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A Rare Maxillary Case of Calcified Epithelial Odontogenic (Pindborg) Tumor in a Young Patient

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Abstract

Aim The calcified epithelial odontogenic tumor (CEOT) is a rare, benign lesion, accounting for approximately 1% of odontogenic tumors. The primary treatment involves surgical excision, with a documented recurrence rate of 14%. This case report aims to contribute to the literature by presenting the radiological and histopathological findings, along with the surgical management, of a CEOT located in an unusual area in a female patient under 20 years old.

Case Report An 18-year-old female patient was referred to our clinic due to an incidental lesion located between the left maxillary premolar teeth. A delayed positive response was noted in the cold vitality test performed on asymptomatic, caries-free premolars. Cone beam computed tomography (CBCT) revealed a radiolucent lesion between the premolars, displacing the root of tooth 24 buccally and expanding the palatal cortex, with central radiopacity. Under local anesthesia, a palatal mucoperiosteal flap was raised, and a window was created in the palatal cortex using a surgical bur. The lesion was then enucleated with a curette, and the flap was sutured. Histopathological analysis showed polyhedral tumor cells among large and small calcification islands, confirming a diagnosis of CEOT.

Discussion Although treatment options may vary based on CEOT location and size, enucleation remains the most common approach.

Conclusion Given the high recurrence rate, long-term follow-up is essential. The patient has been followed up every six months, with no recurrence observed over a two-year period.

Keywords Calcified epithelial odontogenic tumor, Enucleation, Liesegang rings, Maxilla, Pindborg tumor

Introduction

A calcified epithelial odontogenic tumor (CEOT) is a benign, rare tumor that accounts for approximately 1% of odontogenic tumors (1). It is known as the "Pindborg tumor" because it was described by Pindborg in 1955 (2). CEOT is generally seen in individuals between the 2nd and 6th decades, and the number of cases reported under the age of 20 is quite low. The tumor is most commonly seen in the mandibular premolar and molar regions and is rarely seen in the maxilla. These tumors are generally seen to be associated with unerupted teeth (2, 3).

The aim of this case report is to contribute to the literature by presenting the radiological and histopathological findings and surgical treatment of CEOT, which was detected in the maxilla premolar region with a rare localization in a female patient under 20 years of age.

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Case Report

An 18-year-old female patient was referred to Istanbul University, Department of Oral and Maxillofacial Surgery after a lesion was observed between the roots of teeth 24 and 25 on orthopantomography (Figure 1). There were no symptoms, and no caries were seen on both premolars (Figure 2). However, a delayed positive response was obtained on the cold sensitivity test. A lesion resembling an inverted water drop was detected between teeth 24 and 25 on cone beam computed tomography (CBCT). This well-defined lesion pushed the root of tooth 24 towards the buccal side, caused expansion of the palatal cortex and contained radiopacity in its internal structure (Figure 3).

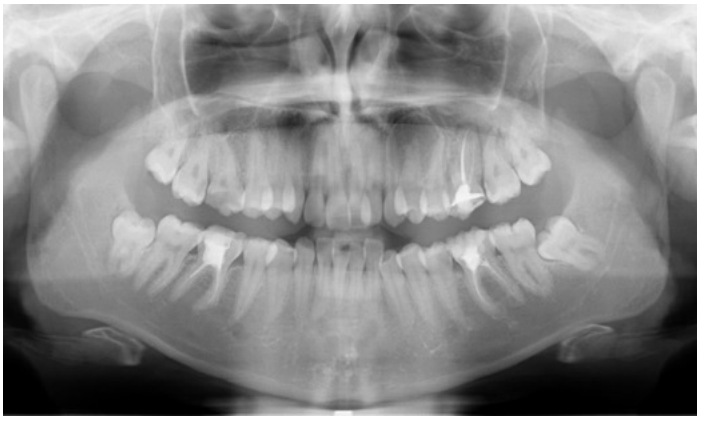

Figure 1: Orthopantomography showing the lesion

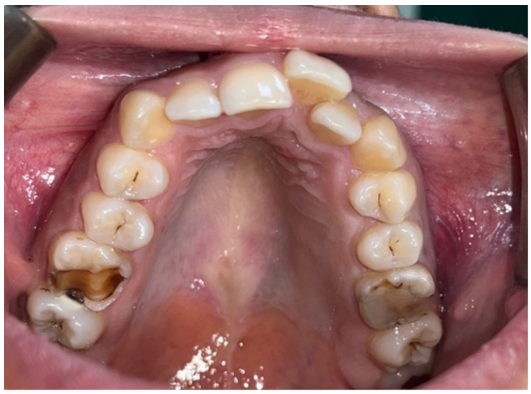

Figure 2: Preoperative intraoral radiograph

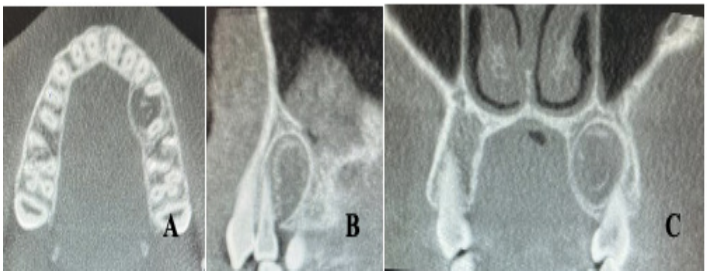

Figure 3: CBCT images A) Widening of the palatal cortex without disturbing the cortical border in the axial section. B) Inverted water drop shape with regular borders in sagittal section. C) Radiopaque foci in the internal structure in the coronal view.

The patient underwent surgery under local anesthesia in appropriate conditions. A mucoperiosteal flap was raised through a palatal sulcular incision. The enucleation of the lesion was performed with a curette through a window opened in the palatal cortex using a handpiece and a round bur. No luxation was observed in the teeth after enucleation. Peripheral bone curettage was carried out, and the operation area was cleaned by irrigation with isotonic saline solutions (Figure 4). The flap was primarily closed by vertical matrix suturing with 3/0 silk (Figure 5). The radiopacity observed on CBCT was confirmed to be calcifications in the lesion. Therefore, the lesion's preliminary diagnosis was suggested by CEOT. The tissue sample was examined histopathologically. In the histopathological examination, polyhedral tumor cells were identified among large and small calcification islands. The diagnosis was confirmed as CEOT (Pindborg tumor) (Figure 6).

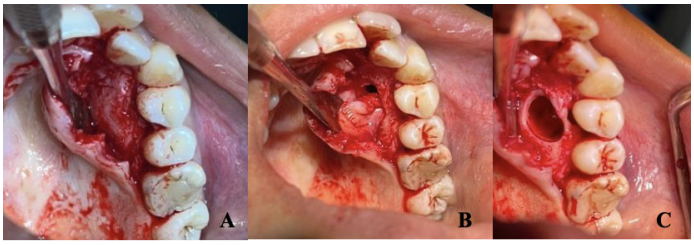

Figure 4: Images during surgery A) Mucoperiosteal flap removal with sulcus incision in the palatal region. B) Enucleation of the lesion using a curette through the window opened in the palatal cortex. C) After peripheral bone curette

At the follow-up visit one week later, it was observed that the wound had healed without any complications, and the sutures were removed. The patient has been under our follow-up for two years, during which no recurrence has been detected clinically or radiologically (Figure 7).

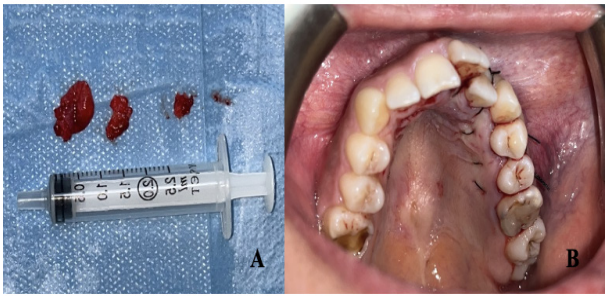

Figure 5: A) The removed lesion and calcified tissue B) The flap was closed primarily with vertical matrix sutures.

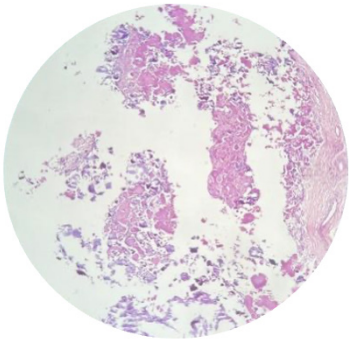

Figure 6: CEOT showing sheets of polyhedral epithelial cells with evidence of calcified areas. (Hematoxylin-eosin stain; original magnification x100)

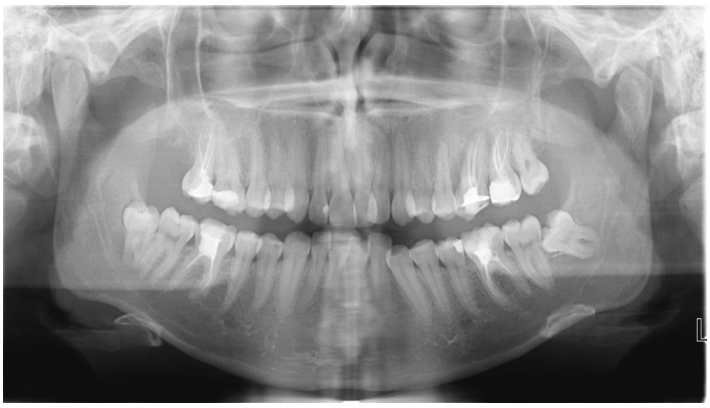

Figure 7: Control orthopantomography

Discussion

The World Health Organization (WHO) classified CEOT as a benign, epithelial odontogenic tumor in 2017 (1). It is a rare neoplasm, accounting for 1% of all odontogenic tumors (4). 90% of CEOT cases are intraosseous and are often seen in the mandible. It has been reported that there is no significant difference between men and women in gender distribution (5).

These tumors are generally slow-growing, painless lesions and have been associated with an impacted tooth in 58% of cases

(6, 7). The case explained in this article was discovered incidentally and is not related to an impacted tooth. It displaced tooth roots 24 and 25 in the lesion area. According to the literature, the radiographic features of these tumors are defined as regularly circumscribed, unilocular or multilocular lesions, and radiopacities characteristically seen in the internal structure (4). In the radiological images in this case, radiopacities were observed in the internal structure of the lesion and are compatible with the literature. CEOT was considered a preliminary diagnosis.

Histopathologically, CEOT is characterized by the presence of varying-sized calcifications, forming concentric laminations known as Liesegang rings (6). Liesegang rings were observed in the histopathological examination of the case explained in this article. Although the treatment of CEOT varies depending on its location and size, the generally preferred method is enucleation (8, 9). There are also cases where the impacted tooth was maintained after enucleation (10). In cases of CEOT, the prognosis is generally good, but recurrence may occur in 14%. Therefore, it is very important to keep such cases under regular follow-up (11).

Conclusion

Although there are different treatment options depending on the location and size of the CEOT, the most commonly used treatment method is enucleation. Since the relapse rate is high, long-term follow-up with patients at short intervals is recommended. The patient is followed up every 6 months, and no recurrence has been observed over a 2-year period.

Declarations

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

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

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