case report, lesion in 53-year-old male patient was not associated with an impacted or unerupted tooth.

Radiographically, according to a study by Kaplan et.al.[8] 58% of CEOTs are unilocular, 27% multilocular, and 15% nonloculated.

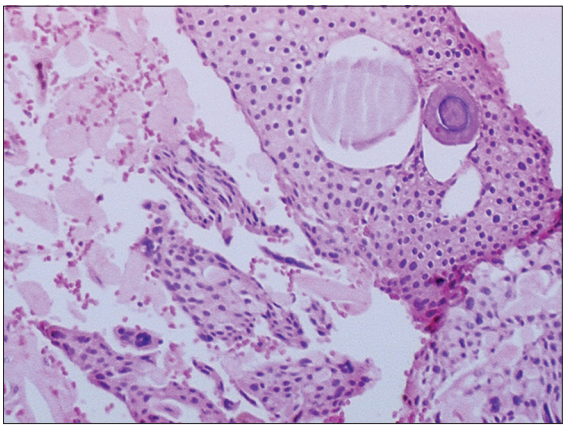

Figure 4a. The histopathologic image of the mass.

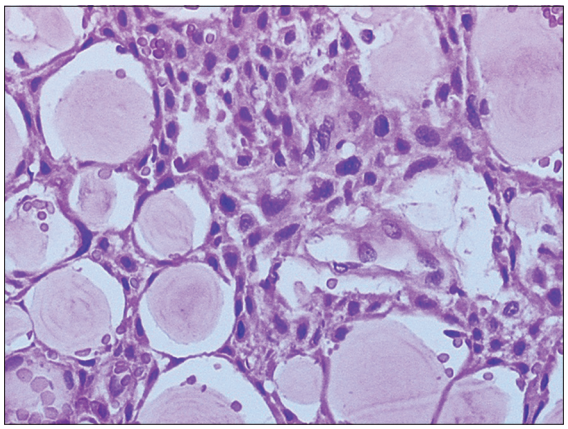

Figure 4b. Histopathological image showing concentric lamellar calcifications which are haracteristic for this lesion.

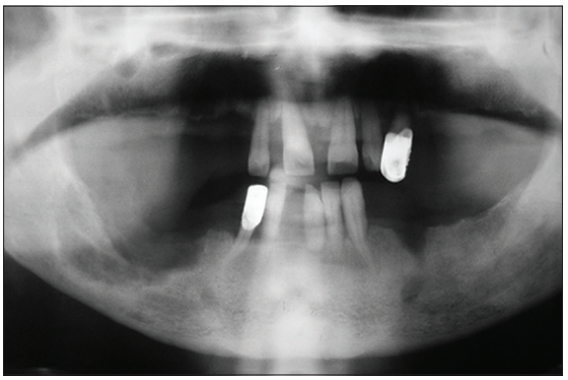

Figure 5. The mass curetted from the mandible posterior region.

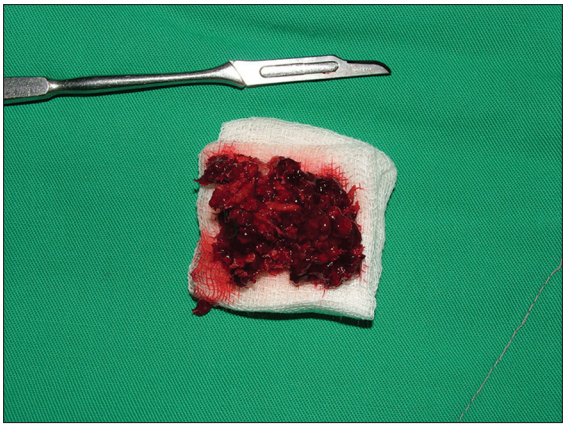

Figure 6. Panoramic radiograph showed satisfactory bone healing at sixth month.

The periphery of the tumor may demonstrate a fine sclerotic rim, and internal aspect frequently contains mineralized structures that appear as amorphous radiopacities of varying sizes. The tumor in this case was seen mainly as multilocular radiolucencies with amorphous opacification and a sclerotic rim. The bigger lesions can be multilobulated, immitating an ameloblastoma, mainly if we do not observe intramural calcifications.[7]

The minority of CEOTs that presents as a radiolucent lesion resemble the presentation of other radiolucent jaw lesions, such as dentigerous cyst, odontogenic ceratocyst, and ameloblastoma or odontogenic myxoma. The more common presentation of the CEOT (mixed radiolucent-radiopaque lesion) will suggest a calcifying odontogenic cyst, ameloblastic fibroodontoma, ossifying fibroma, osteoblastoma or even osteosarcoma if the lesion shows diffuse margins.[1,2,4]

This tumor still has an unpredictable clinical behavior, and can force an aggressive surgical treatment when the local behavior is not benign, or we have a recurrent lesion.[7] Methods of treatment have ranged from simple enucleation or curettage to hemimandibulectomy or hemimaxillectomy.[10] Some authors consider however, that conservative surgery is treatment of choice.[4] In view of the biological behavior of the CEOT, mutilating procedures, such as wide resection or hemiresection of the mandible, seem unwarranted. Enucleation with a margin of macroscopic normal tissue is, therefore, the recommended treatment for lesions involving the mandible. CEOT of the maxilla, however, should be treated more aggressively, as maxillary tumors, in general, tend to grow more rapidly than their mandibular counterpart and do not usually remain well confined.[4,5] Treatment should, however, be individualized for each lesion because the radiography and histologic features may differ from one lesion to another.[4]

The prognosis of the CEOT is good with infrequent recurrence.[1,4,11] Although malignant behavior is extremely rare, five years follow-up of the operated patients should be recommended to assess the healing for this tumor.

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How to cite this article: M. Selim Yavuz, M. Cemil Büyükkurt, M. Melih Ömezli, Sare Şipal. Calcifying epithelial odontogenic tumor: a case report. Cumhuriyet Dent J 2014;17(Supp 1):26-30.