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RESEARCH ARTICLE

CALCIFYINGEPITHELIAL ODONTOGENIC TUMOUR ASSOCIATED WITH IMPACTED CANINE IN MAXILLA – A CASE REPORT

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ABSTRACT

Calcifying Epithelial Odontogenic Tumour (CEOT), also known as Pindborg Tumour is a rare odontogenic epithelial neoplasm. So far, nearly 200 cases have been reported in literature. We are reporting a case of Calcifying Epithelial Odontogenic Tumour in a 25 year old male patient with a painless bony swelling in the maxilla. Approximately, 50% of the cases are associated with an un-erupted tooth or an odontome, and it was the same with our case, except that it was an impacted maxillary canine over shadowed by a retained deciduous tooth. Considering the intrabony location of the lesion and its limited size, we opted for a more conservative surgery. The clinical, radiographic and histopathologic features and the surgical treatment are discussed with relevant references.

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INTRODUCTION

The term Calcifying Epithelial Odontogenic Tumour was first introduced into the scientific literature almost 50 years ago by the late Dr. Jens J. Pindborg (1956, 1958). CEOT is a benign, but locally aggressive tumour. It usually presents as a hard painless mass, generally affecting the mandible more than the maxilla. The characteristic histopathologic description consists of sheets and islands of polyhedral epithelial cells with multiple calcifying bodies with laminated appearance representing liesegang rings (Neville, 2002). Surgical treatment varies from simple enucleation to resection of the affected bone followed by reconstruction of the resected jaws.

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Here, we present a case of CEOT associated with impacted canine in maxilla which is treated by simple enucleation and removal of the impacted tooth

Case Report

A 25year old male patient came for a routine dental examination. The history of presenting illness revealed that the patient had sensitivity in upper front tooth which aggravated on taking hot or cold food and relieved within minutes. No relevant history was present in the past medical and dental history. On examination no significant finding was present extra orally. Intraoral examination revealed retained deciduous in 63, cervical abrasion in 63, sensitive to probing (Fig1). On routine radiological investigation, IOPA in 63 revealed a unilocular radiolucency along the apex with ill-defined borders.

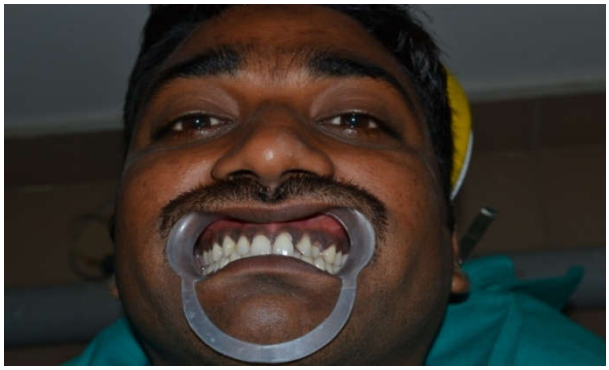

Figure 1.

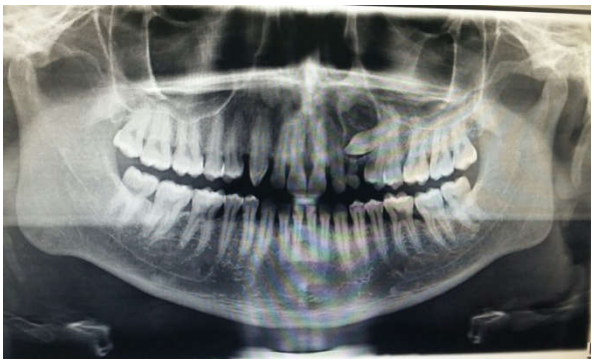

Figure 2.

Occlusal radiograph revealed impacted 23 with unilocular mixed radio opaque and radiolucent areas of size 2x2 cm and a radiopaque mass in the centre. OPG reveals impacted 23 with well-defined radiolucency lesion measuring approximately 2 x1.5cm with radiopacity in the centre of lesion (Fig 2).An axial CT hard tissue window revealed unilocular radiolucency with corticated well defined borders of size 2x2cm and radiopaque mass present in the centre. Posterior border of the lesion is in close proximity to maxillary sinus wall (Fig 3).

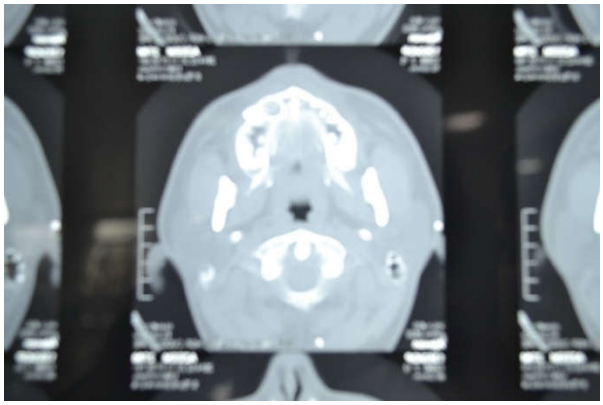

Figure 3.

Surgical removal of the tooth was planned. Infraorbital block, nasopalatine block and greater palatine block were given using lignocaine 2% with adrenaline 1:2,00,000. After obtaining adequate anaesthesia, 2 vertical incisions and sulcular incisions were placed to raise a trapezoidal flap in the 2ndquadrant. The mass was excised and the impacted canine tooth was removed by tooth division (Fig4). The excised mass was sent for histopathological investigation (Fig5). Microscopically, the tumour showed strands and nests of cells with pleomorphic nuclei, prominent nucleoli, uncommonmitoses, and a pronounced eosinophilic cytoplasm all surrounded by a fibrous

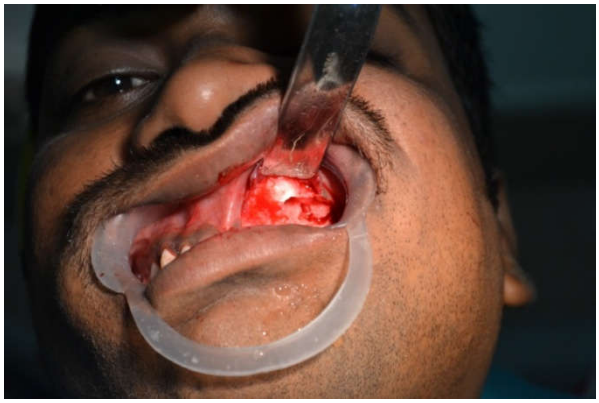

Figure 4.

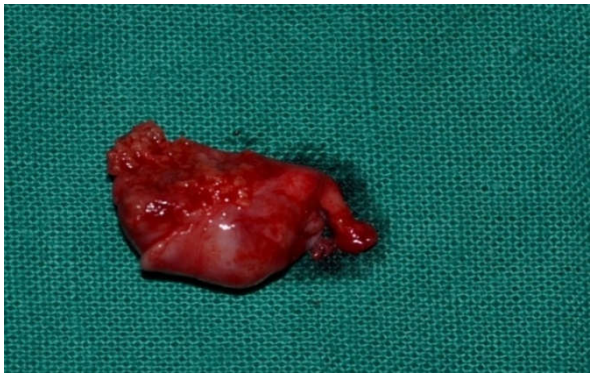

Figure 5.

tissue containing ample eosinophilic material that stained intensely positive for amyloid with Congo red. Many calcified spots were found (3) (Fig6). Following the enucleation of the lesion the patient was followed up for a period of five years with no evidence of recurrence clinically and on routine radiological investigation (Fig 7).

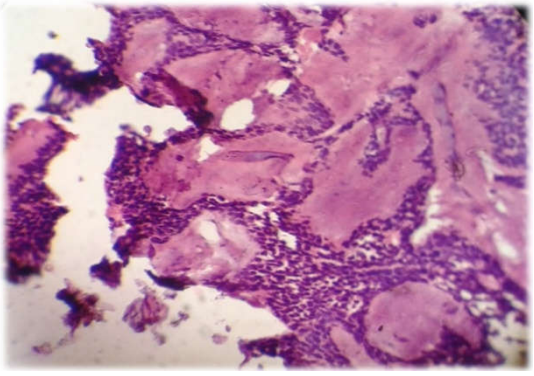

Figure 6.

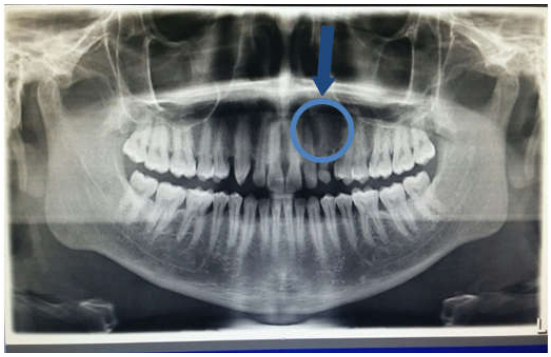

Figure 7.

DISCUSSION

CEOT is an uncommon neoplasm accounting for less than 1% of all odontogenic tumours. This rare tumour was first described as a separate pathologic entity by a Dutch pathologist Jens Jorgen Pindborg in 1955. Less than 200 cases have been reported in literature, with most being reported in the mandible. CEOT has a peak prevalence in the fourth and fifth decades, with an equal sex distribution. It has a marked preference for the mandible and most tumours arise in the molar–premolar region. 52% of the tumours are associated with an impacted tooth, most often the first or second molars (Franklin, 1976). Although Pindborg's tumour is well described in the mandible, descriptions of lesions involving the maxilla are rare. This case features a lesion in the maxilla with an impacted maxillary canine tooth and a retained deciduous tooth treated by less aggressive surgical modalities.

The typical clinical presentation of CEOT is a slowly enlarging mass that causes expansion of the affected site and is asymptomatic. When located in the maxilla, it may be associated with epistaxis, nasal stuffiness, proptosis and headache (Bouckaert *et al*., 2000). In our case the patient is asymptomatic and the diagnosis is made on routine radiological examination after thepatient reported to Dental outpatient Department with complaints of sensitivity. Radio graphically, it has a variety of appearances; 58% of CEOTs are unilocular, 27% are multilocular and 15% are nonloculated. The internal aspect frequently contains mineralized structures that appear as radiopacities (Kaplan *et al*., 2000). Radiographic features of a CEOT may overlap with several other odontogenic or nonodontogenic lesions. The CEOT is commonly associated with impacted teeth and can be confused with a dentigerous cyst which is also seen around an impacted tooth. However, the dentigerous cyst lacks mineralization within the lesion. In contrast to the dentigerous cyst which is more frequently associated with third molars, the CEOT is found around first and second molars. Ameloblastoma and odontogenic myxoma may also present as unilocular or multilocular radiolucencies, similar to the CEOT. These two common odontogenic tumours rarely demonstrate radiographic evidence of radiopacities like CEOT. Further, odontogenic myxoma often has a "soap bubble" appearance with angular trabeculae within the lesion and ameloblastoma is usually associated with root resorption in 81% of the cases. The lesion that strongly resembles CEOT radio graphically is the calcifying odontogenic cyst which presents as a mixed radiolucent radiopaque lesion and is associated with an impacted tooth (Neville, 2002; Marx, 2003). In this case that involved the maxilla, the differential diagnosis included ossifying fibroma and ameloblastic odontoma. Advanced imaging techniques play an important role in evaluating the extent of facial bones and skull involvement and has a crucial role in planning the surgery (Patiño *et al*., 2005). The CT of this tumour usually shows a well-defined mass with thinning of the cortical plates and contains scattered radiopaque foci, confirming more in terms of a CEOT.

The diagnosis of CEOT is based on histopathology. CEOTs are unencapsulated, infiltrating tumours. Epithelial cells appear polyhedral with prominent intercellular bridges having abundant eosinophilic, finely granular cytoplasm with nuclear pleomorphism and prominent nucleoli. Most of the cells are arranged in broad ramifying and anastomosing sheet-like masses with little intervening stroma.

An eosinophilic homogenous material staining like amyloid is characteristic of this tumour with concentric calcified deposits, resembling psammoma bodies called "Liesegang rings." Congo red staining with viewing under polarized light microscopy demonstrates areas of apple green birefringence. These areas depict positive staining of amyloid like substance and are highly characteristic of CEOT. Amyloid also stains positively for crystal violet and thioflavine T.

CEOT has a variable biologic behaviour ranging from very mild to moderate invasiveness (Marx, 2003). The literature shows variations regarding radicalise of the surgical treatment needed. There are very few evidence-based treatment recommendations because of the paucity of cases reported (Cross *et al*., 2000; Rapidis *et al*., 2008). Surgical procedures for treatment may include conservative enucleation, marginal resection or partial resection in larger infiltrating tumours (Cheng *et al*., 2002). In their review of 113 cases, Franklin and Pindborg suggested that marginal resection with a rim of normal tissue is advisable (Franklin, 1976). They advise against a radical surgical approach of wide resection such as hemimaxillectomy (Franklin, 1976). Surgical decision making often depends on parameters of the case such as the anatomic location of the tumour, the size and duration, histopathologic findings, patient's age, and consideration of reconstruction methods following surgical procedure (Franklin, 1976; Bridle *et al*., 2006). The appropriate treatment of the CEOT requires surgical excision with disease-free margins. In the maxilla, the CEOT tends to grow more rapidly and may infiltrate the proximal vital structures, suggesting that more aggressive surgery is required in these specific cases (Lee, 1992). In this case, the CEOT was treated conservatively via removal of the retained deciduous tooth along with surgical removal of the impacted tooth and an enucleation followed by curettage. Recurrence of the lesion is presumed to be due to inadequate removal of neoplastic tissue, which is possible, given the more conservative surgical approach and the follow up period.

No signs of recurrence were reported. Periodic radiographs was also taken to check for recurrence. Local recurrence rates of 10–15% have been reported and malignant transformation is rare, with only three cases reported (Franklin *et al*., 1976; Cheng *et al*., 2002). CEOT has a much lower recurrence rate than ameloblastoma. A follow-up of minimum 5–10 years may be necessary because of the very slow growth rate of this tumour (Franklin *et al*., 1976). In conclusion, this features a case of the CEOT in an unusual location of maxilla. It emphasizes the rapid and unconfined position of maxillary CEOT. Maxillary lesions probably need aggressive surgery in most cases than not as these tumours usually grow more rapidly than their mandibular counterparts and invade the surrounding vital structures. Treatment by surgical enucleation and proper curettage with accurate tumour free margins is needed with periodic clinical and radiographic follow up.

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