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Calcifying Odontogenic Cyst -An Uncommon Occurrence

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Abstract

Calcifying odontogenic cyst (COC) or Gorlin cyst has been defined as a simple cyst lined with ameloblastoma-like epithelium containing focal ghost cells. In 1971, the COC gained international recognition when the World Health Organization (WHO) included it in its classification of "Histological Typing of Odontogenic Tumors, Jaw Cysts, and Allied Lesions.

COC is an uncommon lesion, representing 0.1% of all records and 1.3% of all odontogenic cysts with a predominance at the anterior maxillae in patients of the third decade.

Here we report the case of a 32 years old female patient, who was diagnosed with COC of anterior maxilla. Total enucleation of the lesion was done under general anesthesia.

Satisfactory healing was noticed in the following visits. As it has varied radiological and clinical presentation, it is difficult to arrive to this diagnosis. It has a rare recurrence. It is important to include COC in differential diagnosis of odontogenic lesions involving bone resorption.

Keywords: COC, WHO, Jaw Cysts

Introduction

Calcifying odontogenic cyst (COC) or Gorlin cyst has been defined as a simple cyst lined with ameloblastoma-like epithelium containing focal ghost cells. In 1971, the COC gained international recognition when the World Health Organization (WHO) included it in its classification of "Histological Typing of Odontogenic Tumors, Jaw Cysts, and Allied Lesions"[1].

COC is a hybrid lesion of the jaw exhibiting a diverse variety of clinical behaviours and histopathological features including cystic, solid (neoplastic) and aggressive variations. It consists of a proliferation of ameloblastoma-like odontogenic epithelium and scattered nests of ghost cells that may calcify. Medical and dental literature often states that the COC was first recognized as a distinct clinicopathological entity by Gorlin et al., in 1962[2].

COC is an uncommon lesion, representing 0.1% of all records and 1.3% of all odontogenic cysts with a predominance at the anterior maxillae in patients of the third decade[3].

Case Report

A 32 years old female patient came to our unit with the complaint of pain in the maxillary right anterior teeth region since one week. Patient gave history of swelling in the same region, which started one year back and gradually increased to the current size. There was no history of trauma, paresthesia and lymphadenopathy. There was no complaint of nasal stiffness and epistaxis. Patient had no relevant medical history.

On clinical examination,a swelling of size 4cmx4cm was noted in the right middle one third of face. The skin over the swelling appeared normal. Obliteration of the right nasolabial fold was noted (fig. 1)

On palpation, the swelling was soft in consistency and moderately tender.

On intra oral examination, the swelling was noted extending in the vestibule from 11to 16 regions. The mucosa over the swelling appeared mildly erythematous. Grade II Mobility was noted with respect to 11, 12, 13.

Routine blood investigations revealed normal values. The Panoramic radiograph (fig.2) revealed a well-defined radioluscency extending from periapical region of 11, 12, 13, 14, 15 to close proximity to the right maxillary sinus floor.

Resorbed roots of 11, 12, 13 were noted. A calcified mass was noted in the medial part of the lesion.

CT face revealed a well marginated, unilocular cystic lesion extending from right paramedian region of maxilla to right ipsilatral alveolar ridges. An irregular intraluminal radiodense opacity measuring 11x10x9.2 mm with adjacent small calcifications in the anteromedial aspect of the lesion was noted.(fig.3)

Aspiration of the lesion was performed under local anesthesia, which revealed a clear straw coloured fluid (fig.4)

A total enucleation of the lesion, surgical curettage and extraction of 11, 12 was performed under general anesthesia and platelet rich fibrin was placed and closure was achieved. The sutures were removed on the tenth postoperative day and satisfactory healing was observed.

The enucleated lesion was sent for histopathological examination, (fig .8) which revealed cystic wall bordered by an odontogenic epithelium whose basal layer was made of cubocylindrical cells. Ghost cells and keratinization were noted in the superficial layer. Thus, it was arrived to the diagnosis of calcifying odontogenic cyst.

Discussion

First described in 1962 by Gorlin, COC is a rare lesion of jaw bone. It is defined as a cyst with an ameloblastoma-like epithelium containing calcifications

and focal ghost cells. These cells are also present in dentinogenic ghost cell tumors [3].

In the latest edition of the WHO classification of tumors of the head and neck (2017), Calcifying odontogenic cyst has been classified among the developmental cysts with two entities: intraosseous and extraosseous [4].

Primary clinical presentations vary from loosening of teeth to increasing swelling[3,5].

Incidence is almost equally distributed between the maxilla and the mandible. Reports on gender prevalence vary in the literature but, the occurrence of COCs is approximately equal in males and females. There appears to be a slight predilection for Caucasians[3]. The incidence of root resorption is 75% to 77% of cases[6].

Swelling in the involved region is the most common clinical sign noted in patients with COC. Smaller lesions are usually painless and discovered incidentally during routine radiographic evaluation. When located in the maxilla, patients may sometimes complain of nasal stiffness, epistaxis and headache [7].

Radiographically, COC generally appears as a unilocular lesion with a well-defined margin, and contains calcifications as in our case. The frequency of the multilocular form has been noted as 5% and the presence of calcification, which is an important radiographic feature in the interpretation of COC, is detected in about half of all COCs [8]. The unique feature of COCs is ghost cell keratinisation, similar to its cutaneous variant, a Pilomatrixoma[9]. It is this unique feature that also differentiates it from an ameloblastoma.

However, the synchronous occurrence of COC with ameloblastomas and other odontogenic pathologies including dentigerous cysts [10], ameloblastic fibromas, and odontomas[11] has been reported. COC's are complex lesions and can have substantial variation in the morphology of the epithelium, with several different lesions incorporated, including other odontogenic cysts and/or tumours[12]. Various literatures have sub-classified COCs, however, their behaviour remains the same despite a difference in classification.

Enucleation is the treatment of choice for intraosseous COCs. The extraosseous COC is treated with surgical excision. The reoccurrence rate for both intraosseous and extraosseous COCs is low and the long term prognosis is good[13]. Recurrences have been reported 5 years or more after surgery [14].

Treatment for the central COCs was confined to enucleation, and for the peripheral COCs to local excision. This conservative treatment is consistent with the recommendations of others, despite reports of a few recurrences[15].

Marsupialization could be indicated as a preliminary treatment to COC[16]. Central COC cases not associated with odontomas displayed higher proliferative activity than odontoma associated and peripheral cases[17].

Differential Diagnosis

The differential diagnosis of COC is made with all mixed lesions of the maxillae. It includes benign radiolucent lesions such as dentigerous cyst, adenomatoid odontogenic tumor, ameloblastic fibro-odontoma, and calcifying epithelial odontogenic tumor . Classical histologic findings in COC are an odontogenic epithelium displaying keratinized ghost cells and calcification. Induction of dental hard tissues may be found in solid COC variants[12]. All enucleated specimen need to be examined histopathologically to confirm the working diagnosis, so as appropriate precautious treatment plan and follow-up can be instituted[18].

Conclusion

The COC is a rare odontogenic lesion. As it has varied radiological and clinical presentation, it is difficult to arrive to this diagnosis. It has a rare recurrence. It is

important to include COC in differential diagnosis of odontogenic lesions involving bone resorption.

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Legend Figures

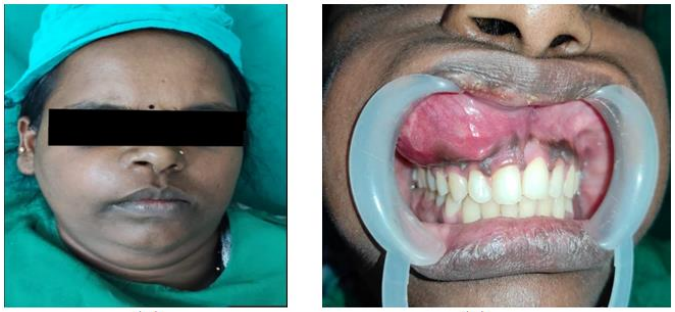


Fig.1: Preoperative Photographs (A) Extraoral, (B) Intraoral

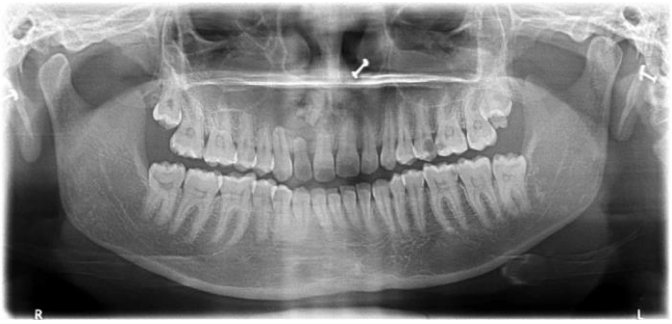


Fig .2: Pre-Operative Panoramic Radiograph

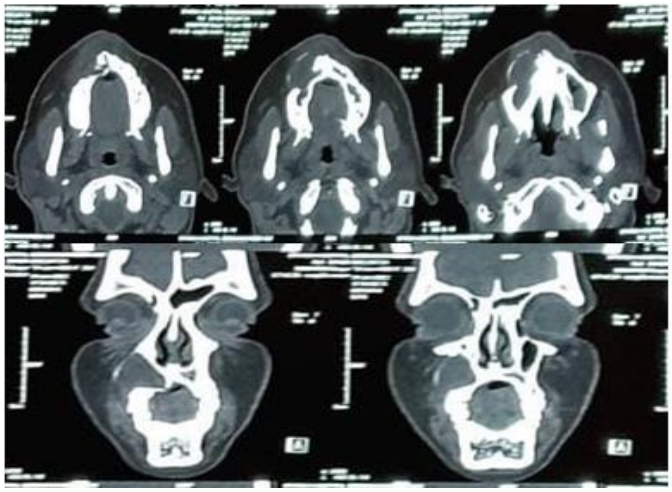


Fig.3: Preoperative Ct: Axial And Coronal View

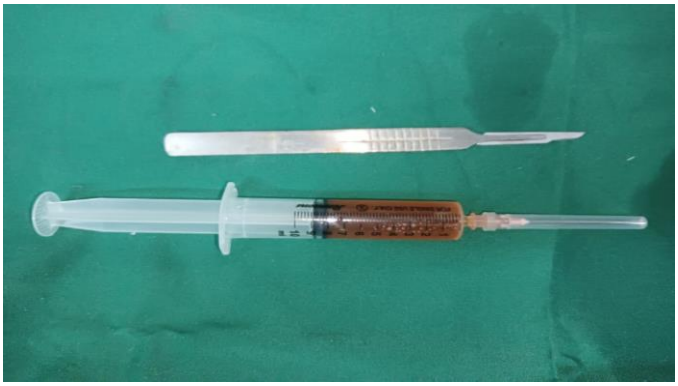


Fig.4: Aspirated Contents of the Cyst

Fig. 5: Enucleation of the Cyst Along With Removal of the Odontome

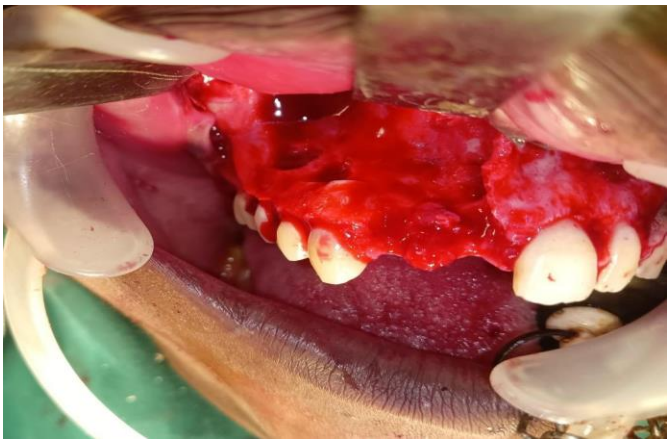
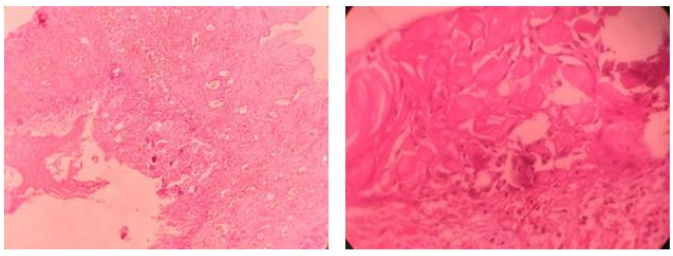


Fig. 6: After Enucleation and Curettage

Fig .7: Closure Achieved Using 3-0 Vicryl Suture Material



(A) (B)

Fig.8: (A) Histopathological Photograph of the Excised Specimen. (B) Photo Showing Ghost Cells and Keratinisation