A rare variant of calcifying odontogenic cyst with ameloblastoma presentation

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SUMMARY

Calcifying odontogenic cyst is a rare entity which was first described by Gorlin, and also accounts for 1% of the jaw cysts according to Shear. Due to its diverse histopathology and variable clinical features, there has been a doubt regarding its nature as a cyst or a neoplasm. In this report we present a case of calcifying odontogenic cyst with mural ameloblastomatous presentation in the left body of the mandible in a 19-year-old male patient.

Keywords: calcifying odontogenic cyst, ameloblastoma.

INTRODUCTION

Calcifying odontogenic cyst (COC) is a rare developmental odontogenic cyst, first described by Gorlin in 1962 (1). According to Shear, it accounts for 1% of jaw cysts (2). COC has been classified under two basic groups, namely cysts and neoplasms. The cystic type of COC comprises the majority of the cases, which are characterized by an uncystic lesion associated with or without an odontoma. They may also show ameloblastomous proliferative activity intraluminally or intramurally (3). Malignant transformation of COC has also been reported (4). The aim of this report is to present the clinical radiological and histopathological features of a calcifying odontogenic cyst with mural ameloblastomatous proliferation which has been rarely reported.

CASE REPORT

A 19 yr old male patient visited our department with pain on the left side of the jaw since 4 months. It was also associated with an extra oral swelling which was diffuse, 3 cm below the altralagral line.
revealed odontogenic epithelium with tall columnar basal cells resembling ameloblasts having reverse polarity & overlying epithelium resembling stellate reticulum. Epithelium shows abberant keratinization & induction activity at epithelial connective tissue interface. Odontogenic epithelium shows proliferation into underlying connective tissue (Mural Proliferation). Connective tissue is collagenous & shows few small odontogenic epithelial islands, bone trabeculae and blood vessels, suggestive of calcifying odontogenic cyst with mural ameloblastomous proliferation (Fig. 6).

**DISCUSSION**

In 1971, the World Health Organisation described COC as “non neoplastic cystic lesion in which the epithelial lining shows a well defined basal layer of columnar cells, an overlying layer that is often many cell layers thick that may resemble stellate reticulum and masses of ghost cells that maybe in the epithelial cyst lining or in the fibrous capsule (5). Calcifying odontogenic cyst occurs intraosseously or extraosseously, with intraosseous being more predominant. Prior to separation of this entity by Gorlin et al, it was often regarded as some form of ameloblastoma (1). The COC is an uncommon lesion demonstrating considerable histologic diversity and presenting with variable clinical behaviors. Although, it is broadly considered to represent a cyst, some investigators prefer to classify it as a neoplasm (6). The question concerning the nature of the cyst appeared to be clarified by Toida, who recently categorized COC into a cyst and neoplasm (7). In the new classification of World Health Organisation(2005), the term calcifying cystic odontogenic tumor was replaced by calcifying odontogenic cyst (COC) which constitutes a benign cystic neoplasia presenting an epithelium with ghost cells which may display calcification in it (8).

According to Praetorius et al., the cystic lesion can be divided into three basic types simple unicys-
tic type, unicystic odontoma producing type, and unicystic ameloblastomous producing type (9). Microscopically ameloblastomatous COC resembles unicystic ameloblastoma except for the ghost cells and calcifications within the proliferative epithelium. Ameloblastomatous COC occurs only intraosseously (10). Ameloblastoma ex COC designates an ameloblastoma which arises from the cyst lining of COC (11). It can also occur intraosseously, appearing as cyst-like radiolucent lesion. Whether these tumors have the same disruptive potential and tendency for occurrence as a typical ameloblastoma is unknown (12).

Calcifying odontogenic cyst with mural ameloblastomatous presentation is a rare histologic variant. Our case did not show any signs of recurrence after surgery, but it is clear that periodic postoperative observations are essential for COC’s associated with ameloblastoma.

Fig. 5. Submentovertex view of the patient showing lateral expansion of mandibular cortex

Fig. 6. Axial CT section showing an expansile lytic lesion in the body of the mandible

Fig. 7. H&E, 40X, Histopathological sections showing odontogenic epithelium and mural proliferation

Fig. 8. Post treatment orthopantomograph

REFERENCES


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