

# Calcifying Epithelial Odontogenic Tumor: Report of a Case

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## KEY WORDS

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## ABSTRACT

The calcifying epithelial odontogenic tumor (CEOT), also known as the Pindborg tumor, is a benign and rare odontogenic tumor accounting for 0.4-3 percent of all odontogenic tumors. Few reports of the incidence of this tumor exist in Iran. The most frequent location is the mandibular premolar and molar area; less frequently the lesion is found in the maxilla. In clinical features, a painless slow-growing swelling will be seen. Microscopically large sheets of epithelial cells in few fibrous stroma, large areas of amorphous, eosinophilic hyalinized (amyloid-like) material and calcification in concentric rings formation within the amyloid-like material will be detected. The authors report a peculiar case of Pindborg tumor characterized by a rare localization of the lesion extending from anterior to the posterior of the upper jaw and by the young age of the patient.

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## Introduction

The calcifying epithelial odontogenic tumor (CEO-T) was first described as an entity by Pindborg in 1956 [1-6]. This tumor is a rare benign neoplasm which comprises about 0.4-3% of all the intra-osseous odontogenic tumors. Although the tumor may show considerable radiographic variation, the most common feature of the lesion is pericoronal lucency with diffuses opaque foci within the lucent area.

The mean age of the occurrence at the time of diagnosis is 40-41 years and there is no significant difference in the occurrence between the sexes [7]. Most cases involve the posterior mandible; there have been few reported maxillary cases [8]. CEOT is a painless slow-growing tumor which causes jaw expansion [9].

Microscopically, CEOT shows sheets of polyhedral epithelial cells in few fibrous stroma. The nuclei show considerable variation, and giant nuclei and nuclear pleomorphism might be seen. In this tumor, the foci of amorphous, eosinophilic, hyalineized (amyloid-like) extracellular material and calcification in the form of concentric rings within this amyloid-like material are often present.

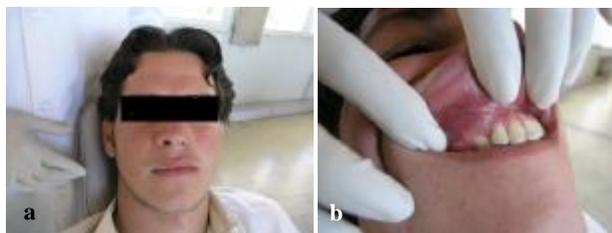
According to the incidence of the lesion in the molar area of the lower jaw and with respect to the resemblance of the radiographic features with ameloblastoma, CEOT can be mistaken with ameloblastoma. Histopathologic features of CEOT may cause pathologists to diagnose squamous cell carcinoma instead of CEOT [10].

In this article the authors report a peculiar case

of Pindborg tumor characterized by a rare localization of the lesion (extending from anterior to posterior) and by the young age of the patient.

**Case Report**

The patient was a 21 year old man who referred to the Oral Medicine Department of Mashhad Dental School with swelling in the right side of the face near the ala of the nose (Figure 1-a). The nasolabial fold was eliminated by the swelling. Intra-oral examination showed a swelling with a hard consistency extending from the distal aspect of upper right central incisor up to the distal portion of the right maxillary first premolar (Figure 1-b). In clinical palatal examination, no abnormal feature was seen.

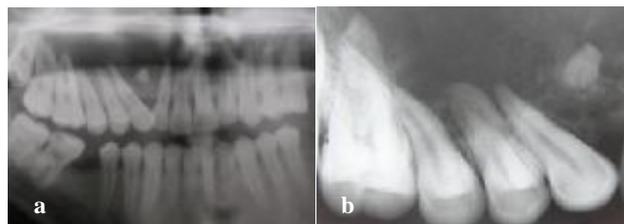


**Figure 1a** Swelling in right side of face near the ala of the nose **b** Intra oral exam showed a swelling with a hard consistency

Few drops of blood were obtained by aspiration of the lesion. The history of the lesion revealed a slow-growing, smaller sized swelling at the beginning reaching to the present size in twenty seven months.

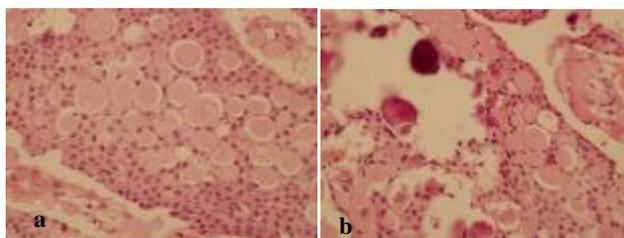
In radiographic evaluation, the maxillary sinuses of both sides were normal. A mixed radiolucent-radiopaque pattern with an opaque mass in the center with an indefinite border was seen between the apices of the upper right lateral incisor and canine causing displacement of the roots of these two teeth and buccal extension up to the upper right first molar tooth (Figures 2-a, 2-b). A biopsy with primitive clinical as well as radiographic diagnosis of calcifying odontogenic cyst (Gorlin cyst) was performed. Microscopically, the polyhedral epithelial cells and

areas of calcification were seen. Primitive pathological diagnosis was CEOT. Surgery was undertaken including a marginal portion of apparently healthy bone and histopathologic as well as immunohistochemical evaluation was performed on the lesion.



**Figure 2a** Panoramic view showed a mixed radiolucent-radiopaque lesion with an indefinite border **b** Periapical view showed an opaque mass in the center

In microscopic view, the tumor was covered by keratinized squamous epithelium. The subepithelial tissue showed sheets of polyhedral cells with clear intercellular bridges. Nuclear pleomorphism was observed in some areas. In some portions, separation of the sheets of cells by a cellular amyloid-like material and calcification was seen (Figures 3-a, 3-b). Immunohistochemical method was positive for pancytokeratin (CK) and epithelial membrane antigen (EMA). Final diagnosis was CEOT. The patient has been under follow-up for the last two years and no recurrence has been seen.



**Figure 3a** Microscopic exam showed sheets of polyhedral epithelial cells with amyloid-like material **b** Foci of calcification between polyhedral epithelial cells

**Discussion**

CEOT was first described by Pindborg in 1956 and Shafer and colleagues called it Pindborg tumor in

1963 [4]. The most common age of occurrence is the fourth and fifth decades of life [1-2]. The mandible is affected twice as often as the maxilla and the premolar- molar area is the most common location [4]. This report adds to the small number of Pindborg's tumor cases located in the maxilla, since over 2/3 of the CEOT cases have been described in the posterior portion of the mandible [8].

Two marked distinctive features were identified in this case: rare localization of the lesion and the young age of the patient. Nascimento et al reported a Pindborg tumor of the maxilla with a significant amyloid-like deposit, and sparse mineral deposits with inconspicuous Liesegang's rings [11]. In our case both components were significant.

Bridle et al have reported an unusual case of such a tumor in the maxilla presenting with abnormal eye signs [12] but in the present case there was no sign of maxillary or eye involvement. Treatment of CEOT consists of surgical removal, which includes a marginal portion of the apparently healthy bone. A minimum of 5-year observation period is suggested. Maxillary CEOT cases require more aggressive surgery, since these tumors tend to grow more rapidly and are not circumscribed [11]. A 14% local recurrence rate has been reported; the prognosis is considered as favorable [13].

In this case surgery was undertaken, including a marginal portion of apparently healthy bone and after two years there was no signs of relapse.

### Conclusion

Although the incidence of this tumor in the anterior segment of the upper jaw is very rare, it may happen as in our case.

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