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Gorlin Cyst or a Benign Odontogenic Tumor: A Diagnostic and Treatment Dialemma

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Case Report

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Abstract: The Calcifying Odontogenic Cyst (COC) represents a heterogeneous group of lesions that exhibits a variety of clinicopathologic and behavioural features. Therefore a proper categorization of the cases is needed for better understanding of each variant. Ameloblastoma is one of the well-known odontogenic tumours that could be associated with Calcifying Odontogenic Cyst. Very few cases of Ameloblastomatous calcifying odontogenic cyst have been reported in the literature. In this report we present a case of ameloblastomatous transformation of calcifying odndontogenic cyst.
Keywords: Gorlin cyst, Ameloblastoma, Calcifying odontogenic cyst.

INTRODUCTION

Gorlin cyst which is also known as calcifying odontogenic cyst (COC), calcifying cystic odontogenic tumor (CCOT), calcifying ghost cell odontogenic cyst, and dentogenic ghost cell tumor, is a rare developmental lesion that arises from the odontogenic epithelium [1] and represents about 2% of all odontogenic pathological changes in the jaw[2-4]. It is clinically characterized as a painless, slow‑growing tumor which equally affects the maxilla and mandible, has a predilection for the anterior region of the jaw bones, and usually arises intraosseously although it may occur extraosseously too. It has a peak incidence during the second and third decades of life with a mean age of 30.3 years and does not demonstrate any gender predilection [5]. Radiographically, Gorlin cyst may appear as a unilocular or multilocular radiolucent lesion with either well‑circumscribed or poorly defined margins and may also be observed in association with unerupted teeth [6] Calcification is an important radiographic feature for the interpretation of Gorlin cyst but is detected in only approximately half of the reported cases[7].

The typical histopathological features of CCOT include a fibrous wall and lining of odontogenic epithelium with either columnar or cuboidal basal cells resembling ameloblasts. Stellate reticulum‑like cells overlay the basal cell layer while ghost cells, which may occasionally become calcified, are also seen in the lining of the cyst [7]. Even after several classifications and sub-classifications, COC remains an enigma. Very few cases of ameloblastomatous COC have been reported in the literature. The treatment of choice for Gorlin cyst is conservative surgical enucleation. However recurrence is frequent especially if associated with neoplastic variant.

The cystic type of COC comprises the majority of the cases, which are characterized by a unicystic lesion associated with or without an odontoma. They may also show ameloblastomatous proliferative activity intraluminally or intramurally (ameloblastomatous COC). The neoplastic variants of COC, which show a solid growth pattern consisting of ameloblastoma-like strands and islands of odontogenic epithelium infiltrating into mature fibrous connective tissue, are further sub classified into ameloblastoma arising from COC (ameloblastoma ex COC) and odontogenic ghost cell tumors [8]. Malignant transformation of COC has also been reported [9].

CASE REPORT

Fig-1: Introral view of the swelling present irt 45,46,47 with bucco-lingual bony expansion

A 40year old male patient named Mr. Murugan reported to our department with the chief complaint of pain and swelling in relation to right lower jaw for the past 2years. On extraoral examination a well-defined swelling measuring around 3*2 cm in size is present in right lower jaw region which is firm on palpation.

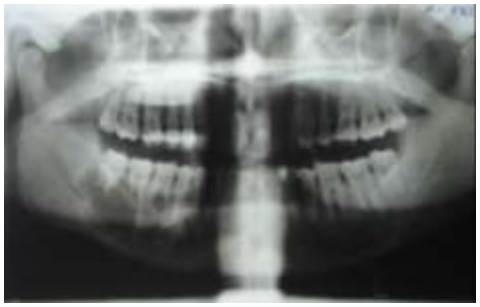

Fig-2: OPG reveals well defined radiolucency in rt. Body of mandible

On intraoral examination a well localized swelling present measuring around 4*2cm in size with obliteration of buccal vestibule from 45 to 48 region. Swelling has well defined borders, firm in consistency and non-tender.

Incisional biopsy was done and the histopathological report was found to be gorlin cyst. Surgical plan was enucleation of cyst under general anesthesia.

Cyst was enucleated. Post op biopsy (histopathological report) turned out to be ameloblastoma associated with gorlin cyst. Patient was kept under constant review. Patient reported to our department after 2 years with a complaint of recurrent swelling in relation to right lower jaw region. On examination there was a recurrent swelling in the right body region of the mandible which is firm in consistency in the same region. We planned for segmental resection of right lower jaw followed by reconstruction.

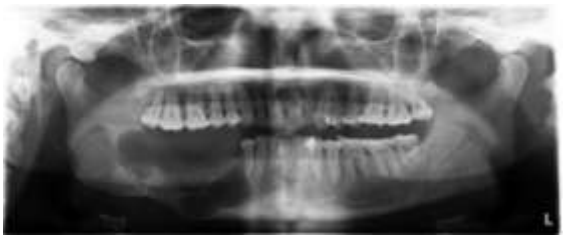

Fig-3: OPG taken 2 yrs later showing recurrent lesion in rt. Body of mandible

Fig-4: Segmental Resection of rt. Mandible for Reccurent lesion

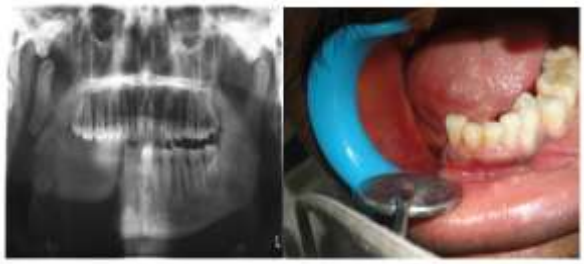

Fig-5: Post Op OPG and clinical picture

DISCUSSION

A cyst is defined as a pathological cavity which may or may not have an epithelial lining and which has a fluid, semi‑fluid, or gaseous content and is not formed by the accumulation of pus. Calcifying odontogenic cyst, as Gorlin cyst was recognized earlier, was first reported by Gorlin *et al*. in 1962 [10]. At that time, it was classified as a cyst related to an odontogenic apparatus.

In 1971, the World Health Organisation described COC as "non neoplastic cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cell layers thick that may resemble stellate reticulum and masses of ghost cells that maybe in the epithelial cyst lining or in the fibrous capsule [11].

Calcifying odontogenic cyst occurs intraosseously or extraosseously, with intraosseous being more predominant. Prior to separation of this entity by Gorlin *et al*. it was often regarded as some form of ameloblastoma. The COC is an uncommon lesion demonstrating considerable histologic diversity and presenting with variable clinical behaviors. Although, it is broadly considered to represent a cyst, some investigators prefer to classify it as a neoplasm [12]. The question concerning the nature of the cyst appeared to be clarified by Toida, who recently categorized COC into a cyst and neoplasm [13].

In the new classification of World Health Organisation, the term calcifying cystic odontogenic tumor was replaced by calcifying odontogenic cyst (COC) which constitutes a benign cystic neoplasia presenting an epithelium with ghost Cells which may display calcification in it [14].

It was later renamed as calcifying cystic odontogenic tumor (CCOT) in the World Health Organization classification revised in 2005 due to its histological complexity, morphological diversity, and aggressive proliferation. CCOT was later recognized by numerous names including Gorlin cyst, calcifying ghost cell odontogenic cyst, and dentogenic ghost cell tumor.

According to Praetorius *et al.* the cystic lesion can be divided into three basic type's simple unicystic type, unicystic odontoma producing type, and unicystic ameloblastomous producing type [15]. Microscopically ameloblastomatous COC resembles unicystic ameloblastoma except for the ghost cells and calcifications within the proliferative epithelium. Ameloblastomatous COC occurs only intraosseously [16] Ameloblastoma ex COC designates an ameloblastoma which arises from the cyst lining of COC [17]. It can also occur intraosseously, appearing as cyst-like radiolucent lesion. Whether these tumors have the same disruptive potential and tendency for occurrence as a typical ameloblastoma is unknown [18].

From the year of description of CGCOC in 1961 till date different terminologies and classifications have been proposed and practiced in the literature-

Terminology	Author proposed
Calcifying odontogenic cyst (COC)	Gorlin *et al.* (1962)
Keratinizing Calcifying odontogenic cyst (KCOC)	Gold *et al.* (1963)
Keratinizing ameloblastoma (KA)	Bhaskar (1965)
Calcifying ghost cell odontogenic tumor (CGOT)	Fejerskov and Krogh (1972)
Cystic calcifying odontogenic tumor (CCOT)	Freedman *et al.* (1975)
Dentinogenic ghost cell tumor (DGCT)	Praetorius *et al.* (1981)
Epithelial odontogenic ghost cell tumor (EOGCT)	Ellis and Shmooker (1986)
Calcifying ghost cell odontogenic cyst (CGCOC)	Toida (1998)
Odontogenic ghost cell tumor (OGCT)	Colmenero *et al.* (1990)
Odontogenic ghost cell ameloblastoma (OGCA)	Shear (1994)
Odontocalcifying Odontogenic tumor (OOT)	Wirshberg *et al.* (1994)
Calcifying cystic odontogenic tumor (CCOT)	WHO classification (2005)

CONCLUSION

Gorlin cyst and its literature review –it is called by different terms since 1961 and its presentation with various histopathological features. Treatment plan should be based on the presentation and its association with any tumours or its aggressive neoplastic variant. Periodic review of the patient is essential if enucleation of cyst was the treatment plan.

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