CALCIFYING EPITHELIAL ODONTOGENIC TUMOUR – CASE REPORT AND REVIEW OF THE LITERATURE

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+ SP Khoo BDS, MSc (Lond), FFDRCSI
* N Jalil BDS, FFDRCSI
+ HB Yaacob BDS, MSc(Lond), FDSRCPS(Glasg.), FDSRCS (Eng.), MBiol

+ Department of Oral Pathology, Oral Medicine & Periodontology
Faculty of Dentistry
University of Malaya
50 603 Kuala Lumpur.

* Dental Specialist, Queen Elizabeth Hospital
Kota Kinabalu, Sabah, Malaysia.

Correspondences to Dr. SP Khoo+.

ABSTRACT

The calcifying epithelial odontogenic tumour (CEOT) is a rare benign odontogenic neoplasm which was first described by Pindborg in 1955. It accounts for less than 1% of all odontogenic lesions. A case of CEOT which presented together with cardiac abnormalities is presented here and the literature of CEOT is reviewed.

Key words:- Calcifying odontogenic tumour, cardiac.

INTRODUCTION

The calcifying epithelial odontogenic tumour (CEOT) is a rare benign odontogenic neoplasm. It was first described by Pindborg as a distinct entity in 1955(1) and accounts for less than 1% of all odontogenic tumours(2). Clinically it manifests as a bony lesion, accounting for the majority of cases or as the extremely rare peripheral type. Numerous terminology have been given to this lesion, such as calcifying ameloblastoma(3), malignant odontoma(3), adamantoblastoma(1), adenoid adamantoblastoma(4), cystic complex odontoma(4), uncommon ameloblastoma with calcifications(1) and others.

The purpose of this article is to report an additional case of the CEOT and to review the literature of this lesion.

CASE REPORT

A 16 year old girl was seen for consultation because of complaints of loose teeth on the lower left quadrant and had requested extraction of those teeth. She had noticed gradual mobility of those teeth over a period of three months. However she only noticed the swelling in the left mandible only three weeks before presenting at the dental clinic. No pain nor numbness was associated with the swelling.

Upon examination there was only very minimal facial asymmetry. There was no lymphadenopathy. Clinically, she presented with a full complement of teeth on both jaws except for all four third molars and a lower left second premolar. She had grossly carious lower right and left first molar. The lower left incisors, canine and second molar were of Grade 1 mobility whilst her lower left first premolar was of Grade II mobility.

A bony hard and firm swelling was noted at the buccal sulcus extending from the lower second incisor to the first molar region. A lower left second premolar was missing and a retained root of lower left first molar was noted. The colour of the mucosa covering the area was normal. There was no ulceration. Her mouth opening was normal.

A dental orthopantomogram revealed an unerupted lower left second premolar associated with a radiopaque mass. The latter was enclosed within a well-defined radiolucent area. The whole lesion extended from the lower left canine to the first molar region(Figure 1).

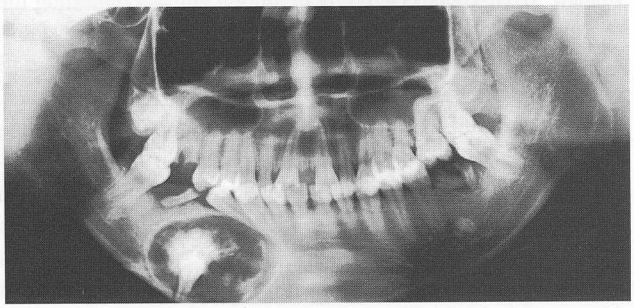

Figure 1. Orthopantomograph of the lesion showing the mass on the lower left mandible in the premolar-molar region, associated with an unerupted premolar.

Her medical history revealed a Congenital Heart Disease for which she was on follow up in Medical Unit. She however, absconded in 1990. Subsequent to the dental examination she was referred to the Medical Unit Specialist where an echocardiogram revealed the following cardiac defect :- a grossly dilated right atrium and right ventricle, a grossly dilated pulmonary artery, a large atrial septal defect and a functional tricuspid regurgitation.

Prophylactic antibiotics were given pre-operatively and the lesion was aspirated. However the findings were negative. A biopsy was carried out. A provisional diagnosis of a calcifying epithelial odontogenic tumour or calcifying odontogenic cyst was made.

The initial histopathological report was made by a General Pathologist and was reported as dentigerous cyst. As the report did not concur with the clinical findings a separate opinion sought.

Histopathologically the tumour was composed of sheets of epithelial cells with a fibrovascular stroma. The epithelial cells were polyhedral in shape and had eosinophilic cytoplasm. Intercellular bridges were present. No mitotic figures were noted throughout the specimen.

Regions of amorphous and eosinophilic material were seen within the sheets of epithelial cells. Calcifications in the form of Leisegang rings were also present. The appearances were consistent with a calcifying epithelial odontogenic tumour (Figure 2).

An operative procedure was subsequently carried out under general anaesthesia with antibiotic cover to remove the lesion. Other retained roots were extracted at the same time. The lesion was totally removed and the area of operation was packed with bismuth iodide pack ribbon gauze. She was then subsequently followed up. Monthly follow-up showed progressive uneventful healing. A 24-month follow-up revealed no evidence of recurrence of the lesion.

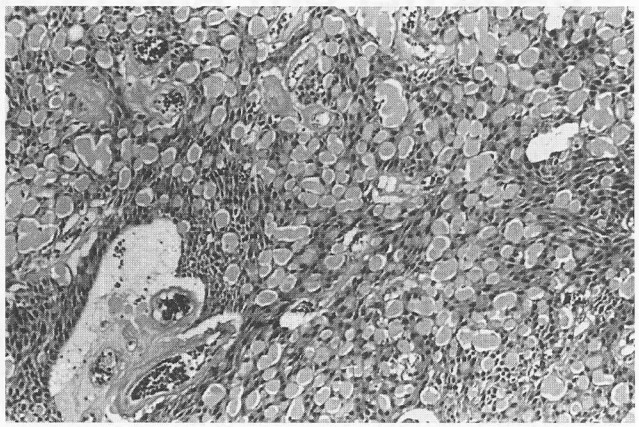

Figure 2. Photomicrograph showing epithelial sheets and demonstration of possible amyloid-like substance between the cells. (Haematoxylin & Eosin) X 200.

DISCUSSION

The calcifying epithelial odontogenic tumour (CEOT) is a rare benign odontogenic neoplasm which was first described by Pindborg as a distinct entity in 1955 (1). It accounts for 0.2% to 1% of all odontogenic tumours (2). Since its initial description, the number of cases in the English literature has totaled 187. There is agreement among authors regarding the clinical presentation of this tumour. In the most comprehensive review of the literature by Franklin and Pindborg (2), 113 cases have been studied.

The tumour is basically one of adulthood. The mean age of occurrence is around 40 years. Chaudhry and his colleagues (5) reported the average age of patients with intraosseous lesions was 45 years whereas those with extraosseous tumour was 38 years. The youngest patients both with extraosseous lesions were a 12 year old boy and a 16 year old girl(6,7). The oldest patient was 92 years old (8).

There is no sex predilection. In reported cases that mentioned race, the great majority of patients were Caucasians. There have only been few reported cases in blacks (5-7).

There is a predilection for occurrence in the premolar-molar region of the mandible. Approximately two-thirds were located there (6,8). The CEOT may occur as intraosseous (87.8%) tumours or as extraosseous tumours(6.1%) with the former presenting most often in the mandible (premolar-molar area) whilst the latter in the anterior part of the jaws (11). The CEOT has also been reported as hybrid tumours (10-12) in combination with adenomatoid odontogenic tumour (AOT) which has been reported to be frequent in women and present at a younger age

Association with impacted teeth was difficult to ascertain, since this information was not uniformly recorded. It would appear that less than half of the reported tumors have developed in close proximity to an unerupted tooth. The typical clinical presentation is a slowly enlarging intraosseous mass which causes expansion of the affected mandible and is asymptomatic.

The varying radiographic features have been described as a) pericoronal radiolucency; b) pericoronal radiolucency with small radiopacities;c) mixed radiolucent-radiopaque lesion not associated with an unerupted tooth; d) "falling-snow" appearance ; e) dense radiopacity. The two commonest presentations were found to be pericoronal radiolucency with diffuse radiopacities in a radiolucent area (2). This makes the clinical diagnosis somewhat difficult due to the similarity with dentigerous cyst. For extraosseous CEOT, bone adjacent to the tumour shows a superficial erosive pattern.

The histomorphologic patern of the CEOT consists of scanty connective tissue stroma that supports clusters of polyhedral epithelial cells with eosinophilic cytoplasm. The nuclei vary in size and staining quality, producing a pleomorphic appearance. Intercellular bridging is present. Small, usually round calcifications with Leisegang rings are present among the epithelial cells and also in the connective tissue. Besides these characteristic features the presence of homogeneous substance that has been variously described as amyloid (13,14), comparable glycoprotein (14), or keratin or enamel matrix(5). A clear cell variant of CEOT has also been described(6,8,15).

The clinical differences among the CEOT types have been thought to be attributed to their origin (10,16,17). It has been shown that the intraosseous CEOT is derived from the stratum intermedium of the enamel organ. In contrast the extraosseous form arises from the dental lamina epithelial rests in gingiva and/or basal cells of the gingival surface epithelium. With the hybrid tumour between CEOT/AOT, the AOT portion arises from all three components of the enamel organ (preameloblasts, stellate reticulum, stratum intermedium(10).

The CEOT is generally considered a benign tumour. The clear cell variant is thought to be more aggressive in behaviour(18,19) Metastasis of this lesion however, has been reported by Basu and colleagues (20). They reported lymph node involvement and evidence of soft tissue invasion.

The management of the CEOT, is largely dependent on the size, site of the lesion and the amount of bone destruction. In the mandible, the recommended surgical approach is enucleation with vigorous curettage in the early stages but with more advanced bone infiltration, resection of the tumour with a normal margin of bone have been advocated(2,8,21). These authors believe that maxillary lesions should be treated more aggressively because they grow faster and because of the proximity to important structures. They also stated that the more amyloid tissue and calcification there is, the less aggressive is the lesion.

The case presented here showed a well-defined radiographic lesion and was completely enucleated. It had been erroneously diagnosed as a dentigerous cyst due to, in part, a misrepresentation of the biopsy specimen sent for examination. However, the histological appearance of the lesion was typical of a CEOT. A large amount of amyloid tissue and calcification was present in the lesion. A conservative approach was adopted. Although various approaches have been suggested by some authors we believe that the treatment should be individualized for each lesion because the radiographic and histologic features may differ from one lesion to another. The case reported here also presented with gross cardiac abnormalities. The authors are not aware of any cases documented in the literature which had an accompanying cardiac abnormality. The management of this patient, thus required close monitoring by the physician and antibiotic cover was necessary.

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