

Case Report

Recurrent CEOT of the maxilla

Geetha Kamath¹, Reji Abraham²

¹Departments of Oral Medicine and Radiology, and ²Orthodontics, Sri Hasanamba Dental College and Hospital, Hassan, Karnataka, India

Received: September 2011
Accepted: December 2011

Address for correspondence:
Dr. Reji Abraham,
Department of
Orthodontics, Sri
Hasanamba Dental College
and Hospital, Vidyanagar,
Hassan 573201, Karnataka,
India.
E-mail: rejiabm@gmail.com

ABSTRACT

Calcifying epithelial odontogenic tumor (CEOT) is a rare benign, but locally infiltrating odontogenic neoplasm. It accounts for less than 1% of all odontogenic tumors. This is a case report of recurrent CEOT in the maxilla. A 35-year-old patient reported after three years of surgical excision of the lesion, with a recurrence. It is of particular concern because of its anatomic location in the maxilla. Maxillary tumors tend to be more aggressive and rapidly spreading and may involve the surrounding vital structures. Adequate resection of the lesion with disease-free surgical margins and long-term follow-up is recommended.

Key Words: Calcifying epithelial odontogenic tumor, maxillary tumors, odontogenic tumor

INTRODUCTION

The calcifying epithelial odontogenic tumor (Pindborgs tumor) is a benign neoplasm of odontogenic origin.^[1] It is a rare tumor accounting for less than 1% of all odontogenic tumors.^[2] It is a benign, though occasionally locally invasive, slow-growing neoplasm. They are localized generally in posterior part of mandible and rarely occur in the maxilla.^[1,2] This article reports a case of recurrent CEOT of the maxilla in a 35 year old male patient.

CASE REPORT

A male patient aged 35 years reported with a painless swelling of eight months duration in the left upper jaw in 2005. On examination, it was hard in consistency with expansion of the cortical plates [Figures 1 and 2]. The first premolar tooth was missing with no history of previous dental extractions. The panoramic radiograph showed a diffuse honeycomb type of radiolucency extending from premolar to

third molar region with few radiopacities [Figures 3 and 4]. The lesion was associated with an impacted tooth, which resembled a premolar and was displaced posterosuperiorly. There was no evidence of root resorption of the adjacent teeth. The lesion was surgically excised and histopathologically diagnosed as calcifying epithelial odontogenic tumor (CEOT).

Microscopic examination of the tissue revealed sheets and strands of polyhedral epithelial cells with hyperchromatic nuclei, mild to moderate pleomorphism and prominent intercellular bridges. Eosinophilic hyaline deposits with calcifications were found within and between sheets of epithelial cells. The patient was not regular for the follow up and reported again in 2008, three years after excision. The patient had noticed a growth in the same region five months back and experienced no discomfort. A computed tomography (CT) scan showed a large, expansile, radiolucent lesion with multiple areas of calcification which completely obliterated the left maxillary antrum [Figures 4 and 5]. The scan showed the tumor extending and involving the lateral nasal wall, orbital floor and the medial pterygoid plate [Figures 6 and 7]. An incisional biopsy confirmed the lesion to be a recurrence of CEOT. No atypias or mitoses were found. The lesion was excised with wide surgical margins and the patient is under observation for the past three years without any signs of recurrence.

Access this article online



Website: www.drj.ir



Figure 1: Swelling of left middle third of face with obliteration of the nasolabial fold at first presentation



Figure 2: Intraoral picture showing expansion of cortical plates in left maxilla



Figure 3: Portion of a panoramic radiograph showing a honeycomb type of radiolucency associated with an impacted tooth

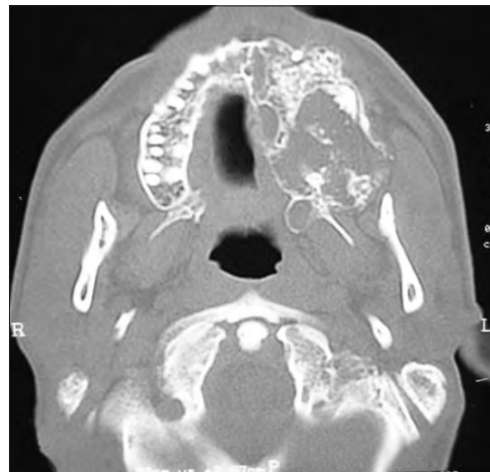


Figure 4: CT taken after recurrence clearly demonstrates the complete loss of architecture of the left maxilla



Figure 5: Axial CT demonstrates radiolucency with numerous radiopacities



Figure 6: Coronal section of CT showing obliteration of the left maxillary sinus

DISCUSSION

CEOT is an uncommon neoplasm accounting for less than 1% of all odontogenic tumors.^[1] This rare

tumor was first described as a separate pathologic entity by a Dutch pathologist Jens Jorgen Pindborg in 1955.^[2] Less than 200 cases have been reported in the literature, with most being reported in the mandible.^[3] CEOT has a peak prevalence in the fourth and fifth decades, with an equal sex distribution. It has a marked preference for the mandible and most tumors arise in the molar–premolar region. 52% of the tumors are associated with an impacted tooth, most often the first or second molars.^[4] Although Pindborg’s tumor is well described in the mandible, descriptions of lesions involving the maxilla are rare. This case features a lesion in the maxilla with an impacted maxillary first premolar tooth.

The typical clinical presentation of CEOT is a slowly enlarging mass that causes expansion of the affected site and is asymptomatic. When located in the maxilla, it may be associated with epistaxis, nasal stuffiness, proptosis and headache.^[5]

Radiographically, it has a variety of appearances; 58% of CEOTs are unilocular, 27% are multilocular, and 15% are nonoculated. The internal aspect frequently contains mineralized structures that appear as radiopacities.^[6] Radiographic features of CEOT may overlap with several other odontogenic or nonodontogenic lesions. CEOT is commonly associated with impacted teeth and can be confused with a dentigerous cyst which is also seen around an impacted tooth. However, the dentigerous cyst lacks mineralization within the lesion. In contrast to dentigerous cyst which is more frequently associated with third molars, CEOT is found around first and second molars. Ameloblastoma and odontogenic myxoma may also present as unilocular or multilocular radiolucencies, similar to CEOT. These two common odontogenic tumors rarely demonstrate radiographic evidence of radiopacities like CEOT. Further, odontogenic myxoma often has a “soap bubble” appearance with angular trabeculae within the lesion and ameloblastoma is usually associated with root resorption in 81% of the cases. The lesion that strongly resembles CEOT radiographically is the calcifying odontogenic cyst which presents as a mixed radiolucent radiopaque lesion and is associated with an impacted tooth.^[3,7] In this case that involved the maxilla, the differential diagnosis included ossifying fibroma, ameloblastic odontoma, and Gorlin’s cyst. Advanced imaging technique plays an important role in evaluating the extent of facial bones and skull involvement and has a crucial role in

planning the surgery.^[8] The CT of this tumor usually shows a well-defined mass with thinning of the cortical plates and contains scattered radiopaque foci. Magnetic resonance imaging (MRI) predominantly shows hyperintense T2-weighted and hypointense T1-weighted images.^[9]

The diagnosis of CEOT is based on histopathology. CEOTs are unencapsulated, infiltrating tumors. Epithelial cells appear polyhedral with prominent intercellular bridges having abundant eosinophilic, finely granular cytoplasm with nuclear pleomorphism and prominent nucleoli.^[7] Most of the cells are arranged in broad ramifying and anastomosing sheet-like masses with little intervening stroma. An eosinophilic homogenous material staining like amyloid is characteristic of this tumor with concentric calcified deposits, resembling psammoma bodies called “Liesegang rings.” Congo red staining with viewing under polarized light microscopy demonstrates areas of apple green birefringence. These areas depict positive staining of amyloid like substance and are highly characteristic of CEOT. Amyloid also stains positively for crystal violet and thioflavine T.

CEOT has a variable biologic behavior ranging from very mild to moderate invasiveness.^[7] The literature shows variations regarding radicality of the surgical treatment needed. There are very few evidence-based treatment recommendations because of the paucity of cases reported.^[10] Surgical procedures for treatment may include conservative enucleation, marginal resection or partial resection in larger infiltrating tumors.^[11] In their review of 113 cases,^[4] Franklin and Pindborg suggested that marginal resection with a rim of normal tissue is advisable. They advise against a radical surgical approach of wide resection such as hemimaxillectomy. Surgical decision making often depends on case parameters such as the anatomic location of the tumor, the size and duration, histopathologic findings, patient’s age, and consideration of reconstruction methods following surgical procedure.^[4,12] The appropriate treatment of CEOT requires surgical excision with disease-free margins. In the maxilla, the CEOT tends to grow more rapidly and may infiltrate the proximal vital structures, suggesting that more aggressive surgery is required in these specific cases.^[13]

In this case, CEOT was treated conservatively via enucleation. Recurrence of the lesion is presumed to be



Figure 7: Section showing involvement of the nasal cavity and inferior orbital wall

due to inadequate removal of neoplastic tissue, which is possible in our case, given the more conservative surgical approach. Local recurrence rates of 10–15% have been reported and malignant transformation is rare, with only three cases reported.^[4,11] CEOT has a much lower recurrence rate than ameloblastoma. A follow-up of minimum 5–10 years may be necessary because of the very slow growth rate of this tumor.^[4]

In conclusion, this report features a case of recurrent CEOT in an unusual location of maxilla. It emphasizes the rapid and unconfined growth of maxillary CEOT. Maxillary lesions probably need aggressive surgery as these tumors usually grow more rapidly than their mandibular counterparts and invade the surrounding vital structures. Treatment by surgical resection with accurate tumor-free margins is needed with periodic clinical and radiographic follow up.

REFERENCES

1. Pindborg JJ. Calcifying epithelial odontogenic tumour. *Acta Pathol Microbiol Scand* 1955;111:71.

2. Pindborg JJ. A calcifying epithelial odontogenic tumor. *Cancer* 1958;11:838-43.
3. Neville BW, Damm DD, Allen CM, Bouquot JE. *Oral and maxillofacial pathology*. 2nd ed. Philadelphia: Saunders; 2002. p. 623-5.
4. Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumor. A review and analysis of 113 cases. *Oral Surg Oral Med Oral Pathol* 1976;42:753-65.
5. Bouckaert MM, Raubenheimer EJ, Jacob 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;90:656-62.
6. Kaplan I, Buchner A, Calderon S, Kaffee I. Radiological and clinical features of calcifying epithelial odontogenic tumor. *Dentomaxillofac Radiol* 2001;30:22-8.
7. Marx RE, Stern D. *Oral and Maxillofacial Pathology*. Chicago, IL: Quintessence Publishing; 2003. p. 635-703.
8. Patino B, Fernandez-Alba J, Garcia-Rozado A, Martin R, Lopez-Cedruj JL, Sanroman B. Calcifying epithelial odontogenic (Pindborg) tumor: A series of 4 distinctive cases and a review of the literature. *J Oral Maxillofac Surg* 2005;63:1361-8.
9. Cross JJ, Pilkington RJ, Antoun NM, Adlams DM. Value of computer tomography and magnetic resonance imaging in the treatment of a calcifying epithelial odontogenic (Pindborg) tumor. *Br J Oral Maxillofac Surg* 2000;38:154-7.
10. Rapidis AD, Stavrianos SD, Andresakkis D, Lagoginnias G, Bertin PM. Calcifying Epithelial Odontogenic Tumor(CEOT) of the Mandible: Clinical therapeutic conference. *J Oral Maxillofac Surg* 2005;63:1337-47.
11. 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;93:287-95.
12. Bridle C, Visram K, Piper K, Ali N. Maxillary calcifying epithelial odontogenic (Pindborg) tumor presenting with abnormal eye signs: Case report and literature review. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;102:e12-5.
13. Lee CY, Mohammadi H, Mostofi R, Habibi A. Calcifying epithelial odontogenic tumor of the maxillary sinus. *J Oral Maxillofac Surg* 1992;50:1326-8.

How to cite this article: Kamath G, Abraham R. Recurrent CEOT of the maxilla. *Dent Res J* 2012;9:233-6.

Source of Support: Nil, **Conflict of Interest:** None declared.