CASE REPORT

CALCIFYING EPITHELIAL ODONTOGENIC TUMOR (PINDBORG TUMOR)

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ABSTRACT
Calcifying epithelial odontogenic tumor is rare. It may mimic both a non-odontogenic or odontogenic tumour causing expansion and destruction of the involved bones. Histopathology is the main stay for definitive diagnosis. Treatment options vary from enucleation to hemi - mandibulectomy or maxillectomy followed by reconstruction. We present a young female patient with CEOT in an attempt to create awareness in its management and to provide evidence based recommendations to add on to the existing literature on this rare tumour.
Key Words: Calcifying epithelial odontogenic tumor, Pindborg tumor, CEOT

INTRODUCTION
The calcifying epithelial odontogenic tumor (CEOT), also known as the Pindborg tumor, is an uncommon lesion that accounts for < 1% of all odontogenic tumors (Petersons, 2004). It was first described as a separate pathologic entity by Dutch pathologist Jens Jorgen Pindborg in 1955. He described it as a benign but locally invasive epithelial tumor, which behaves clinically like an ameloblastoma (Pindborg, 1958). It presents typically as an intraosseous mass that is expansile, painless and exhibits slow growth (Houston and Fowler. 1997). CEOT more frequently affects adults in an age range of 20-60 years, with a peak of incidence between 40 and 60 years. It is usually not found in children and adolescents, with no apparent sex predilection (Franklin and Pindborg, 1976). The mandible is more commonly affected than the maxilla in ratio of 3:1 (Goode, 2004). Radiographically, the most common presentation is a mixed radiopaque/radiolucent lesion, frequently associated with an impacted tooth. However, depending on stages of development, CEOT may present variable radiographic appearances (Franklin and Pindborg, 1976). The lesion usually consists of a radioluscent area, which is well defined. The area is often unilocular when small and larger lesions tend to have honeycomb or soap bubble appearance (Goode, 2004). The mixed radioluscent and radiopaque pattern occurs most often (65%) followed by the completely radioluscent pattern (32%) and least often the totally radiopaque "wind driven snow" pattern (3%). When the tumour is associated with impacted tooth, it may appear as pericoronal radiolucency with or without small radiopacities (Anisha et al., 2010). About 190 cases of CEOT have been reported in the dental literature, with a recurrence rate of about 10-15% (Franklin and Pindborg, 1976; Cicconetti et al., 2004; Anderson's, 1996). This report presents a case of CEOT in an adult patient with CEOT.

CASE REPORT
A 32-year-old female patient presented to the oral and maxillofacial surgery clinic at the University of Nairobi, Dental Hospital. Her chief complaint was that of a slow growing, painless swelling in the lower left jaw that had been there for the past one year. She associated the swelling with an extraction that was done in the same area. Extra - oral examination showed facial asymmetry on the left side. Intra - orally there was a swelling that extended from the 32 to 37 area along the buccal vestibule up to the inferior border of the mandible, with bucco – lingual extension. The lesion was non – tender, bony hard and the overlying mucosa was normal. There was grade two mobility of the involved teeth. An

orthopantomogram revealed an area of mixed radio – opacity and radio – luscency from the distal of 33 extending mesially to 38, showing the classic radiological 'honey comb' appearance of CEOT (Fig. 1). The patient's medical history was unremarkable. Tentative differentials included ameloblastic fibroma and a fibro – osseous lesion. An incisional biopsy done under local anaesthesia and histopathology investigations confirmed CEOT. Resection of the lesion with reconstruction using a stainless steel plate was done (Figs 3, 4). Histopathology reported cells that exhibited prominent desmosomal contacts, with leisegang rings and additional foci of calcification which are features consistent with CEOT (Fig 2). Approximately six months later, the patient underwent an iliac crest harvest and graft in preparation for occlusal rehabilitation. Post – operative radiographic examination showed no signs of recurrence of the tumor.

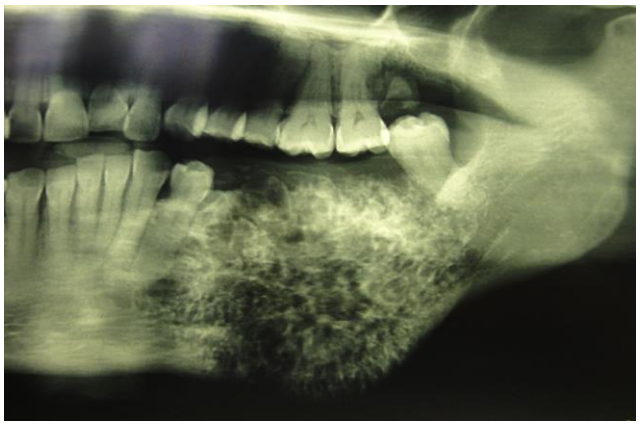

Fig 1: Pre – operative radiograph

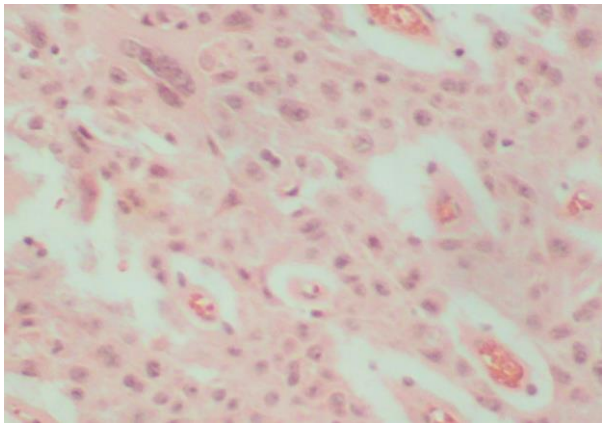

Fig 2: Histopathology using H&E stain, magnification x100

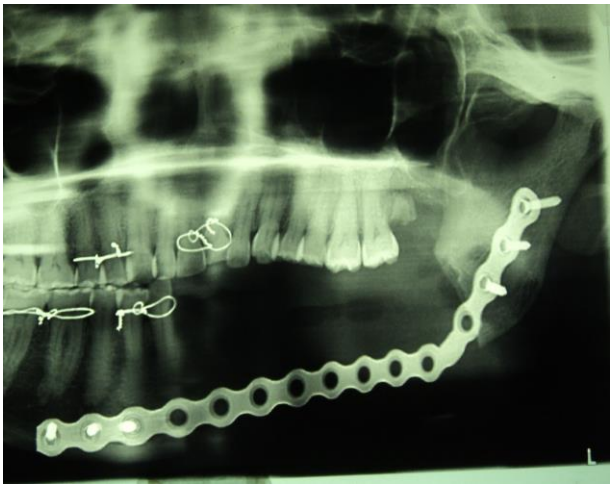

Fig 3: Post – operative radiograph with reconstruction plate insitu

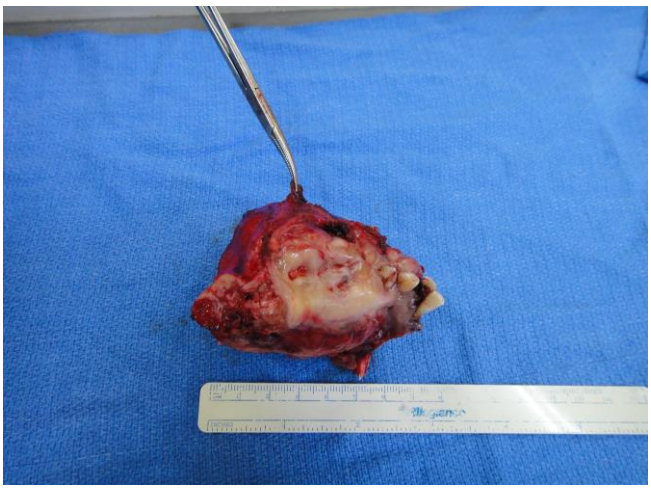

Fig 4: Excised tumor

DISCUSSION

As seen in our case, CEOT, though benign, can cause a significant amount of tissue destruction and disfigurement, especially due to its expansile nature mimicking a variety of

odontogenic and non – odontogenic tumors of the jaw (Lan et al., 1997). In this case, the closest differential diagnosis was a fibro – osseous lesion. Histopathology of the entire specimen confirmed CEOT. Histopathology examination is, therefore the main stay in arriving at a definitive diagnosis. Caution must be taken since CEOT mimics clear cell odontontogenic carcinoma (CEOC), which should form part of the histological differential diagnosis (Angumjar et al., 1996). Literature reports that this intra – osseous tumor manifests as a slow expansile tumor that is painless and when located in the maxilla, patients may sometimes complain of epistaxis, nasal stuffiness and headaches (DeBoni et al., 2006). Few extra – osseous cases have been reported (Wood and Goaz, 1997). There is no consensus on the originating cells. Some pathologists suggest that it is derived from the stratum intermedium, while others believe it originates from remnants of the dental lamina (Anisha et al., 2010). As seen in our case, malignant transformation is extremely rare, with only two reported cases in literature (Veness et al., 2001).

Options in treatment range from simple enucleation and curretage to radical excisions like hemi - mandibulectomy or maxillectomy (Shanmuga and Ravikumar, 2009). Nonetheless, the tumor is generally recommended to be treated identically to the ameloblastoma and odontogenic myxoma, with 1.0 cm bony linear margins and the appropriate attention to soft tissue anatomic barriers (Houston and Fowler, 1997). In our set up due to late presentation most patients present when the tumor is large with expansion of the buccal cortex and mandibular margin as was seen in this case. Regarding conservative procedures, follow up of patients is difficult due to accessibility to nearby hospitals, in addition, to the expenses involved in taking serial radiographs post - operatively hence a radical approach is preferred. Resection of the mandible followed by reconstruction using a stainless steel plate was done in this case. However if presented at its early stages conservative approaches may be attempted with good prognosis. And in general, the prognosis of CEOT is good, with infrequent recurrence (Anderson, 1996). In conclusion, reporting of this benign but locally invasive tumor still remains key in an attempt to create awareness in its management and to provide evidence based recommendations.

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