IJO - INTERNATIONAL JOURNAL OF HEALTH SCIENCES AND NURSING
(ISSN: 2814-2098)
https://ijojournals.com/
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Volume 07 || Issue 01 || January., 2024 ||

Calcifying Epithelial Odontogenic Tumor or Pindborg tumor : about a case in CHU-JDR Antananarivo.

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Abstract

Pindborg tumor or Calcifying Epithelial Odontogenic Tumor (CEOT) derived from the enamel epithelium. It is benign, rare (less than 1% of odontogenic tumors), painless, slowly evolving, hardand is embedded in the bone. It is frequent in subjects between 20 and 60 years old, with a balanced sex ratio. The area of predilection is the mandibular bone. The objective of this study was to diagnose Pindborg tumor and to determine its management. We report the case of a 58-year-old woman, evolving for 10 years in the left ramus. The tumor was removed by enucleation under general anesthesia, then a recurrence occurred after three years. It was painless, bone-like, firm, benign looking, filling the vestibule. CT scan showed a mixed density osteolytic and osteocondensing image in the left ramus, blowing out the cortical bone, partitioned and seat of multiple calcifications. The second procedure consisted of an interrupting mandibular resection and immediate placement of a titanium screw plate for reconstruction.This tumor may be confused with ameloblastoma, ossifying fibroma, and other

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odontogenic tumors with mineralized components. The anatomopathological examination of the surgical specimen confirmed the diagnosis of CEOT. Because of its compartmentalized form, interrupted bone resection is the best technique to avoid recurrence.

Keywords: benign tumor, CEOT, interrupting resection, recurrence.

Introduction

Pindborg's Tumor or Calcifying Epithelial Odontogenic Tumor (CEOT) was first discovered by Jens Jorgen Pindborg in 1955 **(1)**. It is a benign tumor, rare (less than 1% of odontogenic tumors), painless and slow-growing. It is common in people aged between 20 and 60, with a balanced sex ratio **(2)**. Hard, poorly limited, often associated with a molar and integral with the bone, the preferred site is mandibular **(3)**. The aim of this study is to report a case of TOEC in a 58-year-old woman from Antananarivo.

Case report

A 58-year-old woman was referred to us in September 2019 with a recurrent mandibular, symphyseal and left paraspinal swelling **(figure 1.A)**. The first tumoral manifestation had been evolving for around ten years, involving only the left paraspympetal region, anterior to 35. Enucleation was performed under general anaesthetic. Three years after the operation, a swelling reappeared at the same site, extending into the symphyseal region. The swelling was painless, close to the mandibular bone, firm, filling the vestibule, covered with ulcero-necrotic mucosa and a whitish coating, and did not bleed on contact. The floor of the mouth was free but pushed back, with a healthy mucosa; the tongue was mobile **(figure 1.B)**. The patient had no associated sensory-motor disorders, and the cervical lymph nodes were free. The panoramic radiograph or orthopantomogram showed a polygeodic image of the left ramus, with a large symphyseal-parasymphyseal osteolytic zone and osteocondensing zones posteriorly **(figure 2).**

Computed tomography confirmed this radiographic diagnosis and added the notion of cortical effraction and the presence of multiple calcifications **(figure 3)**. This reappearance of a new

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lesion at the same site is a probable sign of recurrence.A second operation was performed, consisting of an interrupted mandibular resection removing the tumour, followed by immediate placement of a titanium screw-plate for reconstruction **(figure 4)**.The post-operative course was complicated by a submental salivary fistula which disappeared after removal of the sutures (figure 5).Pathological examination of the surgical specimen suggested a calcified epithelial odontogenic tumor (Pindborg tumor). The excision margins were healthy.

Comments

CEOTis a tumor usually seen in adults, but it can also develop in children **(4)**.

Because of their almost identical nature, it is difficult to distinguish CEOT from other benign odontogenic tumors such as ameloblastoma, calcifying odontogenic cyst and ossifying fibroma **(5)**. Indeed, CEOT and ameloblastoma are two intraosseous tumors that share the same clinical and radiological appearance.Their location in the mandibular premolar region is similar, as is their slow progression. On CT scan, the images are almost identical: CEOT shows a mixed osteolytic and osteocondensing density in the mandibular body, compartmentalized and the site of multiple calcifications **(6)**, as found in our patient. In ameloblastoma, multilocular soap-bubble osteolysis and multiple root resorption are typical **(7)**. The histological appearance of CEOT is specific: cells with enlarged, squamous-like, multi-nucleated nuclei and a low mitotic index, as well as amyloid deposits and calcifications **(8)**; this is confirmed in our case. Ameloblastoma, on the other hand, derives from Malassez epithelial remnants and shows no calcifications **(9)**.

The indication for treatment depends on the size of the tumour, its relationship with the bony cortices, and whether or not it leads to disruption of the basilar border of the mandible, the posterior border or the upper end of the ramus **(10)**. Conservative enucleation-type treatment is indicated for less aggressive early lesions. This is the case for the management of a calcifying odontogenic cyst or a small ossifying fibroma. For invasive, polygeodic, recurrent and voluminous tumours such as our case, surgical treatment is radical **(9)**: interrupting bone

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resection to avoid recurrence.It is important to respect an exeresis margin of 1 to 2cm, in healthy tissue, while preserving the functions of neighbouring tissues.

Thus, the indications for radical or conservative treatment of CEOT depend on the size and location of the lesion, and whether it is polygeodic or not. Indeed, the risk of recurrence after enucleation is 15 to 30% at a follow-up of 2 to 4 years (8).Interruptive bone resection is indicated for large, recurrent and invasive CEOT**(9)**.However, this technique is highly invasive, leading to functional and aesthetic sequelae. Immediately after tumor removal, a titanium screw-plate is inserted to provide a temporary solution. Adaptation of the manducatory apparatus required re-education, so that the patient was gradually able to mobilize her jaw.The salivary fistula disappeared spontaneously after removal of the sutures.Prognosis was good.

Conclusion

CEOT is a rare benign tumor specific to dento-maxillary structures.It could be confused with ameloblastoma or other odontogenic tumors, and it is the histological examination of the lesion that confirms the diagnosis.CEOT shares the same surgical treatment technique as ameloblastoma. Bone resection with a good margin off the tumor is the best treatment.

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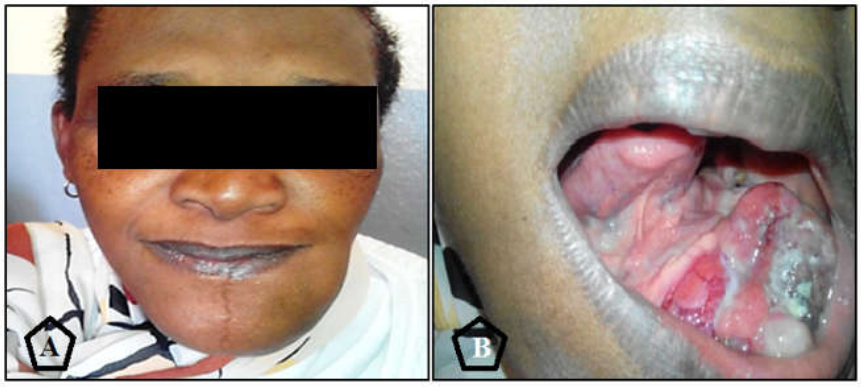

Figure 1: A. Facial asymmetry caused by left mandibular tumor.
B. Endobuccal aspect of the tumor.
(Andriamanantena RH, 2017)

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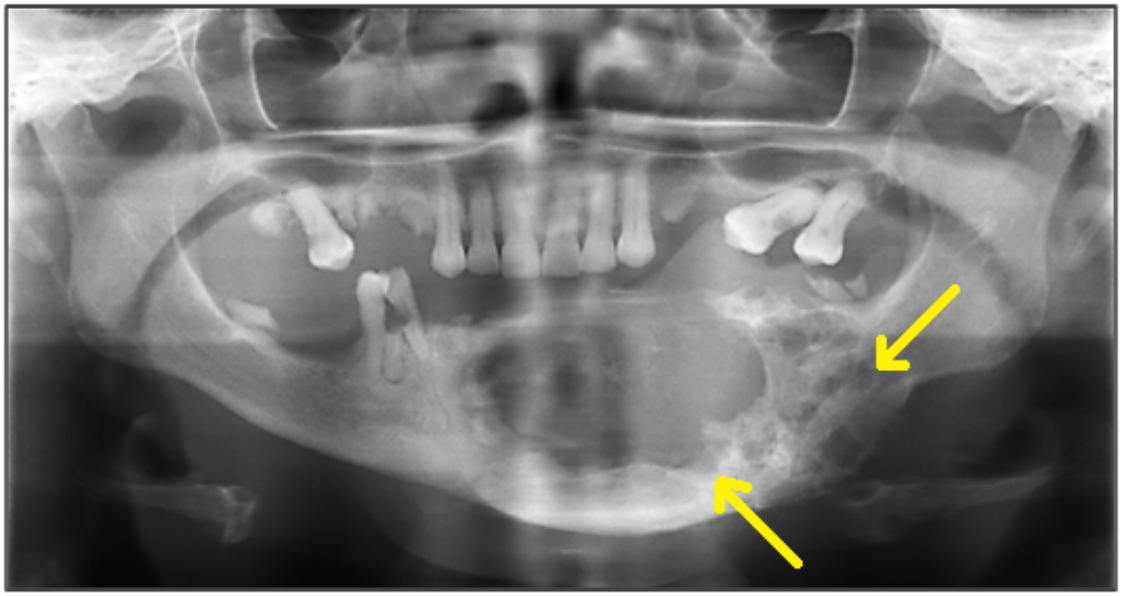

Figure 2: Orthopantomogram showing an image of a large radiolucent zone in the left symphysis-parasymphysis, and polygeodic osteocondensing zones posteriorly with basilar rim involvement.
(Andriamanantena RH, 2017)

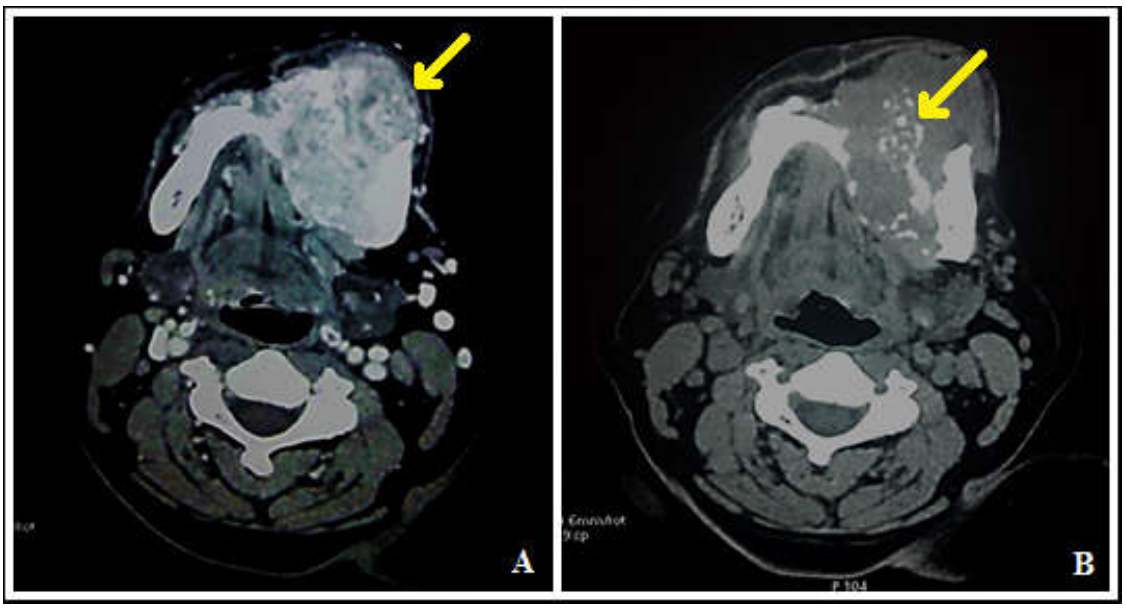

Figure 3: Computed tomography (CT) scan showing mixed osteolytic and osteocondensing density, compartmentalized and the site of multiple calcifications.
A: CT scan with contrast medium injection.
B: CT scan without contrast medium injection.

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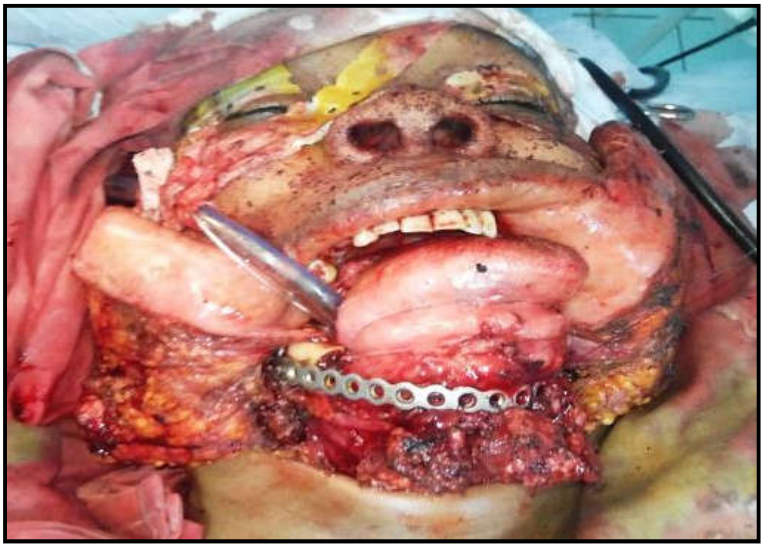

Figure 4: Interrupting mandibular resection removing the tumor, followed by placement of a screwed titanium reconstruction plate.
(Andriamanantena RH, 2017)

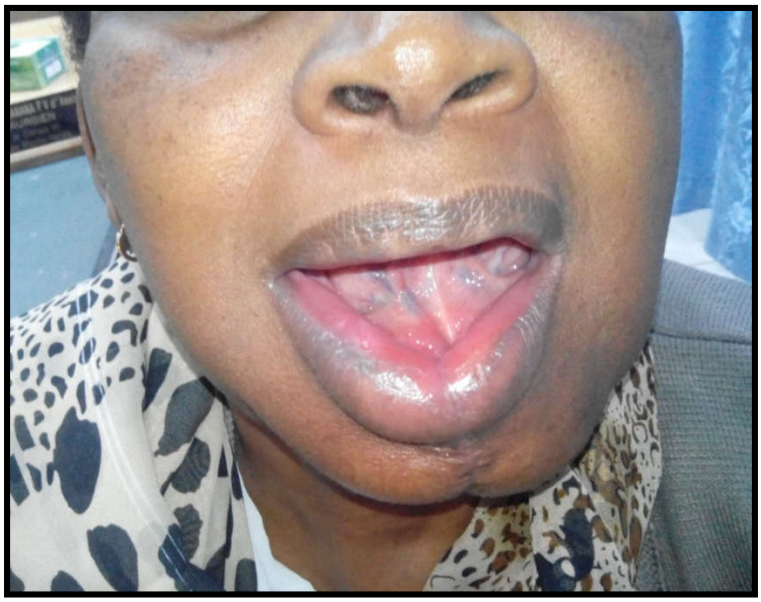

Figure 5: Postoperative follow-up (one month post-op)
(Andriamanantena RH, 2017)