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AMELOBLASTOMATOUS CALCIFYING CYSTIC ODONTOGENIC TUMOR: A CASE REPORT OF A RARE VARIANT

Dental Science

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ABSTRACT

Calcifying odontogenic cyst is considered as a rare lesion and accounts for 1% of jaw cysts. It represents a heterogeneous group of lesions which exhibit a variety of clinicopathologic and behavioral features. It has been categorized as cyst and neoplasm. Calcifying odontogenic cyst is an uncommon developmental odontogenic cyst first described by Gorlin in 1962. It is considered as an extremely rare cyst and accounts for only 1% of jaw cysts reported. Because of its diverse histopathology, here has always been confusion about its nature as a cyst, neoplasm, or hamartoma. WHO classification 2005 has reclassified the lesion as calcifying cystic odontogenic tumor (CCOT) in this report, we present a rare case of calcifying odontogenic cyst -an extremely rare histologic variant in a 29 year-old female in the maxillary anterior region. The lesion was surgically removed. After enucleation no recurrence has been recorded in the ensuing 2 years. Radiological features: CCOT are generally a unilocular lesion, in 5–13% multilocular. Three patterns of radiopacity seen: 1st salt and pepper pattern of flecks, 2nd fluffy cloud like pattern, and 3rd crescent-shaped pattern on one side of the radiolucency in a "new moon"-like configuration. Histopathological features: Stroma showed thick bundles of collagen fibers with spindle fibroblasts, Juxta-epithelial hyalinization, Daughter cysts with ghost cells, Dysplastic dentin in and around some odotogenic islands. With the above histopathologic features, a diagnosis of ameloblastomatous CCOT was made. Results: The patient has been under regular follow up and has not exhibited any signs of recurrence after 2 year of follow up.

KEYWORDS

Ameloblastic proliferation, calcifying odontogenic cyst, Ghost cell, Gorlin cyst, Calcifying cystic odontogenic tumor

INTRODUCTION

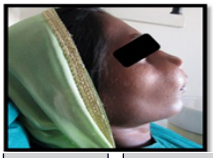
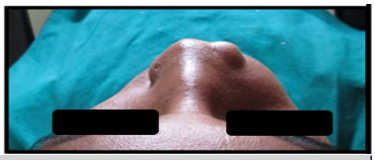
Calcifying odontogenic cyst (COC) was first described as a distinct clinicopathologic entity by Gorlin et al., in 1962. [1][6] Ever since then controversy and confusion have existed regarding its nature. According to the WHO classification in 2005, COC has now been reclassified as calcifying cystic odontogenic tumor (CCOT). [2][5] The lesion shows extreme diversity in its clinical and histopathological features as well as in its biological behavior. CCOTs are frequently associated with odontogenic tumors, a finding which is a rare event in other types of odontogenic cysts or tumors. [3] Central and peripheral forms of calcifying odontogenic cyst occur equally in the upper and lower jaws. [7] Calcifying cystic odontogenic tumor can occur in very young patients, even in the first year of life.

CCOT may clinically be diagnosed as Ameloblastoma, Calcifying epithelial odontogenic tumor, Adenomatoid odontogenic tumor, Ameloblastic fibroodontoma, complex or compound odontoma and dentigerous cyst or as other types of odontogenic cysts. [2]

In this case report, a case of ameloblastoma proliferating type CCOT that occurred in a young female in maxillary anterior area.

CASE REPORT

A 29-year-old female patient visited the department of oral and maxillofacial surgery Narsinhbhai Dental College and Hospital, with a chief complaint of pain and swelling over middle third of face since 3 months. Swelling is well defined on right lateral side of nose which started gradually increased to attain the present size, extending from infraorbital margin to upper lip. Obliteration of nasolabial fold is seen. Palpation swelling is hard & overlying skin normal. Teeth missing upper lateral icisor and upper right canine. No history of pus, blood or watery discharge, color change or parasthesia noted over the swelling, difficulty in opening the mouth, speaking and swallowing food, other associated symptoms like fever, loss of appetite, loss of weight, diarrhea or fatigue all were absent.

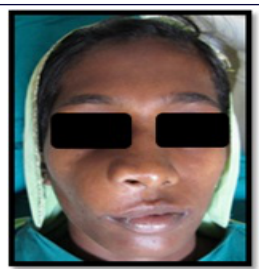
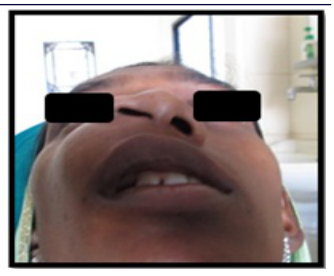



Figure 1: extra oral swelling

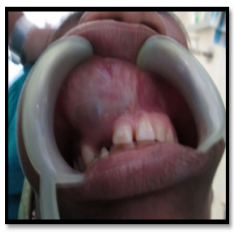


Figure 2: intra oral clinical photograph swelling

The orthopentomogram revealed a multiloculated,large cystic and expensile lesion involving right upper anterior jaw region. The solid tumor measured about 3.5 × 3.0 cm in size. Impacted upper right lateral incisor tooth & upper right canine tooth was also revealed. Based on the clinical feature and radiographical appearance provisional diagnosis of dentigerious cyst was made.

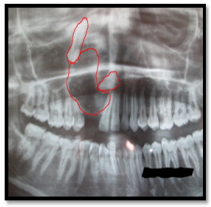


Figure 3: Orthopentogram ; with marking of lining of tumour and impacted teeth

Figure 4: surgical removal of tumour under local anesthesia

The lesion was surgically enucleated along with associated impacted tooth upper right lateral incisor & upper right canine. Excised specimen was sent for further histopathological examination. Excised lesion was cystic, measured about 3.5 × 3.0 cm in size. Excised tissue reveled cystic lining of stratified epithelium with cuboidal to columnar basal cells few showed reverse polarity. Suprabasal cells were stellate reticulum like and few areas showed squamous metaplasia. Ghost cells were seen in few areas. Juxta-epithelial dentinoid like material was seen. Stroma showed thick collagen with hyalinization and numerous odontogenic follicles. A final diagnosis of ameloblastoma proliferating type CCOT was given. The patient has been under regular follow up and has not exhibited any signs of recurrence after 2 years of follow up.

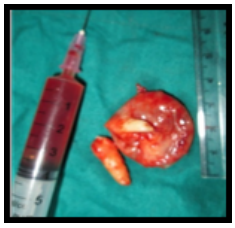


Figure 5: excised lesion with aspirated fluid

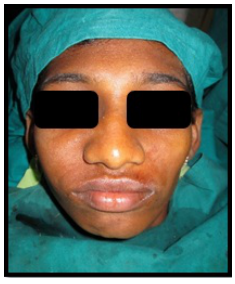


Figure 6: Immediate post-operative photograph showing marked reduction in swelling

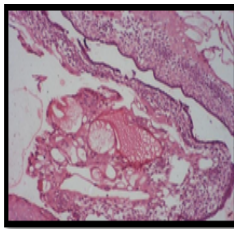


Figure 6: Histopathological section 20x magnification

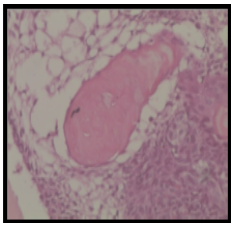


Figure 7: Histopathological section 40x magnification

DISCUSSION

The Calcifying odontogenic cyst accounts for about 1% of all the jaw cysts. Associated with other odontogenic tumors Like Odontomas (20%), Ameloblastoma, and Adenomatoid Odontogenic Tumor. Occur alone or in association with other odontogenic tumors such as, adenomatoid odontogenic tumors and ameloblastomas. [8]

COC was first described in 1932 by Rywkind who reported a lesion of the jaw which resembled cholesteatoma of the ear and thereafter called it as cholesteatoma of the jaw. [4] In 1946, Thoma and Goldman described a lesion which they called a strange variant of ameloblastoma [11]. Gold in 1963 named the lesion as "Keratinizing calcifying odontogenic cyst." Fejerskov and Krogh in 1972 called it "Calcifying ghost cell odontogenic tumor," whereas Freedman *et al*. in 1975 suggested the name "Calcifying cystic odontogenic tumor (CCOT)." [14] Ledesma-Montes *et al*. reviewed the clinical-pathological features of 122 CCOT, DGCT and GCOC cases from 14 institutions in seven countries of three different continents and concluded that of all the cases, Ameloblastomatous proliferating type of CCOT accounted for only three cases (2.46%) indicating the lesion to be very rare. [15]

Central and peripheral forms of calcifying odontogenic cyst occur equally in the upper and lower jaws. Johnson et al reported the occurrence of 60% of the tumors in mandible, 30% in the form of peripheral calcifying odontogenic cysts and anterior part of the jaw was involved in 53% of cases. Hirschberg et al concluded that the upper jaw was affected in 61.5% and the anterior region of the jaw in 75% of the reported cases. [12]

As per Hong et al. two categories for CCOT associated with ameloblastoma: First is the Ameloblastomatous Cystic variant & second is the Neoplastic Variant Associated with Ameloblastoma. [9][10] According to few studies CCOT is more common in females and in maxilla where as there are reports of CCOT occurring more in males and in mandible. Cases have also been reported where calcifying cystic odontogenic tumor is provisionally diagnosed as a residual cyst as well as a Periapical pathology. About 50% of CCOT have been reported as being associated with a unerrupted tooth. Displacement of teeth is often seen. Resorption of roots of the adjacent teeth is a frequent finding and is regarded as an important radiological feature. Local expansion sometimes occurs, and perforation of the cortical plate, when present, may be radiologically demonstrable. [3]

The treatment of cystic lesion involves enucleation with long-term follow-ups. Recurrence depends on completeness of cyst removal. Prognosis is good for cystic CCOT and less certain for neoplastic CCOT. The CCOT may be associated with other odontogenic tumours for that the treatment and prognosis is based on the associated tumors. Buchner suggested that if the COC was associated with an ameloblastoma, its behavior and prognosis would be of the same as an ameloblastoma, not COC. [13]

CONCLUSION

At this present state, it is very difficult to determine whether any individual lesion having a cystic architecture is truly cystic or, in fact, neoplastic in nature. An extensive and systematic analysis of many more cases including immunohistochemical investigations on cell proliferation activity may help resolve this problem. Ameloblastomatous COC is a rare histologic variant. We found very few cases of ameloblastomatous COC in the literature. Our case did not show any evidence of recurrence after treatment, but there is no doubt that careful postoperative observations are necessary for CCOTs which are associated with an ameloblastoma.

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