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Case Report

# Peripheral calcifying epithelial odontogenic tumor – Case report

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ABSTRACT

The calcifying epithelial odontogenic tumor (CEOT), Pindborg tumor is a benign, slow growing, but locally invasive neoplasm. It is known to have a common intraosseous variant and a very rare extraosseous variant. We report an unusual case of an extraosseous variant of CEOT of unusual large size and maxillary anterior location, the treatment was planned considering the clinical, radiological and histological features. Though peripheral types are less aggressive and had no recurrence, in our case regular follow up is required considering the aggressiveness of the lesion and its proximity to important adjacent structures.



## 1. Introduction

The calcifying epithelial odontogenic tumor (CEOT), Pindborg tumor was first recognized by Jens Jorgen Pindborg in 1955.[1,2] This benign, slow growing, but locally invasive neoplasm accounts for 0.4–3% of all odontogenic tumors.[3] It is known to have a common intraosseous variant (94%) and a very rare extraosseous variant (6%).[3] We report an unusual case of an extraosseous variant of CEOT in the maxillary anterior region and a brief review of literature.

## 2. Case report

A 41 year old female patient reported to us with a progressively enlarging asymptomatic swelling in the maxillary anterior region which was of four years duration [Fig. 1a]. Examination revealed a solitary, benign, sessile, non pulsatile expansile mass involving the anterior maxilla extending up to the canine region on either side. The patient had lost her teeth due to gradual expansion of the mass. OPG and CT scan revealed an expansile intensely enhancing soft tissue lesion with intralesional calcification [Figs. 1b, 2a and 2b]. Patient was otherwise healthy and

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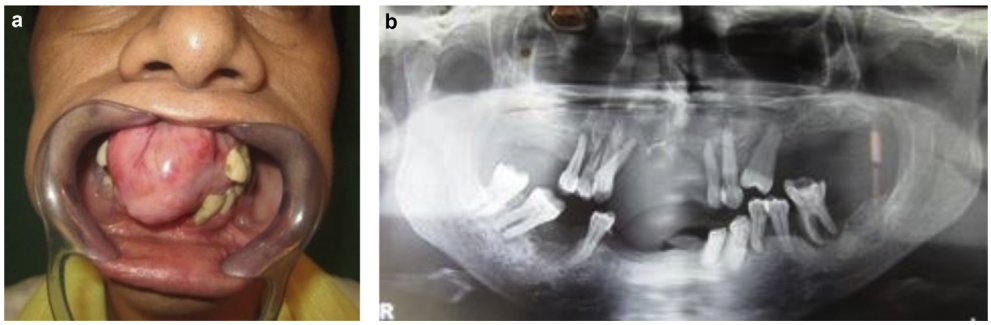


**Fig. 1 – a. Preoperative photograph, b. OPG showing expansile lesion in the anterior maxilla.**

all routine investigations were within normal limits. Incisional biopsy was reported as CEOT. Patient was treated with anterior maxillectomy and lining of defect was done using skin graft. Complete mouth rehabilitation was then carried out with implant supported prosthesis.

Gross specimen consisted of hard palate, and four teeth weighing 80 g and measuring 8 × 6 × 4 cm [Fig. 3a]. Specimen had nodular growth with bosselated surface which was 4 × 4 × 3.5 cm and pale yellow in appearance. Cut section was fibrous white. Histopathological examination revealed mucosa overlying unencapsulated tumor which was composed of nodules of polyhedral cells having eosinophilic cytoplasm, intercellular bridges and large irregular hyperchromatic nuclei surrounded by myxoid to hyalinisedstroma [Figs. 3b and 4a]. Congo red stain was positive [Fig. 4b] and the amyloid like material had undergone Leisgang type of calcification. Foreign body giant cells along with plasma cell infiltration were seen. Bone was eroded in some areas. The final Histopathological report confirmed the initial diagnosis of CEOT. Patient has been on regular follow up for two years without any signs of recurrence [Fig. 5].

## 3. Discussion

Pindborg tumor was previously described in literature as adamantoblastoma (Smith, 1977), adenoid adamantoblastoma (Thoma and Goldman, 1946), ameloblastoma of unusual type with calcification (Ivy, 1948), malignant odontoma (Wunderer, 1953), and cystic complex odontoma (Stoopack, 1957).

It accounts for less than 1% of all odontogenic tumors.[4] Intraosseous tumors (tumors within the bone) are more common (94%) than extraosseus variants (6%).[5] This tumor typically occurs in the fourth to fifth decades (mean age 40 years) and affects both sexes equally. Mandible is affected twice as much as maxilla (2:1)[5] with more than two third of central lesions occurring in premolar and molar region. Clinically it presents as slowly enlarging painless mass concurrent with an impacted tooth causing bone expansion. This lesion is rarely associated with paresthesia. Large lesions in the maxilla have been reported to cause proptosis, epistaxis and nasal air way obstruction.[5] However the peripheral variant is commonly found in the anterior maxilla where it appears as a soft tissue swelling.[6] It occurs during third to fourth decades of life and affects women twice more often than men.[3] Maxillary tumors may present with facial alteration .In this present case, mass in the anterior maxillary region resulted in facial alteration.

The initial consensus regarding the pathogenesis of CEOT was attributed to pindborg in 1955. He stated that the CEOT was indeed of odontogenic origin reduced organ enamel-related due to previously case had been associated to unerupted teeth.[1] However, according to Philipsen et al with the reports of central cases not presenting unerupted tooth and gingival variants, other sources of origin were debated. The

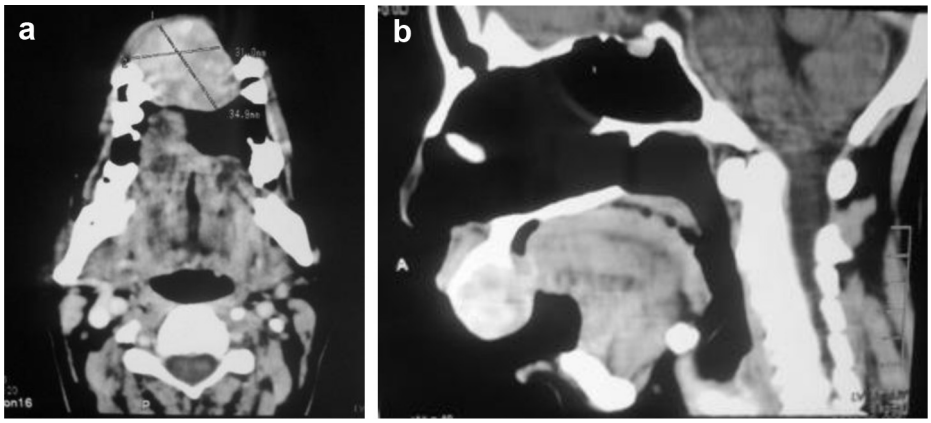


**Fig. 2 – a and b. CT axial and sagittal section showing expansile lesion with intralesional calcification.**