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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.

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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 $8 \times 6 \times 4$ cm [Fig. 3a]. Specimen had nodular growth with bosselated surface which was $4 \times 4 \times 3.5$ 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 hyalinized stroma [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 extraosseous 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.

Fig. 3 – a. Gross specimen, b. Photomicrograph – H&E stain 40×.

Fig. 4 – a. 10× view showing stratified squamous epithelium overlying nodules of eosinophilic epithelial cells, surrounded by hyalinised stroma, b. 40× view showing Congo red stain for amyloid–positive.

soft tissue location of CEOTs strongly suggests that these tumors may arise from rests of dental lamina or from basal cells of the oral epithelium. After disintegration of the dental lamina complex, numerous epithelial remnants (rests of serres) persist in the jaw bones and suprapariosteally in the gingiva when odontogenesis is completed. Furthermore, focal proliferation of the basal layer of the gingival epithelium has also been proposed as a possible origin.^{3,7}

On radiographs, it appears as a unilocular or multilocular radiolucency and is known for sometimes having small

radiopacities within it. In those instances, it is described as having a “driven-snow” appearance.⁵ Kaplan et al⁵ reported root resorption in 4% of 67 cases and emphasized that this feature may aid in the differential diagnosis between CEOT and solid ameloblastoma.

Microscopically, it is characterized by squamous epithelial cells calcifying masses, and homogeneous acellular material admixed with the tumor epithelium and stroma that have been identified as amyloid. The underlying nature of the amyloid-material is still unresolved.^{3,4} It also reveals prominent intercellular bridges and nuclear changes such as pleomorphism, hyperchromatism and prominent nucleoli. The mitotic figures are rare. Spread throughout the epithelium and connective tissue are spherical amorphous calcifications. Several histologic variants of CEOT have been reported, such as clear cell, which is more aggressive with a higher recurrence rate (22%). CEOT has also been found to be associated or combined with other odontogenic lesions such as adenomatoidodontogenic tumor and dentigerous cyst, and it can be found as an intramural growth in the dental follicle,⁸ pigmented Langerhans cell containing,⁹ bone and cementum forming, and noncalcifying. There is one case reported in the literature of a 15-year old white male with a true cystic variant of CEOT.¹⁰

The treatment for CEOT ranges from simple enucleation or curettage to radical and extensive resection such as hemimandibulectomy or hemimaxillectomy. The method of treatment will depend on the size, anatomic location of the

Fig. 5 – Post operative photograph.

tumor and the amount of bone destruction.^{3,5,11} Maxillary lesions should be treated more aggressively because they grow faster and because of the proximity to important structures. An example of the locally aggressive behavior of CEOT was reported in a recent case report by Bouckaert et al¹¹ in which a case of advanced CEOT in the maxilla invaded the orbit, maxillary sinus, and ethmoid sinus and extended to the cranium, which caused compression and abscess in the brain. There were no reported recurrences in the literature in the peripheral type indicating less aggressive behavior and good prognosis. In our case patient was treated with anterior maxillectomy and lining of defect with skin graft.

Franklin and Pindborg reported a recurrence rate of 14%.⁹ It is considered to have a recurrence rate much lower than ameloblastoma.⁹ Malignant transformation is rare, and only three cases have been reported in the literature.⁴ Krolls and Pindborg comparing CEOT with ameloblastoma, postulated that the presence of two products within the CEOT—that is, calcifications and amorphous substance—probably indicates greater tumor differentiation and accounts for the low CEOT recurrence rate. Franklin and Pindborg have correlated the less aggressive nature of the lesion to the presence of amyloid tissue and calcification.⁹

To summarize we have described a case of peripheral 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.

Conflicts of interest

All authors have none to declare.

REFERENCES

1. Pindborg JJ. The calcifying epithelial odontogenic tumor: review of literature and report of an extraosseous case. *Acta Odont Scand.* 1966;24:419–430.
2. Rajendran R. Cysts and tumors of odontogenic origin. In: Rajendran R, Sivapathasundaram B, eds. *Shafer's Textbook of Oral Pathology*. 6th ed. Noida: Elsevier Publishers; 2009:279–281.
3. Philipsen HP, Reichart PA. Calcifying epithelial odontogenic tumour: biological profile based on 181 cases from the literature. *Oral Oncol.* 2000;36:17–26.
4. Cheng YS, Wright JM, Walstad WR, Finn MD. Calcifying epithelial odontogenic tumor showing microscopic features of potential malignant behavior. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2002;93:287–295.
5. Kaplan I, Buchner A, Calderon S, Kaffe I. Radiological and clinical features of calcifying epithelial odontogenic tumor. *Dentomaxillofac Radiol.* 2001;30:22–28.
6. Houston GD, Fowler CB. Extraosseous calcifying epithelial odontogenic tumor: report of two cases and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1997;83:577–583.
7. Soares ECS, Costa FWG, Esses DFS, Filho EPA, Alves APNN. Gingival calcifying epithelial odontogenic tumor: case report and review of the literature comprising a period of 44 years. *J Clin Exp Dent.* 2011;3(5):e491–e496.
8. Ficarra G, Hansen LS, Stiesmeyer EH. Intramural calcifying epithelial odontogenic tumor. *Int J Oral Maxillofac Surg.* 1987;16:217–221.
9. Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumor. A review and analysis of 113 cases. *Oral Surg Oral Med Oral Pathol.* 1976;42:753–765.
10. Gopalakrishnan R, Simonton S, Rohrer MD, Koutlas IG. Cystic variant of calcifying epithelial odontogenic tumor. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2006;102:773–777.
11. Bouckaert MM, Raubenheimer EJ, Jacobs FJ. Calcifying epithelial odontogenic tumor with intracranial extension: report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2000;90:656–662.