Calcifying odontogenic cyst of mandible: A case report


ABSTRACT

Introduction: Calcifying odontogenic cyst was first described by Gorlin et al. in 1962. WHO defined it as “a cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cells thick and that may resemble stellate reticulum, and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule” Calcifying odontogenic cysts are 1.6% of all central odontogenic tumors. These are relatively rare odontogenic tumors occurring in the posterior mandible.

Case Report: This report emphasizes a unique case of a 55-year-old male suffering with pain in lower right molar region and the management of the case.

Conclusion: Even though the recurrence rate of calcifying odontogenic cyst is very rare, there are some case reports of development of ghost cell odontogenic carcinomas from calcifying odontogenic cyst. Our follow-up radiograph did not show any signs of recurrence. However, a clinical study with larger number of cases and long-term follow-up is required.
Calcifying odontogenic cyst of mandible: A case report


Abstract

Introduction: Calcifying odontogenic cyst was first described by Gorlin et al. in 1962. WHO defined it as “a cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cells thick and that may resemble stellate reticulum, and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule.” Calcifying odontogenic cysts are 1.6% of all central odontogenic tumors. These are relatively rare odontogenic tumors occurring in the posterior mandible. Case Report: This report emphasizes a unique case of a 55-year-old male suffering with pain in lower right molar region and the management of the case. Conclusion: Even though the recurrence rate of calcifying odontogenic cyst is very rare, there are some case reports of development of ghost cell odontogenic carcinomas from calcifying odontogenic cyst. Our follow-up radiograph did not show any signs of recurrence. However, a clinical study with larger number of cases and long-term follow-up is required.

Keywords: Calcifying odontogenic cyst (COC), Calcifying cystic odontogenic tumor (CCOT), Ghost cell odontogenic carcinoma (GCOC)

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Introduction

In 2005, calcifying odontogenic cyst was classified as a tumor and designated as a calcifying cystic odontogenic tumor (CCOT) by the World Health Organization. Calcifying odontogenic cyst develops from the reduced enamel epithelium or remnants of odontogenic epithelium. Calcifying odontogenic cysts can occur at any age starting from first decade to seventh decade of life. It occurs more commonly in the second decade of life.
life. It occurs almost equally in both sexes with slight male predilection. The most common sites of occurrence are anterior maxilla 41.2%, posterior mandible 35.3%, anterior mandible (17.6%), and posterior maxilla (5.9%) [1–5].

CASE REPORT

A 55-year-old male reported with pain in the lower right molar region since one month. Pain was insidious in onset, continuous, throbbing type and localized. Intraoral examination revealed missing 38, 48. On palpation, there was tenderness over right mandibular ramus region. Orthopantomogram revealed a well circumscribed unilocular radiolucency in the right mandibular body and angle region, extending from mesial aspect of 46 to 48 which is impacted (Figure 1). Root resorption of 46 and 47 was seen. Endodontically treated 14, 16, 17, 24, 25, 36 and impacted 48. Aspiration has shown serosanguineous fluid. Based on these clinical and radiographic findings a tentative diagnosis of unicystic ameloblastoma or Keratocystic odontogenic tumor was attained.

Surgical enucleation and curettage of the lesion was planned under general anesthesia. Crevicular incision was placed from 45 to 47 and extended onto the ascending ramus of the mandible. Full thickness mucoperiosteal flap was reflected, extraction of 46 and 47 was performed. There was thinning of buccal cortical bone. De-roofing of the cyst was done to expose the cystic cavity. Careful enucleation and curettage of the lesion was performed (Figure 2). Impacted 48 were removed and the entire bony cavity was thoroughly irrigated with hydrogen peroxide and saline. Enucleated tissue specimen was sent for histopathological examination. Postoperatively wound healing was satisfactory and orthopantomogram at sixth month follow-up visit revealed new bone formation with no recurrence (Figure 4), still the case is under follow-up.

HISTOPATHOLOGY

The cystic luminal epithelium is non-keratinized stratified squamous type with luminal proliferations. The epithelium also shows numerous amorphous structures of various size and shape with well-defined borders without nucleus resembling ghost cells are seen. The eosinophilic dentinoid like material and basophilic round to irregular calcified masses are also seen. The cells in spinous layer show intercellular edema with intact desmosomal attachments. The basal cells are cuboidal to columnar with palisading arrangement and budding into underlying connective tissue at focal areas. The connective tissue wall shows densely arranged collagen fibers with mild inflammatory cell infiltrate predominantly lymphocytes. Suggestive of ‘calcifying odontogenic cyst’ (Figure 5).

DISCUSSION

Calcifying odontogenic cysts are usually non-neoplastic with cystic features but, sometimes they appear as solid mass with neoplastic features. The solid lesions are named dentinogenic ghost cell tumor (DGCT), epithelial odontogenic ghost cell tumor (EOGCT). There has been confusion in classification and nomenclature of the lesion [6], because of its diversified histopathological features.
Praetorius in 1981 classified calcifying odontogenic cysts into a cyst entity and a neoplastic entity. The cystic entity was classified into three types [7].

**Type 1:** A simple monocystic type of typical Gorlin cyst, with or without dentinoid calcified tissue

**Type 2:** Monocystic odontoma creative type, presence of ameloblastic fibroma tissue in the cystic wall extending into the surrounding tissue with all the characteristics of the previous type, except that the hard tissue was complex or compound odontoma

**Type 3:** Monocystic ameloblastomatous proliferating type with ameloblastomatous proliferation both in the walls and in the lumen, and dentinoid formation.

Calcifying odontogenic cyst presents as central lesion occurring intraosseously or as peripheral lesion occurring in the soft tissue. The more common are the central lesions [7, 8]. Clinically, Calcifying odontogenic cyst appears as a completely asymptomatic swelling with expansion of cortical plates [5]. In our case, the lesion is central with expansion of buccal cortical plate and the cyst was asymptomatic. Radiographically calcifying odontogenic cysts are seen as a unilocular or multilocular radiolucencies with well circumscribed borders. Unilocular appearance is more common than multilocular appearance. Multilocular appearance accounts for 5% to 13% of all lesions [7, 8]. Calcifications may appear as small opacities giving 'salt and pepper type of pattern' or may even show large solid amorphous masses [5].

In a case series of 11 cases published by Seiji lida et al. ten cases were associated with an unerupted tooth. Adjacent tooth displacement was observed in five cases and root resorption of adjacent teeth was observed in four cases [6]. In the present case the lesion appeared unilocular with well circumscribed borders. It was associated with unerupted 48 and there was resorption of roots in relation to 46,47.

Management of calcifying odontogenic cyst is usually surgical enucleation and curettage [7, 5].

Neoplastic variants of calcifying odontogenic cyst may require an aggressive surgical approach as they have the malignant transformation potential. However, the specific diagnosis is obtained only after histopathological examination.

Recurrence rate of calcifying odontogenic cyst is very rare, only eight cases of recurrence were noted in literature [7].

Ghost cell odontogenic carcinoma is malignant counterpart of calcifying odontogenic cyst, which is rare to occur. Utaroh Motosugi et al. noted ghost cell odontogenic carcinoma arising from calcifying odontogenic cyst in three cases out of 122 cases of calcifying odontogenic cyst [9]. So far 30 cases of ghost cell odontogenic carcinoma have been reported in literature [9]. We opted for enucleation of lesion and thorough curettage of bone cavity.

**CONCLUSION**

Even though the recurrence rate of calcifying odontogenic cyst is very rare, there are few case reports of development of ghost cell odontogenic carcinomas from calcifying odontogenic cyst. Our follow-up radiograph did not show any signs of recurrence. However, a clinical study with larger number of cases and long-term follow-up is required.

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**Author Contributions**

Reddy GV – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Javeed Akhtar Ankolvı – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Arvind U.D. – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Irfan Ali Motiwala – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Phanitej G. – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

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Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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