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Case Report

Aggressive lesion of maxilla destined to be a calcified odontogenic cyst

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ABSTRACT

The lesion which was initially thought as an aggressive lesion turns to be the unique and unusual type of developmental cyst. A female patient being not aware about this turned up with the complaint of tooth ache and difficulty to chew in the right side. Later, after examination and investigations there seems to be calcified structure within the lesion. The lesion was diagnosed as a Calcified Odontogenic Cyst. The present study examines with the advanced imaging modality to study the lesion accurately. The surgical treatment and the follow up of the lesion have been discussed in the article.

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1. Introduction

The calcifying odontogenic cyst (COC) is an unusual lesion with the features of a cyst and the characteristics of a solid neoplasm.¹ The COC is one of the extreme uncommon cysts among odontogenic cyst, which shows wide range of radiographic appearances.² It usually occurs as an intra-osseous lesion but occasionally as an extra-osseous or peripheral variant. It shows an equal distribution between the maxilla and mandible and is commonly seen anterior to the first molar.³ The synonyms for COC are Calcifying Cystic Odontogenic Tumour (CCOT), Calcifying Epithelial Odontogenic Cyst (CEOC), Dentinogenic Ghost Cell Tumour (DGCT) and Gorlin Cyst.⁴

2. Case Report

2.1. Clinical findings

A 60yr old female patient complained of difficulty in chewing food, pain and swelling in the right upper back teeth region for 3 months. Extra orally facial asymmetry

was noted with diffuse swelling in the middle third of face which was hard and tender. Intraorally solitary well-defined swelling noted in maxilla irt 13, 14, 15, 16 region. The oval shaped swelling was about 4×3 cm in size noted. Mobility II mobility irt 13, 14 and grade III mobility irt 15, 16 was observed. The buccal vestibule was obliterated and buccal cortical plate was expanded. Provisionally it was diagnosed as Benign aggressive odontogenic tumour of right maxilla (Figure 1).

2.2. Radiographic findings

Radiographs of the lesion featured with well-defined unilocular radiolucency irt 13, 14, 15, 16 region. Internally heterogenous appearance noted with an area of calcification in the periapical aspect of 15. The borders were smooth, sclerotic rim was appreciated in the posterior region of lesion. Displacement of the roots with root resorption in multiplanar sites were noted with loss of lamina dura of the involved teeth and they were displaced. Buccal cortical plate was expanded. Absence of the floor of the maxillary sinus was appreciated. The right maxillary sinus and nasal cavity were obliterated (Figure 2). Soft tissue window in axial section of Computed Tomography (CT) imaging at

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Fig. 1: Clinical presentation

the level of maxilla showed, well defined unilocular lesion with sclerotic border, the lesion was having the density of a soft tissue and internally calcified material with irregular appearance. The coronal section showed the obliteration of the maxillary sinus and nasal cavity (Figure 3).

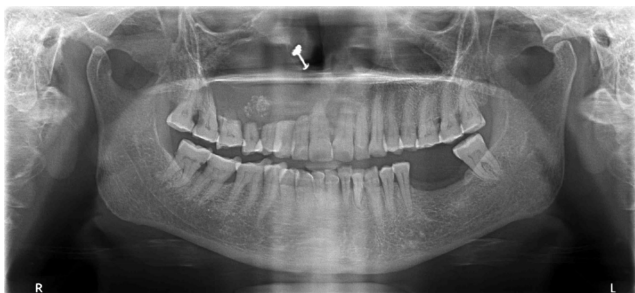


Fig. 2: Pre-operative OPG

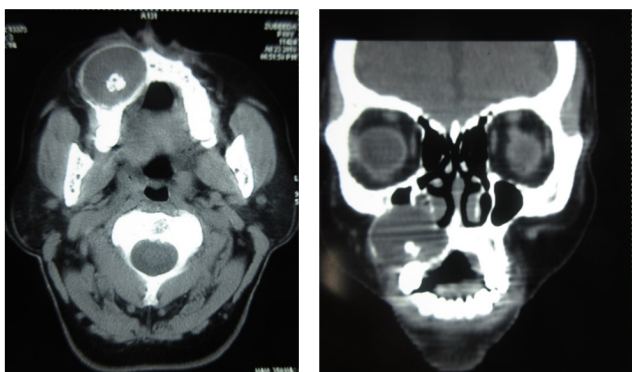


Fig. 3: Coronal Section - Soft tissue window of CT

2.3. Surgical intervention

The lesion was surgically excised along with the extraction of the involved teeth under general anaesthesia (Figure 4 a).

2.4. Histopathological examination

H and E-stained section shows fibrous cyst wall showing extensive ulceration and wall lined by multi-layered cuboidal epithelium, ghost cell formation with elongated rete ridges were noted and it was diagnosed as Calcifying odontogenic cyst of right maxilla (Figure 4 b).

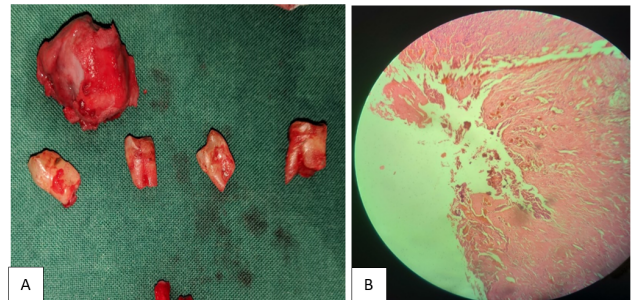


Fig. 4: Excision of lesion (a) & Histopathological section (b)

2.5. Follow up

Patient was examined after 6 months and was subjected to the imaging of Panoramic radiograph. It reveals maxillomandibular complex with density of the treated area was similar to the adjacent area and no evidence of any recurrence of the lesion noted elsewhere in the jaws (Figure 5).

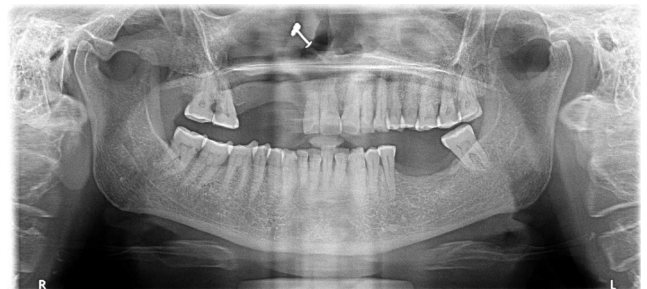


Fig. 5: Post-operative OPG

3. Discussion

The COC has features of both cyst and solid neoplasm making it a little controversial and comprising of two distinct clinicopathologic entities as a cyst and a neoplasm.⁵ The 2005 classification clarified the confusion and determined that both were neoplastic, naming them as calcifying cystic odontogenic tumours for cystic forms

and dentinogenic ghost cell tumours for solid forms, but the 2017 classification changed COC to the developmental cyst variant and the solid form was kept in the mixed odontogenic neoplasm. COCs are simple cysts lined by epithelium with focal accumulations of ghost cells.⁶ Three different types can be found in the cystic variant: simple unicystic, unicystic odontoma-producing and unicystic with ameloblastomatous proliferation.^{7,8} A COC is in the oral cavity lined by odontogenic epithelium of variable thickness containing focal ghost cells.⁹ Most common site is anterior maxilla, followed by posterior mandible. In extraosseous and intraosseous variants of COC asymptomatic swelling is a common clinical feature with the expansion of the cortical plates. The lesion produces calcified structure which is identified as dysplastic dentin.³ Radiographically, COCs are well circumscribed and shows unilocular radiolucency with calcifications of varying density, root resorption and divergence are frequently seen with COC.⁹ The treatment of COC is enucleation and the response depends on any associated odontogenic tumour. It has good and favourable prognosis.¹⁰

4. Conclusion

The ambiguity regarding the interpretation of the lesion still prevails and has become a challenge to diagnose. COC should be considered in the differential diagnosis of the swelling of intraosseous origin in the maxillofacial region. A complete clinical, radiological and histological assessment becomes mandatory to give a correct diagnosis and effective treatment for the lesion. The follow up plays an important role to evaluate further recurrence of the lesions.

5. Source of Funding

None.

6. Conflict of Interest

None.

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