

Clear cell variant of extraosseous calcifying epithelial odontogenic tumor: a case report

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Abstract: Calcifying epithelial odontogenic tumor (CEOT) is a benign epithelial odontogenic tumor occurring most frequently in the posterior part of the lower jaw. Extraosseous CEOT is one of the rarest forms of this tumor, and few such cases involving the maxillary gingiva have been reported in the literature. Here we present a case that showed progressive enlargement in the left maxillary gingival area over a period of 11 years. Clinical examination showed an ulcerated mass measuring 52 × 38 mm located adjacent to the lateral incisor and canine. Histologically, the tumor showed proliferation of sheets and cords of epithelial cells with granular, eosinophilic cytoplasm and round to oval nuclei. In other areas, the epithelial cells exhibited a clear, vacuolated cytoplasm and foci of eosinophilic, homogeneous material representing amyloid deposition. The present case of extraosseous CEOT with clear cells was considered to be a very rare form of this tumor. (J Oral Sci 51, 485-488, 2009)

Keywords: CEOT; clear cell, extraosseous; gingival; Pindborg tumor.

Introduction

Calcifying epithelial odontogenic tumor (CEOT) (also called Pindborg tumor) was first described in 1959 by Pindborg (1). It accounts for less than one percent of all odontogenic tumors.

It is most often encountered in patients aged 30-50 years, but shows no sex predilection. About 65% of all reported cases have occurred in the mandible, most often in the posterior part (2). The most common presenting sign is a painless and slow-growing swelling. In radiographs, the lesion shows unilocular, and more often multilocular radiolucency. It may be completely radiolucent, but the density and size may vary (3-6).

Extraosseous CEOT has been reported only rarely. Only a small proportion of these tumors show the microscopic features of the clear cell variant (7,8). These appear most often as non-specific sessile masses in the anterior gingival tissue (2,3,9). This paper describes the clinical, radiographic, and microscopic features of a rare case of the clear cell variant of peripheral CEOT.

Case Report

In May 2008, a 70-year-old female patient was seen at the Mashhad Faculty of Dentistry. Her chief complaint was progressive enlargement of the left maxillary gingival tissue over the previous 11 years. Clinical and radiographic examinations showed an ulcerated mass measuring 52 × 38 mm located adjacent to the lateral incisor and canine (Figs. 1 and 2).

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An excisional biopsy with 5-mm safety margins included underlying periostium was performed. The appearance of the underlying alveolar bone was normal (Fig. 3). Histologically, the tumor showed proliferation of sheets and cords of epithelial cells with granular, eosinophilic cytoplasm and round to oval nuclei with some considerable nuclear variation. In other areas, the epithelial cells exhibited

clear, vacuolated cytoplasm in a fibrous stroma with large or small areas of amorphous, eosinophilic, hyalinized, amyloid-like extracellular material plus areas of basophilic calcification (Figs. 4 and 5). Foci of an eosinophilic, homogeneous material representing amyloid deposition as revealed by Congo red staining (Fig. 6). PAS staining was negative, indicating an absence of glycogen (Fig. 7).

Discussion

CEOT is generally considered a benign lesion, but



Fig 1 Radiograph of the patient before surgery.

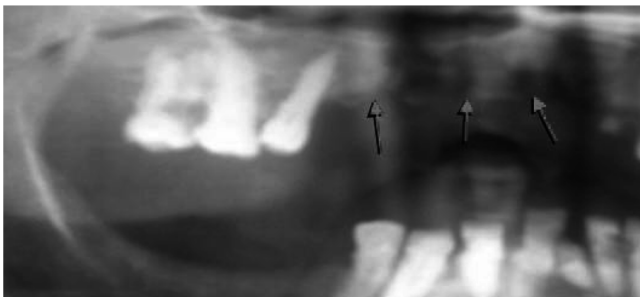


Fig 2 Radiograph of the patient after surgery.

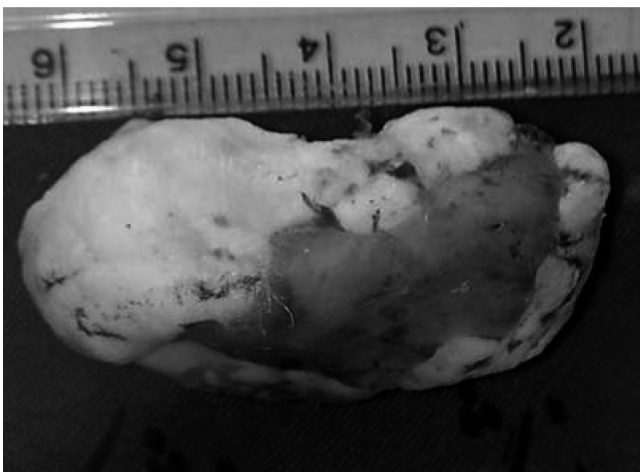


Fig 3 Gross photograph of the peripheral CEOT after excisional biopsy.

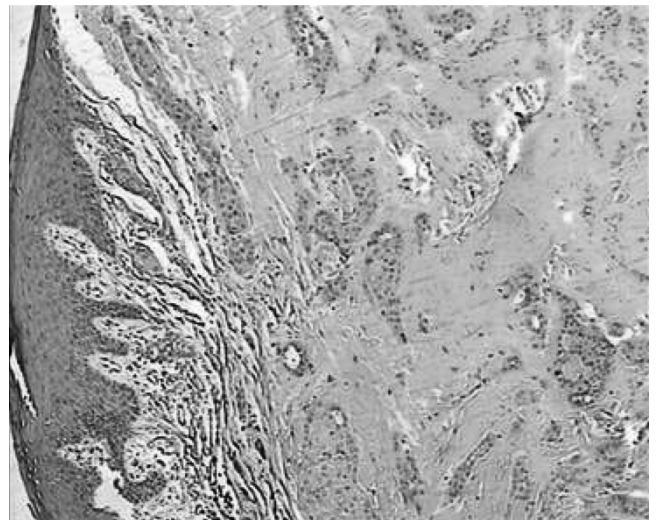


Fig 4 Low-power view demonstrating a thick gingival layer surrounding the extraosseous CEOT.

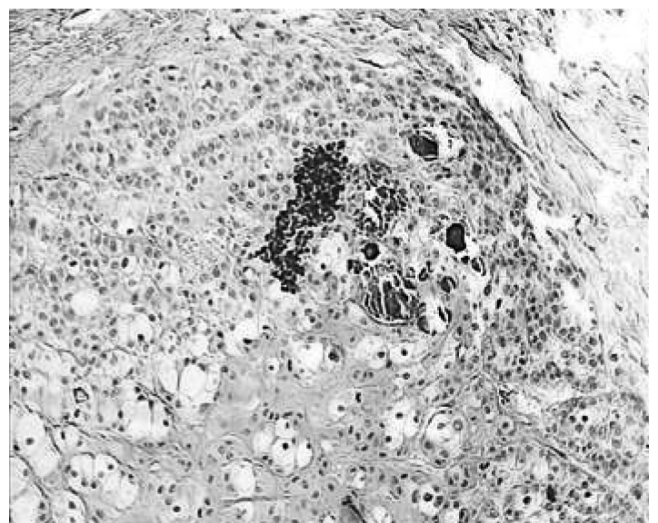


Fig 5 Microscopic view of the epithelial neoplasm composed of sheets and nests of polyhedral epithelial cells with abundant eosinophilic and granular cytoplasm intermixed with clear cells and basophilic calcification (H-E staining; ×400).

occasionally it can invade the surrounding tissues (9). As stated by Buchner and Sciubba (7), follow-up information about extraosseous CEOT is limited, and long-term follow-up reports are rare. Therefore, observation of affected patients over a long period is necessary in order to increase our understanding of the biologic behavior of this rare tumor (3).

Appearance of clear cells in an odontogenic tumor can indicate progressive or malignant behavior in comparison with a non-clear-cell histology (10). In the present case, a clear cell component was observed, but this did not include any significant mitotic figures, nuclear atypism, or pleomorphism as evidence for more aggressive biological behavior.

The clear cell variant of peripheral CEOT is rare, and few cases have been reported to date (Table 1). In 1997, Houston and Fowler (11) reported two cases of extraosseous CEOT in the maxillary gingiva, both of which exhibited a prominent clear cell component. In 1999, Kumamoto et al. (12) reported a case of the clear cell variant of CEOT in the posterior maxilla of a Japanese girl. Histological examination revealed sheets and strands composed of clear vacuolated epithelial cells in a stroma containing intercellular amyloid-like deposition. In 2003, Mesquita et al. (13) reported a case of the clear cell variant of peripheral CEOT located on the maxillary gingiva of a woman, presenting as a 2-cm solitary, firm nodule. Polyhedral and clear epithelial cells and amyloid-like deposition were evident.

The present case was an ulcerated mass measuring 52 × 38 mm located adjacent to the lateral incisor and canine. Histologically, the tumor showed proliferation of sheets

and cords of epithelial cells with granular, eosinophilic cytoplasm and round to oval nuclei.

Surgical management of CEOT varies depending on the

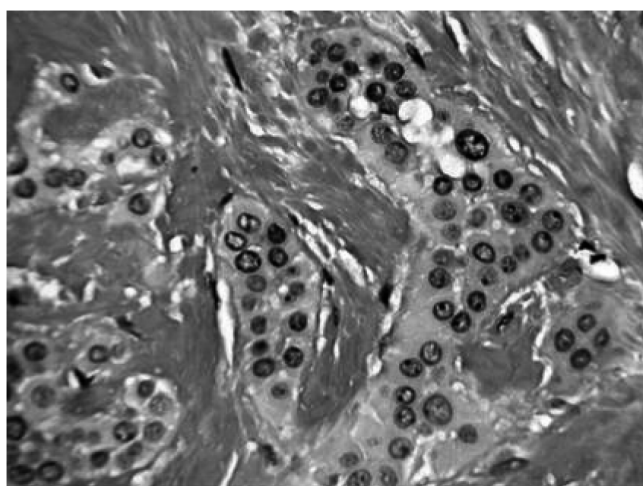


Fig 6 Congo red staining. Pools of amorphous eosinophilic amyloid are present near to epithelial and clear cell sheets ($\times 400$).

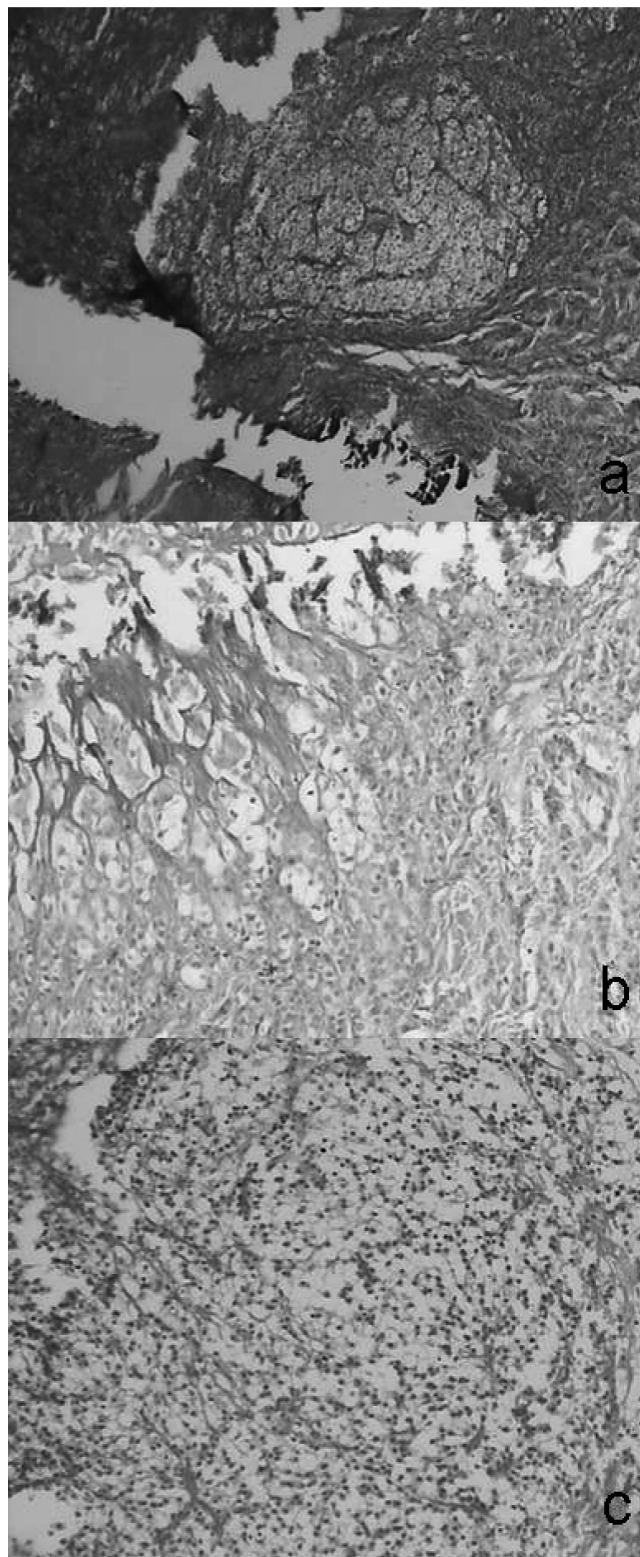


Fig. 7 Microscopic view indicates a group of clear cells negative for PAS staining (a: $\times 4$, b and c: $\times 10$).

Table 1 Reported cases of clear cell variant of peripheral CEOT

Author(s)	Reported year	Race	Age of patient	Sex	Location	Size
Houston and Fowler (11)	1997	USA	64 (Case 1) 27 (Case 2)	Male (Both)	Left palatal mucosa (Case 1) Right posterior mandibular facial gingival (Case 2)	15 × 10 mm (Case 1) 14 × 10 mm (Case 2)
Kumamoto et al. (12)	1999	Japan	14	Female	Posterior maxilla	Not stated
The present case	2009	Iran	70	Female	Maxillary lateral incisor and canine area	52 × 38 mm

size and site of the tumor as well as the extent of bone destruction (6). Appropriate management of peripheral CEOT consists of simple excision (2).

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