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Calcifying epithelial odontogenic tumor: a case report with 3-year follow up

Tumor odontogênico epitelial calcificante: caso clínico com 3 anos de acompanhamento

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RESUMO

Tumor Odontogênico Epitelial Calcificante (TOEC) foi reconhecido originalmente por Thonay Goldman. Ficou conhecido por Tumor de Pindborg devido a sua descrição em 1955 por Pindborg e que foi detalhadamente realizada em 1958. É um tumor raro e importante por sua semelhança a um carcinoma mal diferenciado. Os adultos são principalmente afetados, em uma idade média de 40 anos. O sítio de eleição é o corpo posterior da mandíbula, o qual é acometido em dobro em relação a maxila. Apresenta crescimento lento e usualmente é assintomático, até que o edema se torne obstrutivo. Radiografias mostram área radiolucente, de margens mal definidas e, usualmente com o amadurecimento, áreas de radiopacidade dentro da tumoração. TOEC não são encapsulados e, por isso, localmente invasivos. Seu comportamento assemelha-se ao dos ameloblastomas. Os autores relatam um caso em paciente do sexo feminino, 22 anos de idade, com localização mandibular, reabsorções radiculares e associado a um canino retido (base de mento). Foi realizado tratamento clínico, cirúrgico e realizadas radiografias panorâmicas para diagnóstico e controle do tumor, com acompanhamento de 3 anos.

Palavras-chave: Tumor Odontogênico Epitelial Calcificante; Tumor de Pindborg; Tumores Odontogênicos; Neoplasias Maxilares

ABSTRACT

The calcifying epithelial odontogenic tumor (CEOT) was documented originally by Thonay Goldman. It was known as Pindborg tumor due to its description in 1955 by Pindborg and its description was full detail accomplished in 1958. It is a rare and important tumor for its similarity to a carcinoma badly differentiated. The adults are mainly affected, in a 40 year-old medium age. The election area is the body of the mandible, which is attacked in double than the maxilla. It has slow growth and usually is asymptomatic, until the edema becomes obstructive. Radiographs show radiolucent area, with badly defined margins and, usually, radiopacity areas inside of the tumor, with the mature. The CEOT are not encapsulated and, for that, locally invasive. Its behavior resembles to the ameloblastomas. The authors report a case of a 22-year-old woman with CEOT in the mandible associated to root reabsorptions and an unerupted canine tooth in the mentol base. It was realized clinical and surgical treatments and it was accomplished panoramic radiographs for diagnosis and tumor control, with 3-year follow up.

Keywords: Pindborg tumor; Calcifying epithelial odontogenic tumor; Odontogenic tumors; Maxillary neoplasm.

INTRODUCTION

The odontogenic tumors are a complex group of lesions of several clinical behavior and histological variation. Some odontogenic tumors are composed of odontogenic epithelium, without any relationship to the odontogenic mesenchyme.

The calcifying epithelial odontogenic tumor (CEOT), also known as Pindborg tumor, is a rare lesion that represents about 1% of the odontogenic tumors. Although it has an odontogenic origin, its histogenesis is uncertain. The tumoral cells present morphology similar to the cells of the stratum intermediate of the enamel [1,2].

It was described as a different entity and separate from the ameloblastomas by Gorlin et al. in 1962[3]. Most of the cases do not present painful symptomatology and they are usually diagnosed by swelling of the affected tissues. It occurs mainly in adults, in a 40 year-old medium age. The election area is usually the premolar and molar regions of mandible, which is affected twice more than the maxilla[4].

CEOT can exhibit variable radiographic pattern. The central lesions are usually

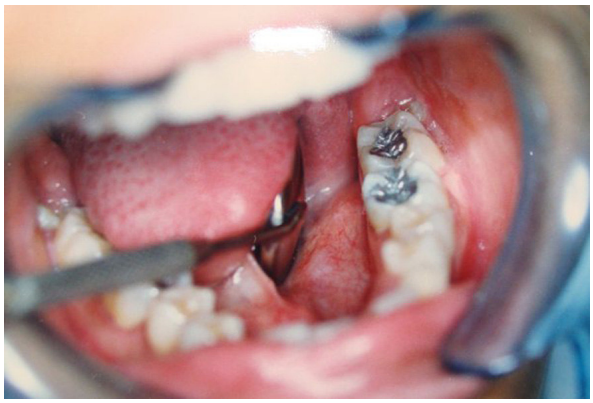
related to well-defined unilocular radiolucency areas or to multilocular radiographic pattern, frequently associated to unerupted tooth[5]. Some other imaging CEOT characteristics are cortical expansion, root reabsorption, dental displacements and poorly defined margins. The calcification presence is constant, however, in variable intensity. When the tumor is associated to unerupted tooth, the calcified material is distributed through its coronary portion[5,6,7].

CEOT presents approximately the same distribution between men and women[4,8]. The known recurrence rate is of 15%, according to Neville et al. in 1998[9]. The recurrence data and the biological behavior of this tumor indicate that the aggressive treatment is not advisable[10].

CASE REPORT

Diagnosis

A 22-year-old woman presented to the Hospital e Maternidade Celso Pierro – PUC Campinas, with discreet edema on the left mandible in premolar and in body of mandible areas with facial asymmetry (Figura 1A). The patient had no spontaneous painful symptomatology; however it was present during palpation. Radiographic examinations confirmed the presence of intraosseous lesion in left mandible that extended from incisors to area of left inferior second molar, with widespread root reabsorption and unerupted left inferior canine on symphysis (Figura 1B). The differential diagnosis included ameloblastoma, dentigerous cyst and CEOT.



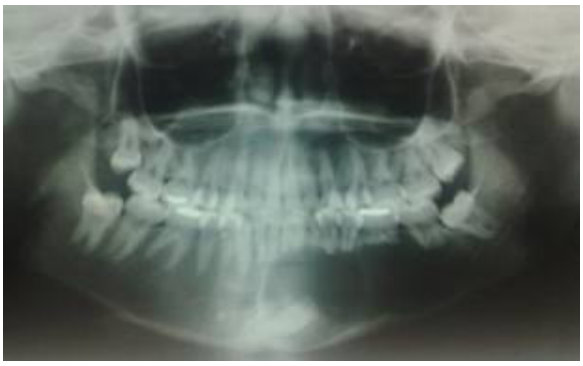


Figure 1. Panel of clinical and radiographic images of the case. A=Edema in mandible premolar and body of mandible areas, B=Panoramic radiographic showing lesion that extends from left inferior molar to contralateral central incisor. Note lesion related to unerupted left inferior canine.

Procedure

The lesion involved teeth were endodontic treated. It was observed that the teeth presented pulp vitality. It was accomplished the exeresis of the intraosseous lesion and the regularization of the teeth apical root. The unerupted left inferior canine tooth was extracted. The surgical wound was sutured and a curative compression was made to contention and decrease of the extra-oral edema. The procedure was accomplished under general anesthesia (Figura 2).

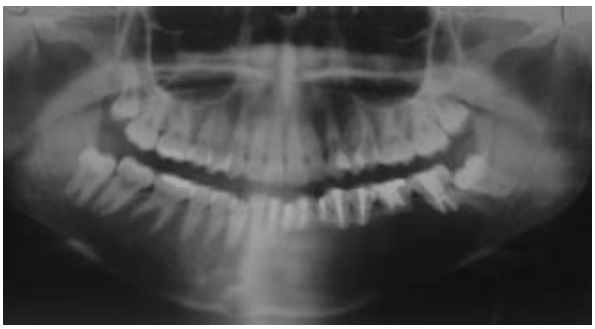


Figure 2. Endodontic treatment in the lesion involved teeth, removal of the unerupted canine tooth and lesion enucleation.

The removed lesion was sent to the pathology service. The found diagnosis was calcifying epithelial odontogenic tumor.

Follow up

Patient annually accomplishes clinical and radiographic attendance. At the 3-year follow up examination, no sign of recurrence

was noted that can be observed in the panoramic radiographic (Figura 3).

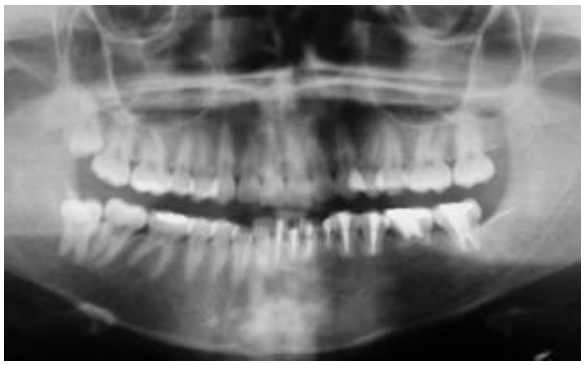


Figure 3. Note in the panoramic radiographic the bone repairing after 3 years of lesion enucleation.

DISCUSSION

CEOT was described for the first time by Pindborg in 1955[11]. It affects more frequently the mandible, with prevalence to molars than to premolars (3:1)[12,13,14,15,16].

In the literature there are few reports of its occurrence in the anterior region of the mandible[15,17]. Although its presence in the maxilla is rare, authors as Lee et al. (1992)[18] and Mohtasham N et al. (2008)[19] described CEOT in the maxillary antrum. The anterior area of the maxillaries is usually associated to peripheral lesions; however in most described cases it was observed that the intraosseous location is the most common in this area[8,13,14,20]. In the presented clinical case, the lesion had an extensive intraosseous location; it committed symphysis and body of mandible with low expansion of the vestibular cortical, without involvement of the adjacent soft tissues The committed age group varies from 8 to 92 years. Kaplan et al. (2001)[2.1] . showed the preference to females, 1.5:1 in 67 cases, and the age group varied between 4th and the 5th decade of life. The lesion affected different etiologic groups, with slight predilection for Caucasians.

The histologic aspect of the tumor consists of a mass of polyhedral giant epithelial cells, with hyperchromatic nucleus, divided by a sparse connective tissue. Some eosinophil homogeneous bodies of amyloid nature that progressively calcify are present between the cited components[11].

The epithelial cells are extremely united and can show cribriform disposition with the cells, united for intercells bridges, in relation with e-caderina and α-catenina expression[22].

Regarding the nature of the amyloid substance, controversy exists concerning the degenerative origin or if it is segregated actively. Labban (1990)[23] and Slootweg (1991)[24] indicate that this calcified amyloid material acts as a stimulus to the tumoral stroma, taking it to secrete a collagen calcified matrix. Different authors describe other five histologic patterns of CEOT[25,26,27].

Clinically, CEOT presents in its evolution an expansible slow growth, usually asymptomatic. Rosa LEB, Jaeger RG em 1990[28] describe important bleeding, and in one of their cases it was accomplished an angiography that showed a highly vascularized tumor.

Pindborg (1958)[29], Krolls (1974)[30], Basu et al. (1984)[31] refer cases of tumor with malignant behavior and ganglionic involvement.

Usually the tumor is discovered on the image exams from the routine appointments.

Radiographically, it is observed a radiolucent image, with different characteristics since CEOT has three evolutionary phases. In a first moment its image is totally radiolucent, similar to a dentigerous cyst (mainly if associated to an unerupted tooth). Therefore, small intratumoral calcifications can be seem in the image, although it is not considered as a pathognomonic characteristic. The last evolution phase is characterized by an image like honeycomb, caused by the bone destruction and the tumor calcification[32]. The lesion described in this report can be included in the first evolutionary phase.

The larger lesions can be multilocular, similar to the ameloblastoma, especially if they have intratumoral calcifications.

The differential diagnosis of this tumor in their two forms (intra and extraosseous) include ameloblastoma, giant cells granuloma, cemento-ossifying fibroma, ameloblastic fibro-odontoma, myxoma, and ameloblastic fibroma[33]. It can also have as differential diagnosis the central mandible hemangioma, if the lesion presents important bleeding[34,35].

If radiographically compared, it can be included in this differential diagnosis the odontogenic cyst, adenomatoid odontogenic tumor and calcifying odontogenic cyst. The differential diagnosis of the extraosseous Pindborg tumor with presence of clear cells (more of the half are of this type) should include tumor of salivary gland, clear cell carcinoma, peripheral ameloblastoma,

oncocytoma and the mucoepidermoid carcinoma[36].

The conservative treatment (enucleation and curettage) has a recurrence rate of 14%. The aggressive treatment (resection marginal or segmented) has no recurrence rate description[35]. The low rate of described recurrence seems to propose a conservative treatment of the lesion[33]. Some authors, like Junqueira et al.[35], differ of this opinion and suggest a more aggressive treatment. Other authors treat accordingly to the lesion size; if small, the enucleation is enough and, if big, a more aggressive treatment is adopted, in order to improve the prognostic.[13,16]

In this case it was proposed a conservative surgical treatment, because the lesion did not demonstrate aggressiveness.

The follow up should be annual and a long period of postoperative attendance is indicated[13,16].

CONCLUSION

Calcifying epithelial odontogenic tumor or Pingdborg tumor is a benign odontogenic lesion, rare, with specific origin, that can appear as a radiologic casual finding. It can be confused with dentigerous cysts and with other osseous tumors, being obligatory the establishment of the correct diagnosis with the pathological exam.

This tumor has unexpected clinical behavior and can be treated surgically in an aggressive way or not, depending on its local condition.

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