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An Uncommon Clinical Presentation of a Calcifying Epithelial Odontogenic Tumor (Pindborg Tumor): Case Report and Review

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Authors' contributions

This work was carried out in collaboration between all authors. Author PT designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors HS and WMF managed the analyses of the study. Authors SS and HFAG managed the literature searches. Author ISFA contributed in review of literature. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

The calcifying epithelial odontogenic tumor (CEOT) is a benign tumor, accounting for 0.4-3% of all odontogenic tumors. It mostly occurs in the 4th to 6th decades of life with equal propensity for both the genders. In about 52% of cases, the tumor is found in association with an unerupted tooth, with lower jaw being more commonly involved. It is usually asymptomatic and often causes bone expansion similar to other odontogenic tumors; therefore, diagnosis depends upon the lesion's histologic features. We report a clinical presentation of CEOT not associated with unerupted teeth and occurring in the anterior mandible of a 16 years old male. The tumor was treated by surgical removal with no sign of recurrence after a period of 6 months. The surgery required complete removal including adequate margins of normal tissue in order to ensure complete removal.

Keywords: Pindborg tumor; calcifying odontogenic tumor; benign odontogenic tumor.

1. INTRODUCTION

The calcifying epithelial odontogenic tumor (CEOT) is a benign neoplasm of odontogenic origin accounting for 0.4-3% of all odontogenic tumors [1]. It was first described in 1955 by the Danish pathologist Jens Jorgen Pindborg [2].

The origin of the tumor is not fully understood. There are theories that suggest its origin is associated with the stratum intermedium of the dental organ and with dental lamina [3]. Clinically, it presents as a painless slow growing lesion, causing expansion of the cortex, and twice more frequently found in the mandible than in the maxilla. However it shows more aggressive behavior when it occurs in the maxilla, where signs/symptom may include epistaxis, proptosis, and a feeling of fullness over the maxillary sinus [4]. In approximately 52% of the cases, the tumor is associated with an impacted tooth [4]. There is a varied age range for its occurrence with equal involvement of both the genders [4,5]. Radiographically, it appears as mixed radiolucent-radiopaque mass with opacities of calcifications. The gradual growth of the tumor causes cortical expansion evident as thinning of the cortexes with fine trabeculae dividing the radiolucent mass into various compartments giving a honeycomb appearance [5,6]. The invasiveness of the tumor ranges from mild to moderate depending upon its biological behavior. Surgical intervention is the treatment of choice [6]. Here, as earlier stated, a better explanation is required. Basically the extent of surgery is based on the extent of tumor invasiveness. Well localized and demarcated tumors will require less extensive surgery than larger tumors that cause thinning of the corticies. The purpose of this paper is to describe a rare clinical presentation of a CEOT and briefly describe our treatment.

2. CASE REPORT

A 16 year-old male patient presented to our hospital with a chief complaint of a 6 month history of swelling in the anterior region of his lower jaw and discomfort during swallowing. The swelling had started spontaneously, with no history of discharge, and had been gradually increasing in size. There was no relevant medical or family history. On clinical examination, an extra oral diffuse swelling was seen in the symphysis region of the mandible measuring approximately 5 x 4 cm in size. On intraoral examination, the swelling extended horizontally from lower right canine to lower left first premolar and vertically from the alveolar margin to deep into the gingiva-buccal sulcus. There was no change in colour of the overlying mucosa; however, the occlusal surface of the swelling was ulcerated because of the continuous occlusal trauma (Fig. 1). On palpation, the swelling was firm, non-tender, with expansion of both the buccal and lingual cortexes. All the teeth associated with tumor were mal-aligned with grade 1 mobility. Pulp vitality was checked using an electric pulp tester showing delayed response in all involved teeth. CT scan (coronal and axial sections) and 3-D CT of the face revealed a well-defined unilocular radiolucency with radiopaque borders in the symphysis region of the mandible that extended from the right canine to the left first premolar (Fig. 2). There was evidence of root resorption and rotation of the teeth that were associated with tumor. Expansion of both the cortices along with perforation of the buccal cortex was also seen.

Based on the clinical and radiographic examination, provisional diagnosis of benign tumor to include: ameloblastoma, ossifying fibroma, odontogenic myxoma, adenomatoid odontogenic tumor or calcifying epithelial

odontogenic tumor was made. Aspiration of the lesion presented yielded a brown serous liquid. An incisional biopsy was and the specimen sent for histological study. The lesion was then diagnosed to be a calcifying epithelial odontogenic tumor (Fig. 3).

Surgical intervention was performed under general anesthesia. After the naso-tracheal intubation, a sulcular incision was made, and a full-thickness mucoperiosteal flap elevated to expose the tumor. A surgical enucleation was performed. The tumor mass was detached from the surrounding bone by blunt dissection and removed, followed by extraction of teeth involved in the tumor. Clinically the tumor was found to have a thick wall that surrounded dentin-like material and enamel that appeared to be compatible with a compound odontoma (Fig. 4). The closure was accomplished in a single layer with horizontal mattress sutures using 3-0 vicryl. The patient was kept on prescribed antibiotics and analgesics for 5 days and 0.12% chlorhexidine mouthwash twice a day for 2 weeks. The postoperative phase was uneventful and the patient was discharged after 1 week. No signs of recurrence were noted after I year follow-up (Fig. 5).

Fig. 1. Preoperative
(i) Front view, (ii) Lateral view, (iii) Intraoral view

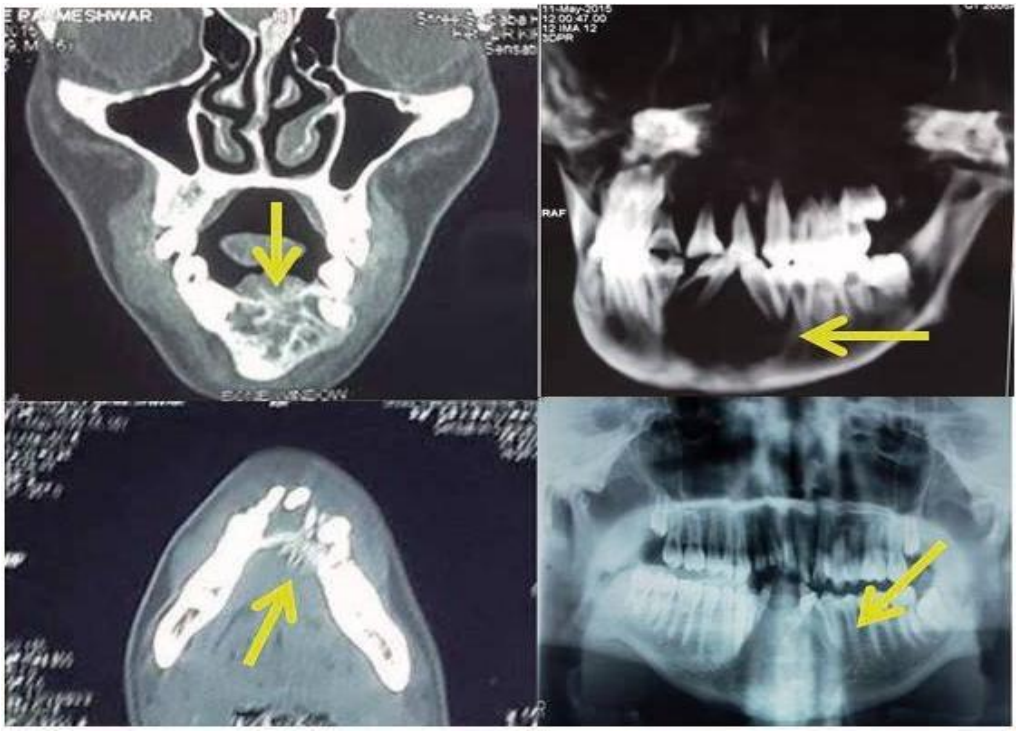

Fig. 2. Preoperative radiographs
A CT scan study of the mandible confirming the expansion of the buccal and lingual cortical plates along with perforation of the buccal & lingual cortex in the mandibular anterior region. Radiographic examination showed a unilocular radiolucent lesion with irregular border extending from lower right canine to left lower first premolar region

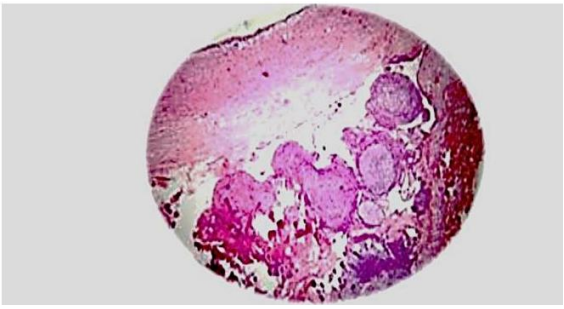

Fig. 3. Histological view of CEOT

Deep-stained calcifying materials and eosinophilic amyloids. The epithelial cells shaped like strands surround the calcifying materials. Area stained with (H & E staining x 40) showing Liesegang calcifications

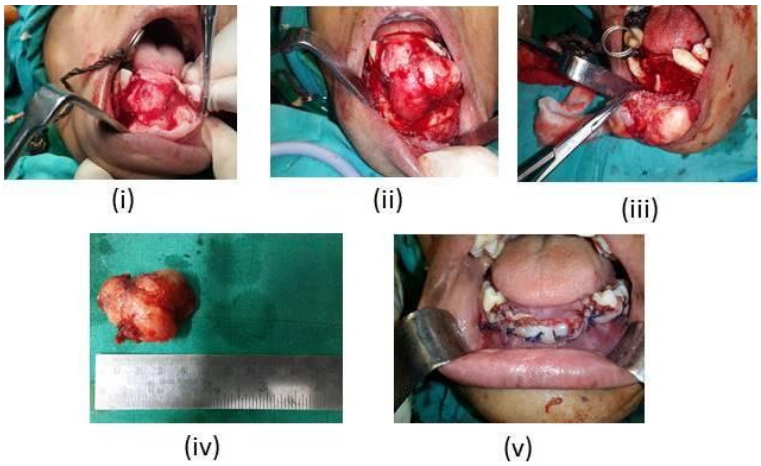

Fig. 4. Intraoperative

(i) Sulcular incision & flap reflection, (ii) Exposed tumor, (iii) Separation of tumor from both cortices, (iv) Tumor removed, (v) Closure

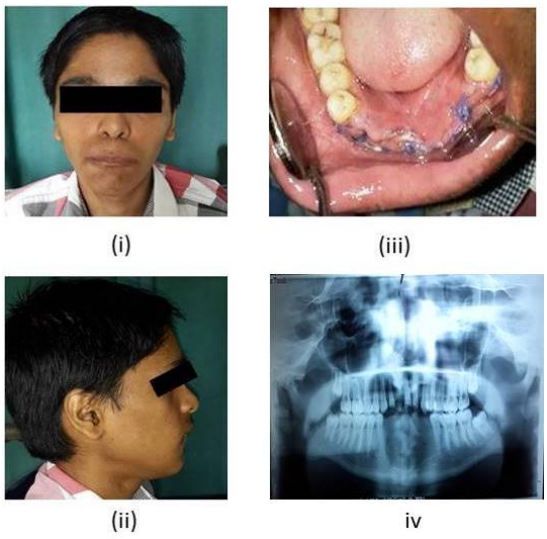

Fig. 5. Postoperative

(i) Front view, (ii) Lateral view, (iii) Intraoral view, (iv) Radiograph

3. DISCUSSION

CEOT is classified as either a central (94% intraosseous) or peripheral (6% extra-osseous) type [6]. The central type is twice more common in mandible than in the maxilla with the high prevalence rate in the premolar-molar region [6]. In our case the site of the tumor was in the mandibular anterior region extending from right cuspid to left 1st bicuspid region where it is less commonly seen. However an extraosseous variant of CEOT is dominant in the anterior region of jaws [6].

A wide age spectrum is seen with the occurrence of CEOT. Franklin and Pindborg reported a study of 113 patients of CEOT in which the age ranged from 8 to 92 years with mean at 40 years [7]. According to Cicconetti and colleagues, CEOT more frequently occurs between 40 to 60 years with peak incidence in the 5th decade [3]. The age of the patient in the present case was 16

years, much younger than the reported age range.

The CEOT is benign, slow growing, and expansile in nature. They are often asymptomatic and discovered on routine radiographs though occasionally patient may report with pain [2]. The increasing pressure of the tumor may lead to tooth tipping, rotation, migration, and/or mobility secondary to root resorption [4]. In our case, the patient complained of a small swelling of 6 months duration in the lower anterior region of jaw that gradually increased to approximately 5 x 4 cm in size. The vertical growth of the swelling had reached the occlusal level which led to traumatization of the mucosa overlying the swelling by the opposing teeth. There was no pain associated with the growth but there was obvious difficulty in deglutition. The considerable size of the growth along with the bicortical expansion made it visible extra orally as a swelling over the anterior mandible. All the teeth associated with tumor were mobile and showed various degrees of rotation due to bone loss and root resorption.

The histogenesis of this tumor remains evasive and different hypothesis exist such as its derivation from the stratum intermedium layer of the enamel organ in the tooth development stage, or from remnants of the primitive dental lamina found in the initial stage of odontogenesis [8,9]. The pathogenesis is not clearly understood; however, it commonly appears in connection with unerupted teeth. This case report presents a rare lesion that was not associated with unerupted teeth as similarly reported by Saha et al. [10].

The clinical behavior of CEOT is similar to that of intraosseous ameloblastoma, however, the CEOT has less tendency to penetrate into the medullary bone as compared to an ameloblastoma which is more aggressive [11]. The radiographic appearance of the CEOT is variable and may be unilocular (58%), multilocular (27%) or non-loculated (15%) depending upon its stage of development [12]. In the initial stages of its development it is completely radiolucent mimicking a dentigerous cyst because of its relationship with an impacted tooth [13]. Small calcifications start appearing in the delayed phase of development followed by osseous destruction and large radiopaque calcified material formations resembling “wind driven snow” in appearance [14]. The present case appeared as a unilocular radiolucency with the lesion surrounded by a well-defined radio-opaque border, and expansion of both cortices with perforation of the buccal cortex. Root resorption is unusual (4%) however was visualized in the present case along with tooth displacement [15].

The differential diagnosis included adenomatoid odontogenic tumor, calcifying odontogenic cyst, ameloblastic fibro-odontoma, and odontoma. Our case was unique clinically and radiographically. The oval calcifications resembled “Liesegang rings” which are pathognomonic of this tumor [16,17].

Methods of treatment range from curettage, complete extirpation of the tumor, or resection. Extirpation of the pathological mass with a margin of healthy tissue is usually recommended for mandibular lesions [18]. CEOT of the maxilla should be treated more aggressively as maxillary tumors are fast growing and are usually not well confined [11]. Conservative extirpation of the lesion was the treatment of choice for our patient considering his young age and the adverse functional and cosmetic effects of more radical procedures.

Although CEOT has been known to have a recurrence rate of 14%, much lower than ameloblastoma [19,20]. Our patient did not show any signs of recurrence during 1-year period of follow-up. However a longer follow-up period of a minimum of 5-10 years is required because of the tumor’s slow rate of growth.

4. CONCLUSION

Because CEOTs tumors can vary in their presentation they need to be fully evaluated clinically and radiographically and a biopsy performed to establish a definitive diagnosis. Surgical removal can usually be conservative because the tumor is not especially aggressive. Long-term follow-up with clinical and radiological examinations are necessary because the tumor is slow growing and can reoccur.

CONSENT AND ETHICAL APPROVAL

This work has been approved by the Ethical Committee and informed written consent to the work was taken by the patient.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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