Calcifying epithelial odontogenic tumor: a case report

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Calcifying epithelial odontogenic tumor (CEOT) is a rare benign lesion. A case of CEOT in the left mandible of a 46-year-old man is presented. This case is exceptionally unusual because it was left untreated for 10 years, indicating in some way the course of the tumor. Histologic findings both at the time of first diagnosis and at the final treatment 9 years later were identical. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;112:e117-e120)

The calcifying epithelial odontogenic tumor (CEOT), also named Pindborg tumor after the pathologist who described it in 1955, is an uncommon benign locally invasive tumor. It usually involves the premolar-molar area of the mandible, there is no gender predilection, and the peak incidence is found between the fourth and fifth decades of life. A peripheral soft tissue variant of the lesion has been described as well as a malignant one.

CASE REPORT

A 46-year-old caucasian male patient, an economic refugee, presented to the outpatient department complaining of a swelling in his lower left jaw. The mass was painless, increasing slowly in size over the years, and lately causing difficulty in mastication. The mass was first noticed by the patient more than 10 years before. The patient reported that a biopsy was taken from his lower left jaw 9 years before. The
histopathology report confirming the presence of a CEOT and a pantomogram of bad quality, but still revealing the radiographic appearance of the lesion, could be retrieved from that period (Fig. 1). The patient refused treatment at that time.

Nine years later, extraoral examination showed slight fullness and asymmetry of his lower left face. The lower border of the mandible on palpation gave the impression of a dome-shaped expansion in the premolar area. There was no cervical lymphadenopathy clinically. Intraorally, the lesion, firm in palpation, occupied the left mandible from the canine to the retromolar region with expansion of the lingual and buccal plates. The bridge work on that side appeared almost sunk into the lesion. The inferior alveolar nerve was unaffected (Fig. 2).

The orthopantomogram revealed a mixed radiopaque-radiolucent endosseous lesion of the left mandible extending from the left lower canine to the area in front of the angle of the mandible. The lower border of the mandible was also affected.
The computerized tomographic (CT) scan images depicted an intraosseous lesion causing expansion of the mandible in all directions, erosion of both plates, and lingual extraosseous extension (Fig. 4).

The diagnosis of CEOT was confirmed again by an incisional biopsy. Under general anesthesia, via a submandibular approach, a left segmental mandibulectomy extending from the left lower incisor to the area in front of the angle was performed.

Reconstruction was achieved using a 13-hole reconstruction plate with 3 screws on each side (Fig. 5). The postoperative period was uneventful. The patient was regularly seen on an outpatient basis for 1 year (Fig. 6). Thereafter, the patient failed to present himself to the outpatient clinic.

**Histopathology report**

The tumor was composed of closely packed polyhedral epithelial cells in a scanty fibrous, often hyalinized, stroma. The epithelial cell frequently had nuclear pleomorphism, anisonucleosis, and hyperchromatism. The mitotic activity was minimal or absent. The intracellular rounded masses of tumor filled with eosinophilic homogenous material become progressively calcified (Figs. 7 and 8).

**DISCUSSION**

CEOT is a rare benign lesion representing <1% of all odontogenic tumors. No more than 150 cases have been reported in the English-language literature. The histogenesis of the CEOT tumor remains controversial. Origin from the reduced enamel epithelium, stratum intermedium, rest of dental lamina, and basal cells of the oral epithelium have been proposed.

The clinical presentation depends on the site of involvement, but it typically is that of a slowly enlarging mass causing expansion of the cortical plates, without pain or altered sensation.

The radiographic appearance is not characteristic. Early tumors may appear radiolucent. As the tumor matures, areas of calcification develop. The lesion...
may be unilocular or multilocular with a mixed radiolucent-radiopaque picture, occasionally giving the “soap bubble” appearance. The presence of an impacted crown 3, 4 may or may not be a feature. CT scan images may be helpful in delineating the extent of the tumor, the involvement of cortical plates, basal bone, and extraosseous extension. Ameloblastoma, calcifying follicular cyst, and malignant CEOT should be considered in the differential diagnosis of Pindborg tumor.\textsuperscript{3,5,11} Definite diagnosis is based on histologic examination.\textsuperscript{5,12}

CEOT is managed surgically. The tumor is not encapsulated, and most authors agree that the resection should include a safe margin of clinically and radiographically healthy bone.\textsuperscript{1,10} CEOT is a rare lesion, and large series with follow-up are lacking. Consequently, treatment policy and recurrence rate are still debatable, although most authors agree that 1 cm of safe margins is adequate.\textsuperscript{1}

The case we have presented is a typical CEOT regarding site of occurrence, age, histology, and clinical and radiographic features. The uniqueness is based on the fact that the tumor was left untreated for at least 10 years. Histology and radiographic image were available from the time of first diagnosis as well as the time of treatment 10 years later.

The histology is similar in both instances, and comparing the 2 pantomograms the tumor has the same appearance and a minimal change in its size, considering the quite large time interval. This might suggest that the lesion exhibits an early stage of more or less fast grow followed by a period of maturity during which it remains almost unchanged. In planning the surgery and the rim of healthy bone to be excised, effort should be made to be made to preserve the continuity of the mandible and minimize the extent of healthy bone excision.

REFERENCES