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CALCIFYING EPITHELIAL ODONTOGENIC TUMOR-UNUSUAL OCCURENCE IN ANTERIOR MAXILLA : A CASE REPORT

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ABSTRACT

The calcifying epithelial odontogenic tumor (CEOT) is a rare benign odontogenic neoplasm epithelial in origin that accounts for approximately 1% of all odontogenic tumors. Epithelial cells of Pindborg tumour arise from remnants of the cells in the stratum intermedium layer of the enamel organ in tooth development.It mostly occurs in the mandibular posterior region of jaw. The occurrence of this tumour in the anterior maxilla (an uncommon site) makes it rarer. Although the present case was asymptomatic, root resorption, swelling and displacement of adjacent teeth necessitated its surgical removal. The lesion was surgically enucleated and histopathological examination confirmed calcifying epithelial odontogenic tumour, showing abundant calcifications in the form of Liesegang rings

INTRODUCTION

Nearly 200 cases of calcifying epithelial odontogenic tumor (CEOT) have been reported in the literature (Bouckaert *et al*., 2000) since Pindborg described it as a separate pathologic entity in 1955 (Philipsen *et al*., 2000). The eponym Pindborg Tumor was first introduced to the literature in 1967 to describe this interesting and unique odontogenic tumor (Kaplan *et al*., 2001 and Houston *et al*., 1997). World Health Organization in 1992 classified it as a benign odontogenic tumour, which is exclusively epithelial in its tissue of origin. The differential diagnosis for CEOT should include adenomatoid odontogenic tumour (AOT), calcifying odontogenic cyst (COC), ameloblastic fibro odentoma (AFO) and odontoma. It is a locally aggressive but slow growing neoplasm that occurs as intraosseous (96%) and extraosseous (4%) variants (Franklin and 1976; Goode *et al*., 2004 and Reichart *et al*., 2004). The histogenesis of this tumour remains elusive. They tend to occur over a wide age range but usually predominate in the 3rd to 6th decades of life with almost equal sex predilection. The intraosseous CEOT are found primarily in the mandible (mandible: maxilla ratio 2:1), especially in the premolar and molar regions.

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Around 53% of these tumours are associated with unerupted teeth, most common being the mandibular molars. The extraosseous variant exhibits preponderance to the anterior gingival regions (Neville *et al*., 2001; Houston and Fowler, 1997). The incidence rate of calcifying epithelial odontogenic tumor is 0.4 – 1% of total odontogenic tumor from which less than 8% occurs in anterior maxillary region. CEOT are slow growing, expansible, painless masses that cause expansion of the cortical plates but occasionally patients may report with pain, epitasis, nasal stuffiness etc.

The extraosseous variant usually presents as a nodular swelling. Radiographically, they demonstrate irregular unilocular or multilocular radiolucency containing radiopaque masses of varying sizes and opacity. If an unerupted tooth is associated with the tumour, the radiopacities tend to cluster near the tooth crown (Neville *et al*., 2001; Houston and Fowler, 1997). The intraosseous variant is often easily enucleated and varies in size from 1 to 4 cm in diameter. The mass is usually greyish white in colour, bisection of which reveals multiple calcified particles which produce a crunching sound on cutting. The tumour may be solid or contain minute cystic spaces with the associated unerupted tooth being present within the tumour mass (Goode *et al*., 2004; Neville *et al*., 2001).

Case Report

A 60-year-old male reported with a swelling in upper left front region of jaw since 5-6 month. Clinical examination revealed it a painless, firm gingival swelling in the left anterior region of maxilla measuring 8 × 10 cmswhich was progressively increasing in size (Figure 1).

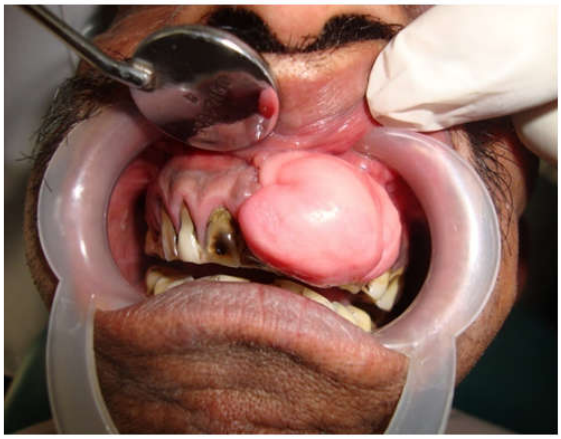

Figure 1.

The intra oral periapical radiograph revelled thinning of buccal and lingual cortical bony plates (Figure 2). Based on history, clinical examination and radiographical diagnosis a provisional diagnosis of calcifying epithelial odontogenic tumour (CEOT), adenomatoid odontogenic tumour (AOT), calcifying odontogenic cyst (COC), ameloblastic fibro odentoma (AFO) and odontoma was made. Routine laboratory parameters were normal hence incisional biopsy was taken measuring 3x2x2 cms (Figure 3).

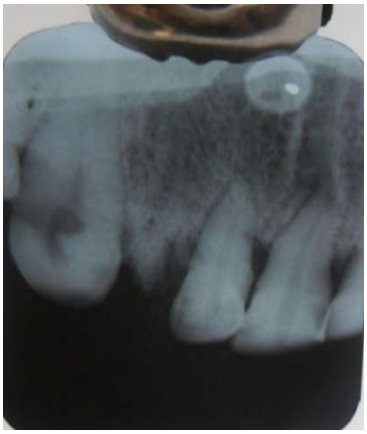

Figure 2.

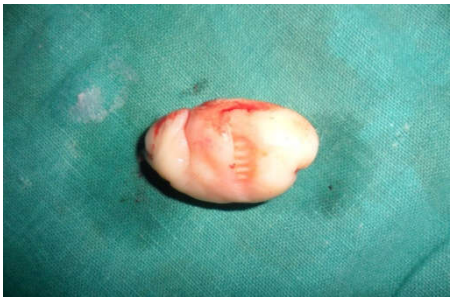

Figure 3

The microscopicexamination revealed an epithelial neoplasm composed of sheets and nests of polyhedral epithelial cells with an abundant eosinophilic, granular cytoplasm. Cellular outlines were distinct and intercellular bridges were noted. Considerable nuclear polymorphism was a frequent finding. Extracellular amyloid-like substance and calcified concentric deposits known as Liesegang rings were also identified (Figure 4).

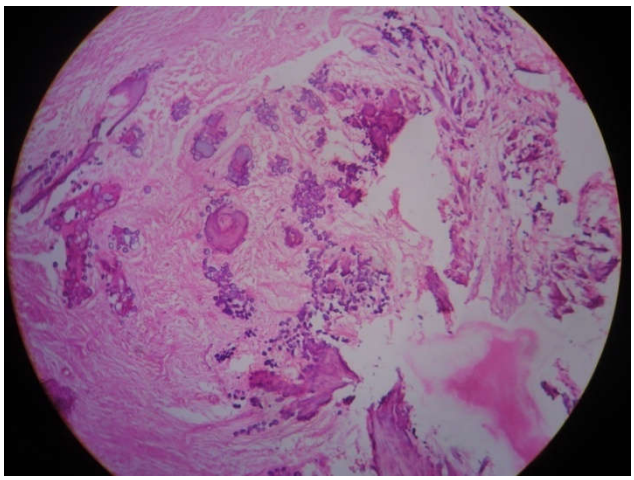

Figure 4.

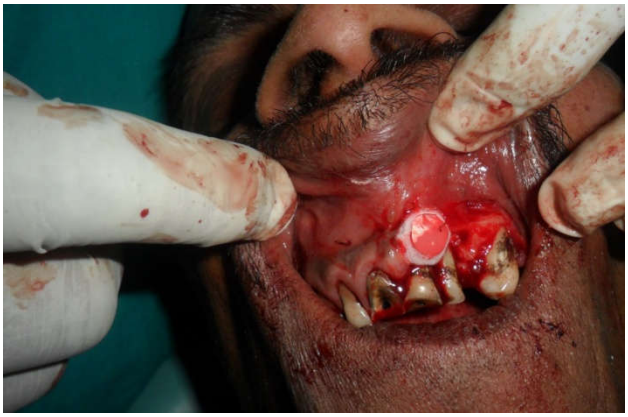

Figure 5.

The treatment plan was dependent on multiple factors such as size and location of neoplasm, general condition of patient and operator skill. Small, intrabony mandibular lesions with well-defined borders are treated by simple enucleation or curettage followed by judicious removal of a thin layer of bone adjacent to the tumour. Large tumours require aggressive approach by segmental resection, hemimandibulectomy and hemimaxillectomy, which cause bone discontinuity requiring reconstruction procedures such as grafting or distraction osteogenesis. Recurrence rate of 10−20% following conservative treatment is reported. Malignant transformation and metastasis is rare. The treatment of choice in this case was total enucleation of the lesion with a follow up of 1 year (Figure 5). The gross examination of the surgical resection revealed a cystic cavity filled with blood, focal areas of yellowish vegetations and extra-osseous tumoral mass, surrounded by a myxomatous capsule. During specimen bisection, calcified particles were frequently observed producing a crunching sound while cutting. The patient has

remained asymptomatic and experienced no recurrence during the 1 year post operative period .Thus the post operative period was uneventful.

DISCUSSION

The literature reports 67 cases of CEOT in maxillary region only 13% of the patients complained of pain or discomfort (Kaplan et al., 2001). When located in the maxilla, patients may sometimes complain of nasal stuffiness, epistaxis and headache (Goode et al., 2004; and Cheng et al., 2002) which was common in this patient. CEOT occurs most commonly between 20 to 60 years of age with mean around 40 years. In 113 cases reviewed by Franklin and Pindborg (Franklin et al., 1976) patients ranged from 8 to 92 years of age with mean at 40 years. In 2004, Cicconetti and colleagues reported that tumour more frequently affects adults in the age range of 40 to 60 years with peak incidence in the 5 Th decades with an equal sex distribution (Cicconetti et al., 2004).

This article describes an unusual lesion involving anterior maxillary region in thesixth decade of life. However, the age range of patients with CEOT varies between 8 and 92 years (mean age of 36.9 years) at the time of diagnosis (Kaplan et al., 2001) showed a female predilection of 1.5:1 in 67 cases, with peak age in the fourth and fifth decades, and lesion occurrence in different groups with a slight predilection for Caucasian individuals (Bouckaert et al., 2000). Ninety-four percent of the lesions are central and intraosseous and 6% are extraosseous. CEOT may lead to tooth tipping, rotation, migration, and/or mobility secondary to root resorption. This lesion is often symptomless and discovered on routine radiography. Alternatively it may present symptomatically as a slow-growing, painless, expansile, bony swelling with cortical bone resorption and finally perforation, as was seen in the case reported. The diagnosis of CEOT is also based on histopathological examination revealing polyhedral neoplastic cells, which have abundant eosinophilic finely granular cytoplasm with nuclear pleomorphism and prominent nucleoli. Most of the cells are arranged in anastomosing sheet like masses. An extracellular eosinophilic homogenous material staining like amyloid is characteristic of this tumour with concentric calcific deposits called Liesigang Ring (Franklin et al., 1976). The case described also depicted calcific foci in abundance with fused amorphous calcified aggregate.

Conclusion

Based on history, clinical examination, radiographical and mainly on histopathological diagnosis (the section shows sheets of polyhedral cells with prominent intercellular bridges and calcification in the form of liesegang ring) confirmed it to be a case of calcifying epithelial odontogenic tumour ofextraosseous type.

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