CASE REPORT

Calcifying Odontogenic Cyst of Maxilla- A Rare Case Report

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ABSTRACT

The calcifying odontogenic cyst (COC) is an uncommon developmental lesion of the jaws, which was reclassified as a cyst in the 2017 WHO classification of head and neck tumours. COC mainly confined to the anterior maxilla, at the canine and premolar region. It is most frequently seen in females, with an increased incidence during the second and third decades of life. The diagnosis of COC is generally multi-factorial, involving clinical, radiographic and biopsy details.
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INTRODUCTION

Cystic lesions commonly affect the jaws, but dentigerous cyst and keratocysts are mostly prevalent when it comes to the maxillary bone. On the other hand, calcifying odontogenic cysts (COC) also known as Gorlin's cyst are rare lesions comprising 0.1% of all records and 1.3 % of all odontogenic cysts respectively (1). Since then, several aggressive diseases of COC and its benign counterparts have been reported with no clear etiology; there remains a belief that these lesions may be derived from remnants of the dental lamina or present in cell rests. COC can present in different clinical forms, such as unicystic, multicystic and extraosseous variants, that could explain why a clear pathogenic mechanism is still controversial. (2)

CASE REPORT

A medically fit 32-year-old female presented with history of swelling on the right side of her face for the last 6-7 months. She reports that the swelling was small in the beginning and gradually increased to its present size. The growth has stopped for the last 2 months. The patient also described mild pain associated with touching the swelling. There is no history of pain, pus discharge, or any other discomfort except facial asymmetry. Past medical, dental, and family histories were non-contributory. Extraoral examination revealed gross facial asymmetry on the right side of the face. The swelling was diffuse in nature and extended superiorly from the infraorbital margin to the line joining the lobule of the right ear to the corner of the mouth. Anteriorly it extended from the ala of the nose and posteriorly to the line corresponding to the lateral canthus of the eye. Swelling was tender upon applying pressure to the swelling (Figure 1a).

Local examination of the swelling intraorally showed a diffuse swelling in the right palatal region, extending anteriorly from the upper right front tooth region to posteriorly to the upper right back tooth region (tooth 16). It was hard in consistency and non-tender with expansion of palatal cortex. (Figure 1b) Based on history and clinical presentation a provisional diagnosis of Adenomatoid odontogenic tumor of right maxillary sinus was considered.

The patient was referred for radiographic investigations. An orthopantomogram showed an impacted 13 with haziness of the right maxillary sinus. A diffuse radiolucent lesion was seen in the right maxilla with a well-defined radiopaque border (Figure 2). Cone beam computed tomography revealed haziness of the right maxillary sinus and destruction of the buccal and lingual cortical

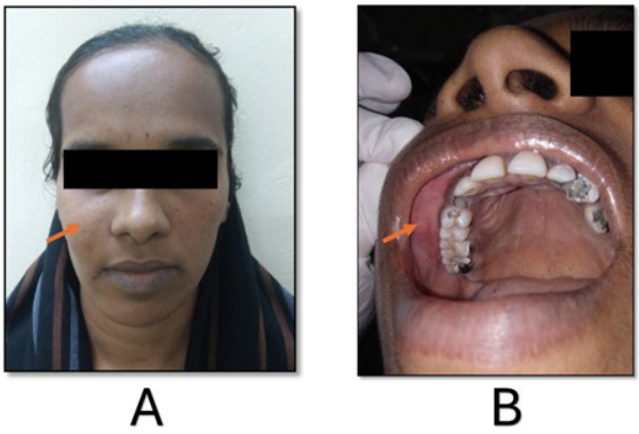

Figure 1: (A) Swelling on right side of the face. (B) Intraoral swelling on right maxillary region

Figure 2: OPG showing a diffuse radiolucent lesion on the right maxilla

plates on axial and coronal sections, respectively (Figure 3). Differential diagnosis of dentigerous cyst, calcifying odontogenic cyst, calcifying odontogenic tumor, unicystic ameloblastoma, and odontogenic keratocyst was considered.

Patient was referred to the department oral and maxillofacial surgery for incisional biopsy. The stained studied sections show a cystic cavity lined by epithelium, with basal columnar cells, superficial stellate reticulum-like cells and presence of ghost cells with some of them undergoing calcification. The fibrous connective tissue wall showed focal areas of dentinoid like material. Above features were suggestive of Calcifying odontogenic cyst (Figure 4a).

Considering the clinical, radiographic and histopathological features a final diagnosis of Calcifying odontogenic cyst of right maxillary sinus was given. Patient was posted for surgery under general anaesthesia and cyst enucleation was done. (Figure 4b). Patient was recalled after 15 days after surgery and evaluated for recovery. The patient was evaluated 6 months and 12 months after surgery to check for recurrence, and there was no evidence of recurrence.

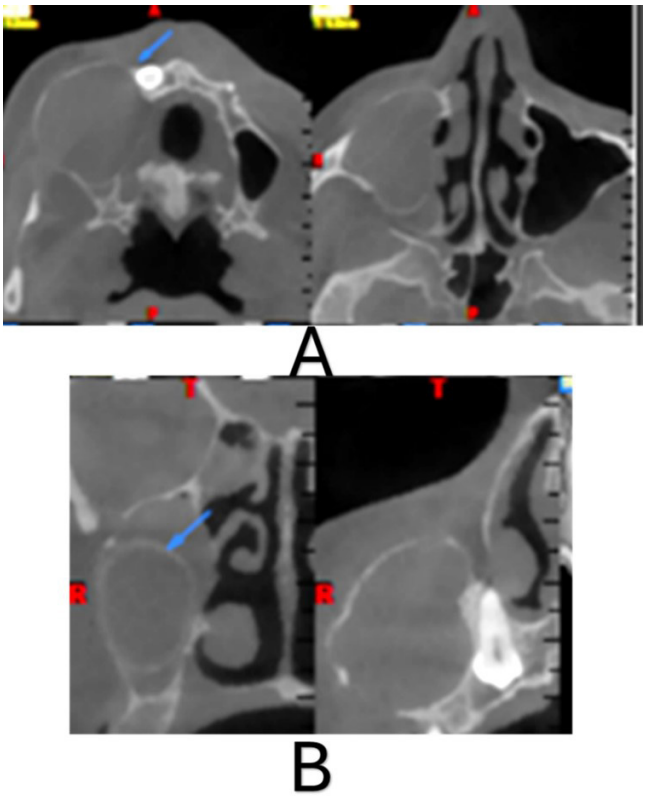

Figure 3(A and B): CBCT scans showing destruction on buccal and lingual cortex, with well-defined radiopaque lesion on the right maxillary sinus region.

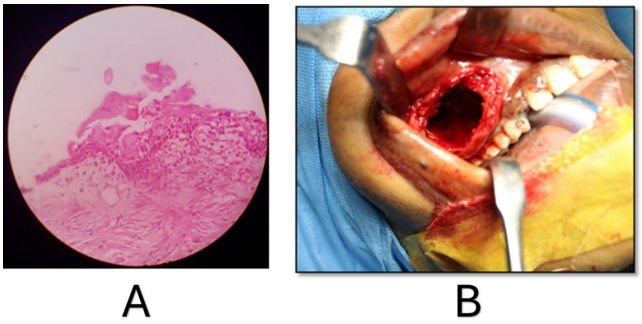

Figure 4: (A) A cystic cavity lined by epithelium, with basal columnar cells, superficial stellate reticulum- like cells and presence of ghost cells with some of them undergoing calcification. (B) Intraoperative picture showing cystic cavity

DISCUSSION

Calcifying odontogenic cyst, also named as Gorlin cyst, is one of the rare odontogenic lesions that has been presented with distinctive histopathological features. COC usually presents as a painless and slow-growing lesion in the jaw; the anterior portion of both maxilla and mandible is mostly affected. In the series by Arruda et al., COC accounted for 1.3% of odontogenic cysts, and 53% occurred in females and 47% in males. These lesions usually appear as unilocular radiolucencies in radiographs and may present associated calcifications in some instances (1,3).

In the study by Uchiyama et al., COCs were usually unilocular, with well-defined borders and regular margins. Root resorption was present in seven of nine cases, whereas tooth divergence in eight of nine cases. Impacted teeth were noted in six of nine cases, similar to those found in our case. Odontomas can also be very frequently associated with COC (4).

The most recommended COC treatment includes enucleation of the cyst followed by curettage; it includes enucleation followed by removal of a 1- to 2-mm layer of bone around the periphery of the cystic cavity with a sharp curette or bur. (5) This procedure was followed in the reported case as it aims to limit the risk of recurrence.

CONCLUSION

Calcifying odontogenic cyst, also named as Gorlin cyst, is one of the rare odontogenic lesions that has been presented with distinctive histopathological features. COC usually presents as a painless and slow-growing lesion in the jaw; the anterior portion of both maxilla and mandible is mostly affected. In the series by Arruda et al., COC accounted for 1.3% of odontogenic cysts, and 53% occurred in females and 47% in males. These lesions usually appear as unilocular radiolucencies in radiographs and may present associated calcifications in some instances (1,3).

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