

# CASE REPORT

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## PERIPHERAL CALCIFYING EPITHELIAL ODONTOGENIC TUMOR –A CASE REPORT AND BRIEF REVIEW OF LITERATURE

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**ABSTRACT:** Calcifying Epithelial Odontogenic Tumor (CEOT) is an extremely rare, benign neoplasm, accounting for approximately 1% of all odontogenic tumors. Peripheral CEOTs commonly resemble oral hyperplastic or reactive lesions and are histologically similar to their intraosseous counterparts. We report an unusual case of peripheral CEOT. A 40-year-old female presented with firm, gingival swellings localized in premolar area of the mandibular lingual gingiva. The presumptive diagnosis was giant cell granuloma. The masses were surgically excised under local anaesthesia with bone curettage. Morphologic features, histochemical and immunohistochemical tests revealed peripheral variant of calcifying epithelial odontogenic tumor. So gingival masses must be carefully evaluated for clinical and histologic evidence of neoplasia.

**KEYWORDS:** Calcifying epithelial odontogenic tumor, peripheral CEOT

**INTRODUCTION:** Peripheral odontogenic tumours are benign uncommon focal overgrowths of the oral soft tissue, usually occurring in the gingiva and the mucosa overlying the jaw. Peripheral Calcifying Epithelial Odontogenic Tumor (CEOT) is a rare benign tumour. CEOT is a locally invasive neoplasm characterized by deposition of amyloid-like material and its calcification. Very few cases of the extraosseous are reported till now and the gender ratio of male to female is 6:5. Mean age of onset is 30 years. Clinically, the lesion appears as a painless firm gingival mass, 0.8–1.7 cm in diameter, with a normal to red colour. The surface may be ulcerated. The mandible is the most common location with a slight predilection to the anterior and premolar area. Four of the nine reported cases have exhibited the unique microscopic features of a peripheral epithelial odontogenic tumor.<sup>2-8</sup>

**CASE REPORT:** A 34 year old systemically healthy female patient was reported to the Oral and Maxillofacial department, complaining of an asymptomatic slow-growing swelling which gradually increased to the present size within a period of 6-month duration in the lower left anterior lingual region. The onset was spontaneous and there was no history of trauma or

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functional impairment. Extraoral examination and palpation revealed cervical lymph nodes of normal size and consistency. On intraoral examination the painless, firm, sessile swelling of 3x2 cm was located in the lingual vestibular gingiva between right mandibular central incisor and left mandibular first premolar teeth, covered by a normal oral mucosa.(Fig .1)

Radiographically the lesion showed a characteristic saucerisation or cupping resorption of bone appearing as unilocular radiolucency on a IOPA film causing resorption at interdental areas and shows numerous scattered radiopaque foci of varying size and density suggesting of a peripheral lesion.<sup>9,10</sup> There is slight displacement of teeth with respect to lower left lateral incisor and canine .Occlusal radiograph shows buccal displacement with respect to lower canine

(Figure 2 and fig 2.a)

A clinical differential diagnosis of Fibroma ,Peripheral ossifying fibroma and Peripheral giant cell granuloma was considered and the decision was made to excise the lesion under local anaesthesia. During the surgical procedure, the lesion was totally enucleated and the bone surface was curetted and sent for histopathological examination. On gross examination the lesion was blackish brown in colour, hard in consistency, measuring 4.2x1.5x0.5 cms in dimension.(Figure 3)

Histopathological examination showed sheets of closely packed polyhedral odontogenic epithelial cells showing hyperchromatic nuclei and eosinophilic cytoplasm with mild pleomorphism. Amyloid like material between odontogenic epithelial cells, numerous calcified structures resembling liesegang rings were also seen. A final diagnosis of Peripheral Calcifying Epithelial Odontogenic Tumor was considered.

**DISCUSSION:** Calcifying epithelial odontogenic tumors (CEOTs) located in the oral and maxillofacial region are well-recognized examples of odontogenic tumors that display a broad spectrum of clinical and histopathologic features. On the other hand, the extraosseous variant of CEOT is extremely rare, with only few cases being described in the literatures.

The calcifying epithelial odontogenic tumor (CEOT) was first described by Pindborg <sup>11</sup> as a distinct entity in 1955. The eponym "Pindborg Tumor" was first introduced into the literature in 1967 to further describe this interesting and unique odontogenic tumor.<sup>12</sup>

The first reports of extra osseous CEOT date back to 1966 when Pindborg published two cases of gingival growth in the anterior jaw region of young patients. Since their recognition, 21 cases of these tumors have been published in the English literature. The initial consensus regarding the pathogenesis of CEOT was attributed to Pindborg in 1955. He stated that the CEOT was indeed of odontogenic origin and had been associated to unerupted teeth.

Clinically, peripheral CEOT is a slow-growing solitary painless mass. There is a slight female gender preference, with most cases occurring in middle aged women. Anatomically, a maxilla to mandible ratio of 1:2 has been reported for the peripheral types and mandibular lesions are mainly found in the premolar/molar region. In contrast, the present case occurred in the anterior region of the jaws. Peripheral CEOTs mainly appear as nonspecific sessile exophytic masses in anterior gingival tissue and can mimic a wide variety of oral lesions, including, pyogenic granuloma, peripheral giant cell lesion, peripheral ossifying fibroma, and fibrous hyperplasia. However, the etiology of these conditions is usually known and careful examination is necessary when any local irritant is defined or the lesion recurs after surgical treatment.

In the present case, a provisional diagnosis of peripheral ossifying fibroma was made. This diagnosis was based on the clinical and radiographic appearance of the mass, including

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normal-appearing mucosa in contrast to the red or purple color characteristic of pyogenic granuloma and peripheral giant cell lesions, respectively. Peripheral giant cell lesions usually produce a superficial 'peripheral-cuff' radiolucency that is not observed in ossifying fibroma. On the other hand, the observation of radio-opacity in the latter has been associated with mild resorption of the crest of the ridge. As observed in the present case, Peripheral CEOT may appear as radiolucency with scattered radiopaque foci. Histologically, small amounts of mineralized material might be observed, which is a common finding in peripheral CEOT types. Over the years, the histology of CEOTs has been well documented. Ai-Ru et al. subclassified this tumor into four distinct microscopic patterns, although two or more types may be present in the same tumor. Type 1 consists of sheets, nests and masses of polyhedral epithelial cells exhibiting prominent intercellular bridges. Type 2 is characterized by a cribriform arrangement of tumor cells. Type 3 consists of scattered or densely populated tumor cells. Type 4 is characterized by small nests and cords of epithelial cells. In addition, several cellular variants such as clear cell, pigmented, Langerhans cell containing bone and cementum forming, myoepithelial cell, and noncalcifying subtypes have been reported. According to the histologic classification of Ai-Ru et al., Peripheral CEOTs can be divided into three main variants: clear cell, conventional, and hybrid. Only one case was reported by Etit et al., in which the CEOT presented unusual features of ameloblastoma.

In the present case, the slow gingival growth of the mass, small size, and clinical appearance similar to that of peripheral ossifying fibroma may lead to an incorrect clinical diagnosis and inadequate treatment. Abrahão et al. reported a unique case of bilateral peripheral CEOT presumptively diagnosed as bilateral pyogenic granuloma that was surgically excised without bone curettage and recurred bilaterally one year later. Thus, although peripheral CEOT is a nonaggressive tumor and recurrence is rare, a favorable prognosis depends on complete excision of the tumor and effective curettage. Furthermore, malignant transformation is an extremely rare phenomenon.

The clear cell odontogenic carcinoma<sup>13-15</sup> should also be considered in the histologic differential diagnosis of the CEOT. Although this neoplasm may exhibit nests composed entirely of clear cells with a frequent deposition of calcium salts, usually in the form of Liesegang rings which is not observed commonly with the CEOT. In addition, an extraosseous clear cell odontogenic carcinoma has not yet been reported.

In summary, Pindborg tumors are uncommon odontogenic tumors and even though extremely rare peripheral CEOTs, should be included by dental surgeons in the differential diagnosis of gingival lesions.

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Fig. 1



Fig. 2

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Fig .2.a



Fig .3

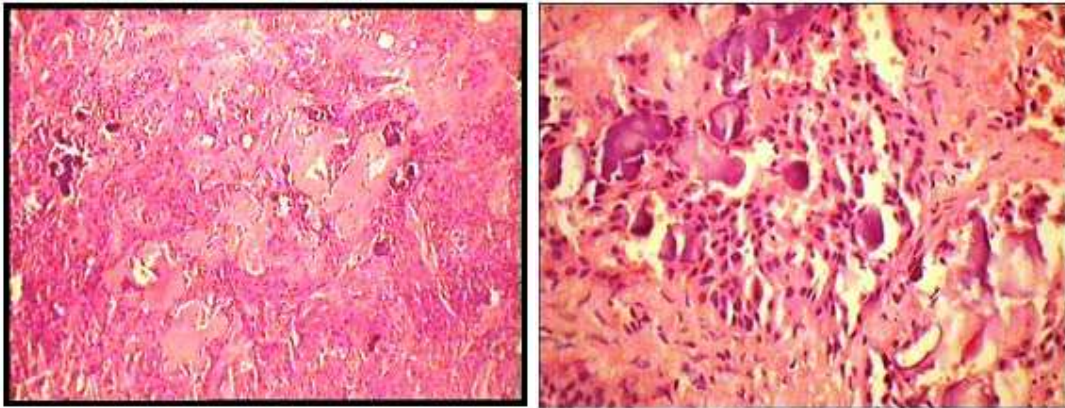


Fig .4