Ameloblastoma Proliferating Type Calcifying Cystic Odontogenic Tumor: A Case Report

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ABSTRACT

Calcifying odontogenic cyst(COC) is considered as a rare entity and comprises of 1% of Jaw cysts. It is a developmental odontogenic cyst first described by Gorlin et al. in 1962. There has always been a dilemma about its nature as a cyst or a neoplasm. As large number of reports has been published, it was proposed that COC was indeed a heterogeneous group of lesions with distinct histopathological findings. WHO classification in 2005 has modified the term COC to Calcifying cystic odontogenic tumor (CCOT). Here, we report a case of CCOT with proliferating ameloblastoma in a 16-year-old female in the left mandibular molar area.

KEYWORDS: Ameloblastic proliferation, Calcifying Odontogenic cyst, Neoplasm

INTRODUCTION

Gorlin et al, first described Calcifying odontogenic cyst (COC) as a specific odontogenic lesion. Since then controversy and confusion, have existed regarding the relation between non-neoplastic, the cystic lesion and solid tumor masses that share a common cellular and histomorphologic features.[1]

COC, first described by Gorlin and colleagues, are now included in the group of odontogenic tumours in a World Health Organization (WHO) international classification proposed in 1992, as a tumor of odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation.[2] This diversity led to a "dualistic" concept which graded COC as a cyst and neoplasm and renamed it as Calcifying cystic odontogenic tumor (CCOT).[3]

CCOT has occurrence constituting about 0.37% to 2.1% of all odontogenic tumors.[4] It is well known that this lesion often occurs in association with odontogenic tumours such as complex odontoma and ameloblastoma, and all recent histopathological classifications of CCOTs advocate categorizing the variants associated with these tumours.[2] Malignant transformation of COC has also been reported.[5] Although the association of ameloblastoma with this lesion is important, there are only a few reports that detail the clinical and radiographic features of CCOTs associated with ameloblastoma.[2]

In this paper, we report a case of ameloblastoma proliferating type CCOT that occurred in a young female in mandibular molar ramus area.

CASE REPORT

A 16 year old female patient reported to the Department Oral Medicine and Radiology with acomplaint of pain and swelling on the left side of the face since 3 months. Patient noticed a swelling on the left side of the face which gradually increased to attain the present size. Pain was associated with the swelling which was dull, intermittent and localized.

The clinical examination revealed a single diffuse bony swelling on the left side of the face. It was hard, tender on palpation, margins were well defined with a smooth surface(Figure 1). Intraorally swelling caused obliteration of buccal vestibule. It had a well defined smooth surface, was fixed to the underlying bone leading to thinning of lingual cortical plate and cortical expansion. Submandibular lymph nodes on the left side were palpable, enlarged, soft, mobile and tender.

Panoramic radiograph (OPG) revealed a unilocular radiolucency distal to 36 extending along the ramus to coronoid process of mandible, measuring about 4 x 3 cm. The radiolucency was well defined surrounded by sclerotic border. Cortical expansion with thinning of cortex was evident. Cortical expansion of the lower border of the mandible was noted. 37 was partially formed and pushed to the inferior border of the mandible below the root apex of 36 (figure 2).

3D construction view of CBCT showed hollowing of the entire ramus on the left side from distal of 36 extending to the coronoid process. Inferior border of mandible was intact but the superior border showed discontinuity. 37 on

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left side was pushed to the inferior aspect of the mandible(Figure 3).

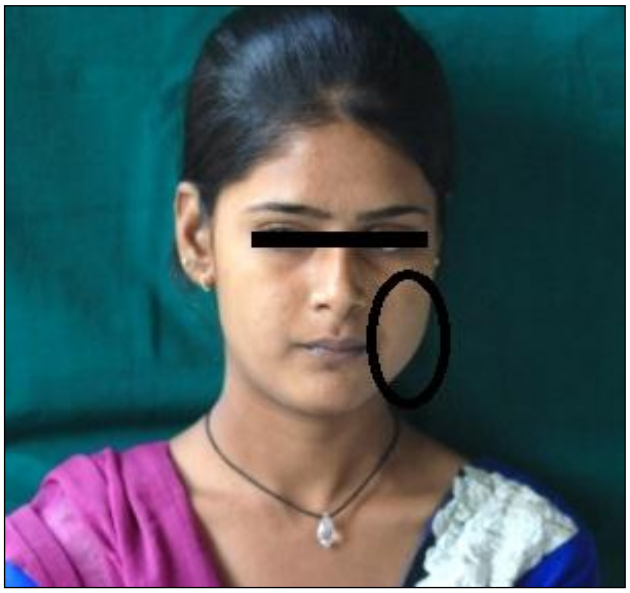

Figure 1.Extra-oral photograph

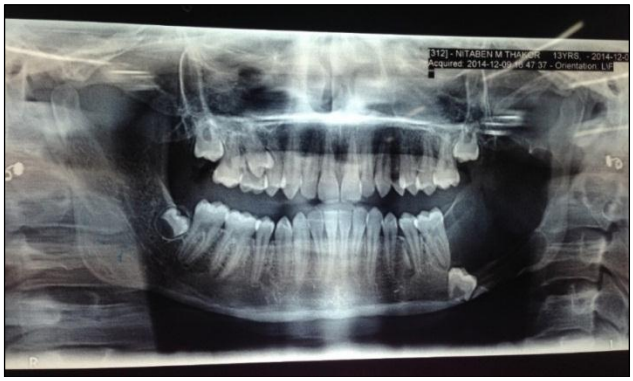

Figure 2. OPG showed unilocular radiolucency on the left side of mandible extending distal to 36 and involving the ramus and coronoid process.

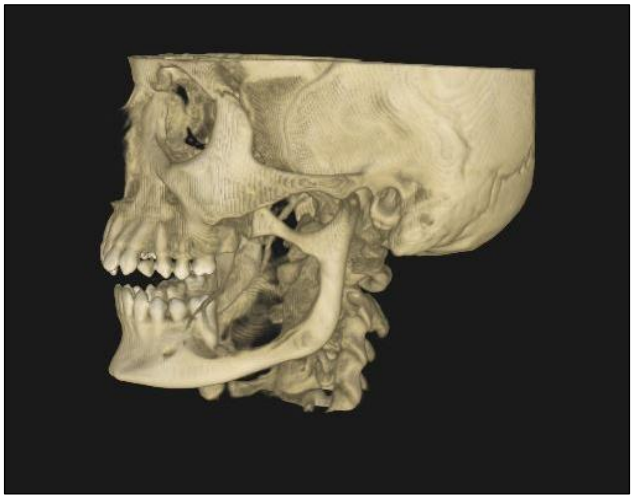

Figure 3. 3D reconstruction view of the left side of the mandible showed hollowing of the entire ramus area extending from distal to 1st molar teeth to the coronoid process.Inferior border of mandible was seen intact with the superior border appearing discontinuous.

Incisional biopsy was performed. The microscopy revealed a cystic lining comprising of columnar to cuboidal basal cells and suprabasal stellate reticulum like cells. Squamous metaplasia and ghost cells were seen. With the above histopathological features diagnosis of CCOT was given (Figure 4).

The lesion was surgically enucleated along with the associated tooth (37) under general anesthesia. The excised specimen was sent for further histopathological examination. Excised lesion was cystic, measured about 6 × 3.5 cm and was attached at the cemento-enamel junction of 37.(Figure 5) Histopathological examination

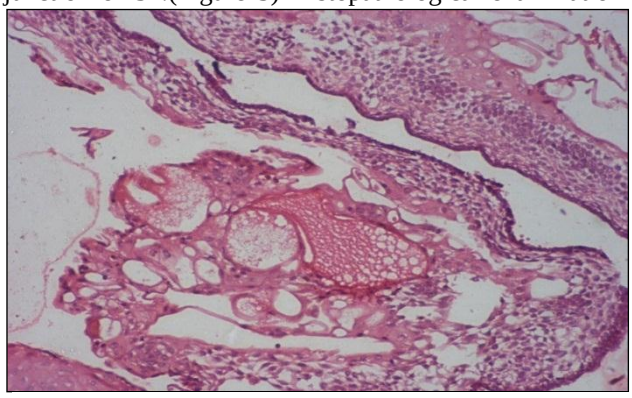

Figure 4. 4X- H&E section showing cystic lining with basal cuboidal cells along with superficial stellate reticulum like cells and with ghost cells at the superficial part of the lining.

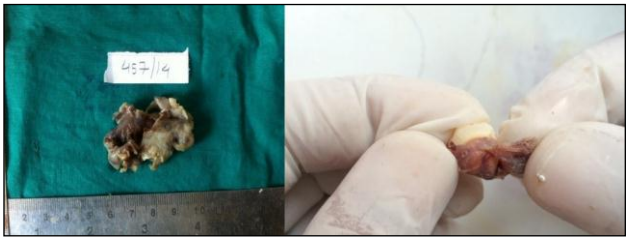

Figure 5. Main excised lesion- Grayish white in color, firm in consistency, measuring about 6 X 3.5 cm. Tooth specimen - mandibular 2nd molar attached with soft tissue at cemento-enamel junction.

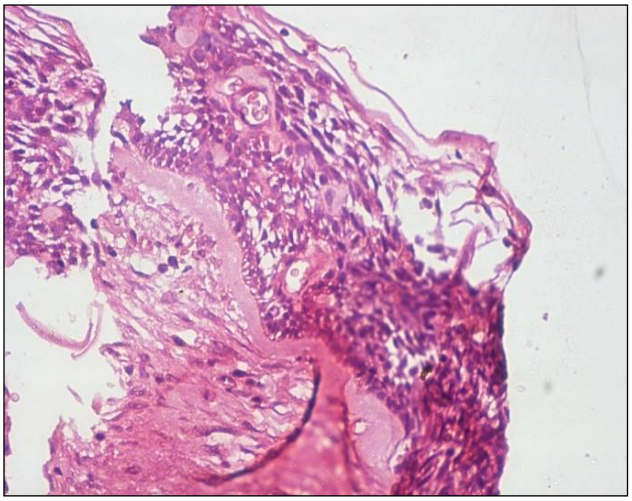

Figure 6. 10X- H&E section showing cystic lining with basal cuboidal and stellate reticulum like cells along with ghost cells at the superficial part of the lining and juxta-epithelial dentinoid like material.

of excised tissue reveled cystic lining of stratified epithelium with cuboidal to columnar basal cells few showed reverse polarity. Suprabasal cells were stellate reticulum like and few areas showed squamous metaplasia. Ghost cells were seen in few areas. Juxta-epithelial dentinoid like material was seen. Stroma showed thick collagen with hyalinization and numerous odontogenic follicles. A final diagnosis of ameloblastoma proliferating type CCOT was given (Figure 6-8).

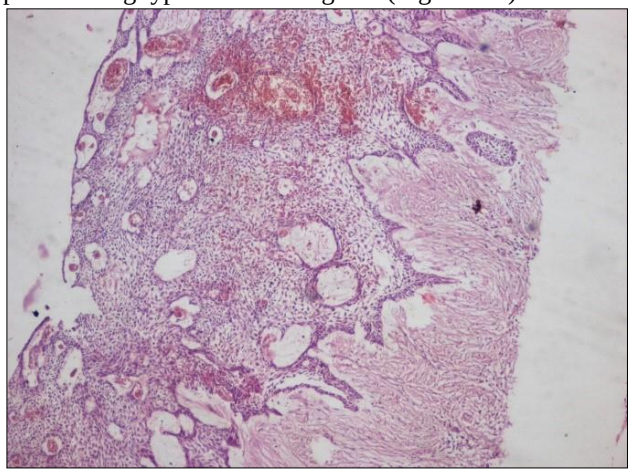

Figure 7. 10X H&E section showing proliferating cystic lining and odontogenic island within the capsule.

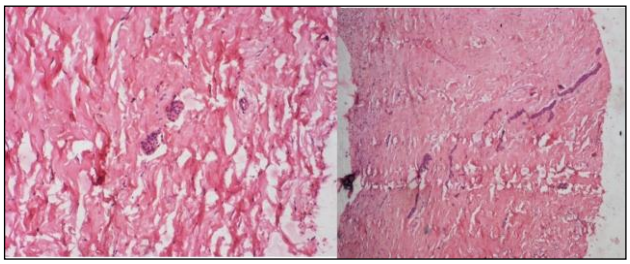

Figure 8. 10X H&E sections showing odontogenic epithelial strands in fibrous stroma.

DISCUSSION

In 1971, the WHO classification of odontogenic tumors defined COC as "a non-neoplastic cystic lesion in which the epithelial lining shows well defined basal layer of columnar cells, and superficial squamous cell resembling stellate reticulum. Ghost cells are found in the epithelial lining or with in the fibrous capsule. The ghost cells may become calcified. Dysplastic dentin may be laid down next to basal layer of epithelium."[5]

The COC is an uncommon lesion that demonstrates considerable histologic diversity and presents variable clinical behaviors. Although, it is widely considered to represent a cyst, some investigators prefer to classify it as a neoplasm.[6] In 1992, the WHO classified this lesion with odontogenic tumors and named it as CCOT.[3] Some CCOTs appear to represent non-neoplastic cysts; other members of this group, variously designated as dentinogenic ghost cell tumors or epithelial odontogenic ghost cell tumors, having no cystic features, may be infiltrative or even malignant, and are regarded as neoplasms.[6]

Since it has been accepted that CCOT occurs as two different lesions- cystic and solid variants, both having different prognostic significance.[5] Several classification of CCOT subtypes have been proposed, but most of them have limitation in separating cystic and neoplastic variant.[7]

Most of the CCOTs are seen as unilocular radiolucent lesions. Only 5-13% of the cases show multilocular appearance.[8]

CCOT has been reported to coexist with other odontogenic tumours, such as ameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma and so on. Among them, ameloblastoma may be one of the most important tumour in terms of its histology and prognosis and all recent histological classifications have established a category for the variant of CCOT associated with ameloblastoma.[2]

In the present case, although the basal cells showed ameloblastic proliferative activity and odontogenic islands in cystic stroma, they did not completely meet the histopathologic criteria of early ameloblastoma as suggested by Vickers and Gorlin.[9] Hence, the case was diagnosed as ameloblastoma proliferating type CCOT.

S Iida et al found a case of CCOT with solid parts of ghost cells and ameloblastomatous proliferation seen in the connective tissue of the cyst wall.[2] Hong et al reported 92 cases of CCOT from the files of AFIP registry of Oral Pathology, out of which only 11 cases (14%) were ameloblastomatous CCOT.[5] Kamran Nosrati et al also reported a case of CCOT which showed acanthomatous islands in the connective tissue of lining of the cyst.[6]

A simple unicystic CCOT is characterized by well-defined darkely stained basal cells, an overlying layer of stellate reticulum like cells and few ghost cells that may or may not show calcification. Juxtaepithelial dentinoid formation seen occasionally. However, it may be difficult to distinguish ameloblastomatous CCOT from ameloblastoma arising from CCOT (ameloblastoma ex CCOT).[5] The multicystic CCOT and odontogenic ghost cell tumor can be clearly distinguished by the inductive activity seen around the odontogenic epithelial proliferation and formation of ghost cells. This inductive effect of the tumor also distinguished the CCOT from ameloblastoma ex CCOT.[5] Keeping this in mind we classified the lesion as ameloblastoma proliferating type CCOT. Ameloblastoma ex CCOT occurs intraosseously, appearing as cyst like, radiolucent lesion. Whether these tumors are potentially destructive as typical ameloblastoma and have the propensity for reoccurrence is unknown. Buchner suggested that if CCOT is associated with an ameloblastoma, its behavior and prognosis would be the same as an ameloblastoma, not CCOT.[7]

CCOTs occurs usually as an intraosseous (70%) and as extraosseous (16-22%) lesions. They are seen in

individuals over 50 years of age. About 50% of CCOT have been reported as being associated with an unerupted tooth. Displacement of teeth is often seen. Resorption of the roots of the adjacent teeth is a frequent finding and is regarded as an important radiological feature. Local expansion sometimes occurs, and perforation of the cortical plate, when present, may be radiologically demonstrable.[10]

In the case reported by S Iida et al, the lesion was associated with the unerupted lower second molar which was displaced inferiorly to the position below the first molar.[2] This was in accordance to the present case.

The site predilection of CCOT is mostly in mandibular molar area and expand in anterio-posterior direction. Some case showed root resorption of teeth of affected area. In the present case, the lesion also showed intact inferior border of mandible but superior border was discontinuous with involvement of coronoid process.

The treatment of cystic lesion involves enucleation withlong-term follow-ups. Recurrence depends on completeness of cyst removal. Prognosis is good for cystic CCOT and less certain for neoplastic CCOT. The CCOT may be associated with other odontogenic tumours for that the treatment and prognosis is based on the associated tumors.[4] After complete surgical excision of the reported case patient was followed up for six months with fair prognosis and no recurrence.

CONCLUSION

Ameloblastoma proliferating type CCOT is a rare histolopathological variantof all CCOTs. The need of the hour is proper categorization of the other variants of CCOT according to clinical, radiological, and histopatholical characersitic for better understanding of pathogenesis of each variant.

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