Case Report

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CALCIFYING EPITHELIAL ODONTOGENIC TUMOUR: REPORT OF TWO CASES

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ABSTRACT:

Calcifying epithelial odontogenic tumor (CEOT) is an uncommon, locally invasive, benign odontogenic tumor of epithelial origin accounting for only about 1% of all odontogenic tumors. The present case report describes two cases of intraosseous CEOT of maxilla and mandible respectively, with an emphasis on clinical, radiographic and histopathological features.

Keywords: CEOT, Pindborg tumor, Impacted, Maxilla

INTRODUCTION:

The calcifying epithelial odontogenic tumor(CEOT), also known as a Pindborg tumor, is an odontogenic tumor first described by the Danish pathologist Jens Jorgen Pindborg in 1955.[1] It is an uncommon odontogenic tumour accounting for only approximately 1% of all odontogenic tumours. It usually presents as a slow-growing, asymptomatic painless swelling but locally invasive.[2] CEOT can present as an intraosseous lesion (central type) in the majority of cases (95%) and extraosseous or peripheral lesions account for fewer than 5% of cases. CEOTs are thought to originate from the stratum intermedium or reduced enamel epithelium of odontogenic epithelium.[7] Treatment includes the surgical removal of the lesion, with a recurrence of about 14% in the reported cases.[3-5] The prognosis is considered good.

CASE DETAIL:

CASE 1: A 28 year old male patient reported to the Department of Oral Medicine and Radiology with a complaint of swelling involving left upper back tooth region since 3 months. The swelling was gradually increasing in size and painless. It was not associated with any discharge or numbness. His past medical and dental history were non-contributory. Extraoral examination revealed no gross swelling. Intraoral examination showed a well-defined swelling, 4 ×4 cm in size, extending from distal of maxillary left second premolar to the maxillary tuberosity anteroposteriorly. Medio-laterally, it was extending from upper left buccal vestibule to palate, 2cm lateral to the midline. The left upper buccal vestibule was obliterated. Mucosa over the swelling showed indentations mark of the lower antagonist teeth. Left Maxillary second and third molars were missing. On

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palpation, the swelling was non tender and firm to hard in consistency.

The intraoral periapical radiograph showed a well-defined unilocular radiolucency extending from left maxillary second premolar to the end of alveolar ridge. Impacted second molar was displaced superiorly. There were few calcifications seen within the lesion.

A panoramic radiograph showed a well-defined unilocular radiolucency measuring approximately 3 cm × 3 cm in size extending from the distal of maxillary right second premolar to the maxillary tuberosity. Thinning of the walls of the left maxillary sinus was seen with an impacted second molar tooth displaced superiorly to the floor of sinus. The third molar was also displaced posterosuperiorly. Root resorption was seen in the first molar. Few calcifications were also seen within the lesion.

The axial, coronal and sagittal view of computed tomography revealed a hypodense, 27×24×26 mm in size, expansile lesion involving the alveolar margin of left maxilla with extension upto left maxillary sinus surrounding the crown of impacted left second molar. Both buccal and palatal cortical bone expansion with thinning and perforation was present. Areas of calcification within the lesion were also noted.

Incisional biopsy was done and histopathology sections showed sheets, cords and islands of polyhedral epithelial cells in a fibrous stroma. Cells were closely packed with distinct cellular outline, eosinophilic cytoplasm, prominent intercellular bridges and slight nuclear pleomorphism. Abundant areas of amorphous, eosinophilic, hyalinised amyloid like material was seen dispersed within the tumour cells. Numerous areas of calcification were noted. Few isolated calcifications showed typical Liesegang ring appearance. Based on the histopathological features, a final diagnosis of calcifying epithelial odontogenic tumor was made. Surgical excision of the lesion was done which further confirmed the diagnosis of CEOT. Patient is kept under regular follow up and no signs of recurrence has been reported in period of 1 year.

CASE 2:

A 19 year old male patient reported with the complaint of swelling in the lower left back tooth region since 2 months. The swelling was asymptomatic and gradually increasing in size. The past medical and dental history was non-contributory. Extra-oral examination revealed no gross swelling. Intraoral examination showed a well-defined swelling, 3 ×3 cm in size, extending from the distal aspect of mandibular left second premolar to the retromandibular region anteroposteriorly with no evidence of mandibular left first and second molar clinically. Bucco-lingual extent was from the depth of the buccal vestibule to the depth of the lingual vestibule. Mucosa over the swelling showed indentations mark of the upper antagonist teeth. On palpation, the swelling was firm in consistency and non-tender.

Radiographic examination with panoramic radiography and contrast enhanced computed tomography was conducted. Panoramic radiograph revealed a well-defined radiolucent lesion in the left mandibular body region extending from the distal aspect of mandibular second premolar to the ascending ramus mesio-distally. Supero-inferiorly it extended from the occlusal level to the inferior border of the mandible. Interior of the lesion showed the presence of impacted and displaced mandibular left first molar with focal specks of calcification in its occlusal aspect. There was also evidence of impacted and displaced mandibular second molar to the ramus region. CECT of the maxillofacial region revealed expansile lytic lesion in the mandibular body region on left side showing pericoronal relation with the impacted tooth and calcific foci within. Expansion and thinning of cortex with cortical breach on the buccal side was seen.

Incisional biopsy and histopathologic examination was conducted which was suggestive of calcifying epithelial odontogenic tumor. The patient is planned for surgical excision.

DISCUSSION:

WHO defined CEOT/Pindborg tumor as a locally invasive epithelial odontogenic tumor, characterized by the presence of amyloid material that may become calcified. CEOT represents less than 1% of all odontogenic tumor and occurs in patients between 20 to 60 years of age, with a mean around 40 years.[2-6] Most cases are intraosseous, approximately 6% arise in extraosseous locations. Extraosseous lesions arise most commonly in the anterior gingival region. Intraosseous tumours affect mandible more commonly than maxilla with a ratio of 2:1. It is most often located in the premolar-molar region of the mandible and associated with one or more impacted tooth in half of the cases. Chrcanovic BR, Gomez RS in their review of 339 cases found that CEOT reported in 4th to 5th decade, more common in mandible (60%) than maxilla (40%) and associated with impacted tooth half of the time.[5] However, present cases were reported in young patients with age less than 30 years.

CEOT commonly manifests as a painless slow growing lesion causing bone expansion, tooth movement and root resorption. Maxillary lesions may cause symptoms such as epistaxis, nasal stuffiness and headache. Even though the impacted tooth was displaced to the floor of maxillary sinus, our patient did not report any history of epistaxis, nasal stuffiness or headache. Our case also showed root resorption of 1st molar and displacement of 2nd and 3rd molar.

Various radiographic features have been described in literature for CEOT, which include pericoronal or nontooth related radiolucency, mixed radiolucent-radiopaque or dense radiopaque lesions, unilocular or multilocular lesions.[9] Characterstic radiographic appearance is seen as many small irregular trabeculae traversing radiolucent area which gives a

characteristic "driven snow" appearance on the radiograph due to scattered flecks of calcification.[8] Present cases also showed few areas of calcification on the radiograph.

CT and 3D reconstruction are helpful in determining the exact extension of the tumor, displacement of the tooth and visualization of the internal structure, which plays a role in appropriate diagnosis and treatment planning. CT imaging helps in the process of interpretation, but the final diagnosis of CEOT is based on histological examination.

The histologic pattern of CEOT is typical and well defined. The tumor consists of polyhedral cells arranged in masses, sheets, islands, cords, rows or strands in a scanty connective tissue stroma. The cells are pleomorphic with well-defined borders, prominent nucleoli and abundant finely granular cytoplasm filled with an eosinophilic “amyloid-like" material, which gradually becomes concentric calcified deposits, resembling psammoma bodies called the "Liesegang rings," which is considered as pathognomonic for this tumor.[7] All these features did exist in the reported cases.

The differential diagnosis of CEOT depends on radiographic appearance. The radiographic appearance of CEOT varies with development and thus it can present as a well-defined radiolucency, mixed radiolucent-radiopaque or completely radioopaque mass. In case of radiolucent lesion- dentigerous cyst, odontogenic keratocyst, ameloblastoma, odontogenic myxoma; whereas in mixed radiolucent radiopaque lesion- Calcifying odontogenic cyst, adenomatoid odontogenic tumor, complex odontoma, ameloblastic fibro-odontoma, fibro-osseous lesions, osteoblastoma should be considered.

The treatment methods can range from simple enucleation or curettage to resection. Enucleation with a margin of normal tissue is usually recommended for mandibular lesions. CEOT of the maxilla should be treated more aggressively as maxillary tumors grow more rapidly and are usually not well confined. Our patient underwent enucleation and no recurrence is reported in 1 year of follow-up. The recurrence rate may range from 14% to 20%. The malignant behavior is extremely rare.[10]

CONCLUSION:

CEOT is a rare odontogenic tumour with different types of presentation which can lead to under diagnosis or misdiagnosed as less aggressive pathology. One must be aware of its range of presentations for correct diagnosis and management of odontogenic jaw lesions.

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FIGURES:

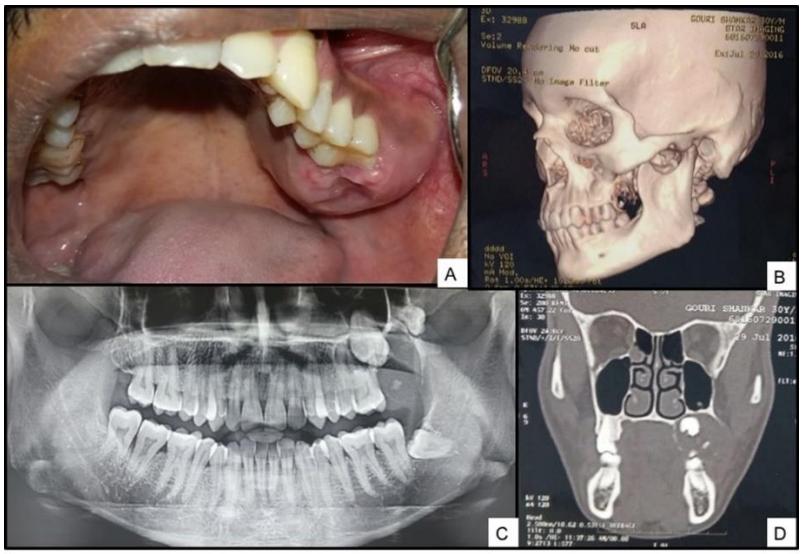

Figure 1: A) Intraoral photograph showing swelling in left posterior maxilla.
B) 3D CT showing osteolyetic lesion in left posterior maxilla.
C) OPG showing unilocular radiolucency displacing 2nd and 3rd molars. Calcifications were also seen within the lesion.
D) CT(coronal section) showing osteolytic lesion with perforation of cortical plate with impacted 2nd molar

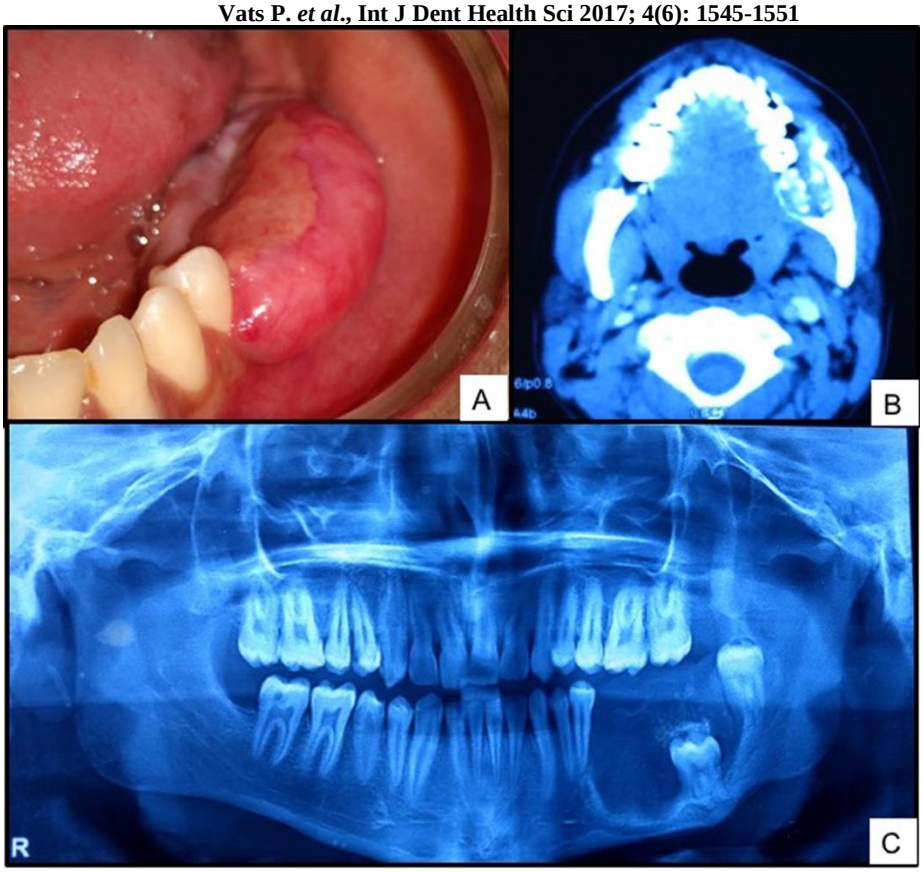

Figure 2: A) Intraoral photograph showing swelling in left back tooth region.
B) CT showing osteolytic lesion with flecks of calcification in internal structure.
C) OPG showing well defined, radiolucent lesion in body of mandible with impacted 1st molar. Area of calcification is seen above impacted tooth.

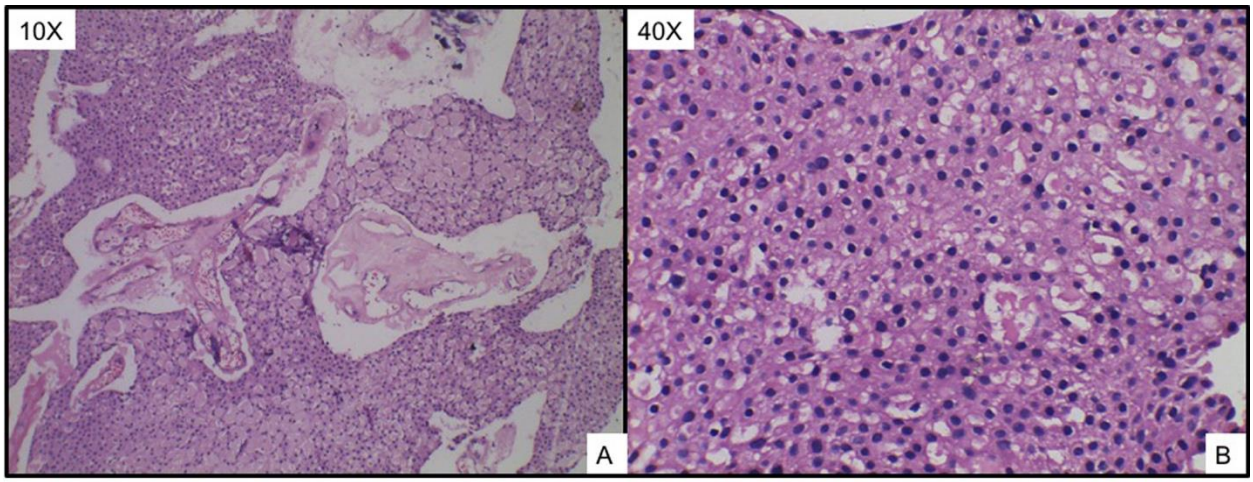

Figure 3: Microphotograph shows (A) abundant areas of amorphous, eosinophilic, hyalinised amyloid like extracellular material dispersed between tumour cells; (B) lesion composed of sheets of polyhedral epithelial cells exhibiting distinct cellular outlines, prominent intercellular bridges and nuclear pleomorphisim.

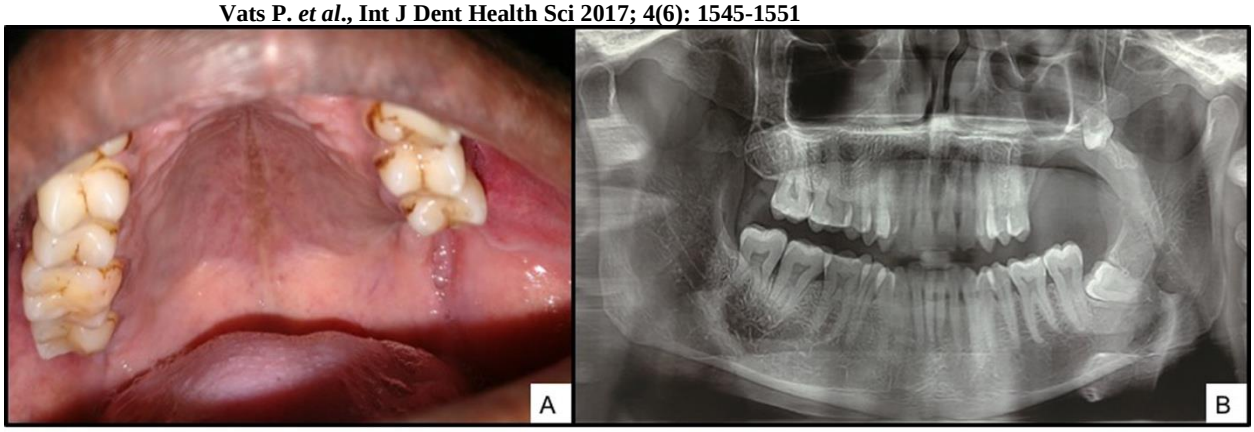

Figure 4:Follow up after 1 year for case 1