Calcifying Epithelial Odontogenic Cyst in Maxilla: A Case Report

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The present article reports the case of a calcifying odontogenic cyst Type I variant in the maxilla of a 65-year-old woman. The lesion is unusual in that it has features of a cyst but also has many characteristics of solid neoplasm. It is classified into 2 types Type I-cystic variant and Type II-solid tumor variant. The calcifying odontogenic cyst may present some problems of differential diagnosis, both clinically and histopathologically. The histopathologic examination showed that structures characteristic of calcifying odontogenic cyst. The lesion was benign. After surgical removal of the cyst, the wound healed uneventfully, and the region was free of symptoms.

Keywords: Anesthesia, Odontogenic cyst, Tumor

INTRODUCTION

Calcifying odontogenic cyst is a rare lesion of the jaws. Gorlin et al.1 first described the condition. Gold2 recommends more descriptive term of calcifying and keratinizing odontogenic cyst. The condition has been referred to as keratinizing ameloblastoma3 or melanoticameloblastic odontoma.4 One of the histological features of the condition is the presence of ghost cells. Fejerskov and Krogh5 are of the opinion that the lesion initially presents as a solid tumor consisting mainly of ghost cells, and that the cyst development is a secondary phenomenon. They suggested a descriptive term of ghost cell odontogenic tumor for the lesion. A case is presented here in which cyst occurred in maxilla of 65-year-old female.

CASE REPORT

A patient of 65 years had a present swelling in right labial vestibule of 3 months duration. The problem began with mobility of upper right first premolar tooth around 1.5 months back. Then there was no swelling or any problems associated with the involved tooth. She went for dental consultation, and the tooth got removed. 1 week after the removal of tooth, she noticed a swelling on the right upper labial vestibule in the apical region of 15-16. Swelling was fluctuant and nontender. Then there were no associated dental problems. She consulted the dental surgeon again, and the doctor aspirated some fluid from the swelling. After aspiration swelling decreased and it again reappeared after 2 days when the dental surgeon referred the case to the oral surgeon with an orthopantogram (OPG). No history of diabetes, hypertension, or any other systemic diseases with relevant history of medications, allergy, or prior hospitalization was found. He had visited many dental surgeons for many dental treatments, and all were uneventful without any relevant history. No history of habits, mixed diet, and all bowel and sleeping habits are normal. No history of jaundice, clubbing, cyanosis, edema, or lymphadenopathy. The temperature was 98.4°F, pulse-82/min, respiratory rate-22/min, and B.P-122/84 mm of Hg.

LOCAL EXAMINATION

Soft tissue examination

Inspection

Diffuse swelling in the right upper labial vestibule extending from 12 to 15 regions. The mucosa over the swelling appeared normal. A bluish tinged area is visible in the lower part of the swelling near the labial part of the alveolar ridge.

Palpation

Soft, fluctuant, and non-tender swelling. Blanching of the overlying mucosa was observed felt.

Hard tissue examination

Inspection

Missing 12, 13, and 14 (H/O extraction) and poor periodontal status.

Palpation

Grade I mobile 11 and Grade II mobile 15 (Figure 1).
Investigations
X-ray: OPG reveals a well-circumscribed radiolucency with radiopaque borders of around 3.0 × 2.5 cm extending from mesial portion of the root of 21 to mesial of 16 root (Figure 2). Impacted 13 was also found in relation to the radiolucency. Small radiopaque were also found in the radiolucent area. The radiolucency was found involving the coronal portion of impacted 13.

Aspiration revealed around 2 ml of thin dark brown exudate from the swelling.

Provisional diagnosis
1. Dentigerous cyst
2. Unicystic ameloblastoma
3. Calcifying odontogenic epithelial tumor.

Treatment plan
1. Complete enucleation of the lesion along with removal of an impacted tooth
2. Intranasal antrostomy (if needed).

SURGICAL PROCEDURE

The procedure was planned to do under general anesthesia (GA). Under GA, patient was draped and painted. Local anesthesia was administered with 1:200000 lignocaine with adrenaline along the upper right labial vestibular region. Incison was placed along the alveolar ridge carefully not puncturing into the cystic contents and a releasing incision angulated upward from the horizontal incision between 22 and 23. A small releasing incision was also placed at the posterior limit of the horizontal incision along the labial mucosa apical to 16. Mucoperiosteal flap was reflected carefully separating the cystic lining from the labial mucosa with the help of mayo scissors. The cystic cavity was not found to involve the maxillary sinus. The whole cystic lining was curetted out from the bony wall carefully. The impacted canine was removed with forceps without the need of any bone removal as the bone around the tooth was eroded. The cyst was removed in toto along with the impacted tooth attached with the cystic lining. The bony cavity was thoroughly curetted and cleaned with normal saline. A soframycin pack was placed inside the cystic cavity, and the end of the pack was taken out through the labial mucosa by a stab incision over the labial mucosa. Flap was repositioned and sutured in place with 3-0 vicryl. The specimen was put in 10% formalin and sent for histopathological examination. Recovery was uneventful. Patient was kept as IP for 2 days and discharged after removal of the pack and being relieved of all the complaints. Patient was instructed to maintain proper oral hygiene and was prescribed antibiotics (Figures 3 and 4).

HISTOPATHOLOGICAL EXAMINATION

Given section shows stratified squamous nonkeratinized cystic epithelium with focal areas of intraluminal proliferation showing acantholysis and ghost cells. Associated fibrovascular connective tissue has moderate collagen fibers, odontogenic epithelial islands, and peripheral areas of bony spicules. Suggestive of calcifying epithelial odontogenic cyst (CEOC).

DISCUSSION

The CEOC is known to involve maxilla and mandible with equal frequency. The age of occurrence varies from 3 years to 80 years with definite peaking in the second decade. The cyst is usually asymptomatic unless secondarily infected. Some cases were reported where the cyst concomitantly occurred with other odontogenic lesions. Praetorius described four types of conjunctural lesions with the cyst namely,
dentin-producing ameloblastoma, odontoameloblastoma, ameloblastic fibro odontoma, and complex odontoma. The presence of ghost cells characterizes the histological appearance of the lesion. Ultrastructurally, the ghost cells contain coarse fibrils which may be due to incomplete keratinization of the cells. As the lesion matures, the ghost cells undergo dysplastic calcification. Sometimes, dentinoid- or osteoid-like material is elaborated near ghost cell connective tissue junction. This may be due to an Inductive effect of the epithelium. The calcifications if present will appear as scattered radiopaque flakes in radiograph. The other conditions which simulate this radiographic appearance are pindborg tumor, ameloblastic fibro odontoma, and adenomatoid odontogenic tumour. Maxillary sinus involvement of CEOC seems to be a rare occurrence. The patient in the case described by Gorlin was a 14-year-old female in whom the cyst encroached upon the right maxillary antrum and was associated with complex odontoma. Sudder-math described a cyst occurring in 22-year-old male without any other associated lesions. Enucleation of the cyst has provided total eradication of the lesion in all cases apart from the four published cases, in which the lesion recurred after surgical removal.

REFERENCES


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