

Dentinogenic Ghost Cell Tumor: A Rare Case Report in Maxillary Sinus

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Abstract

Ghost Cell Tumors (GCT) are a class of lesions that can exist in the form of a cyst or solid neoplastic tumor, and in terms of behavior, they can be benign, locally invasive, or metastatic. They are identified by the presence of ame-loblastic epithelium, ghost cell, and calcification. These lesions are mostly prevalent in the lower jaw, anterior mandible area. They are distributed equally between the male and female population in the second, sixth, and eighth decades of life. Since the recurrence rate is very low, it seems that a conservative approach is sufficient for most cases. Here, we present a rare case of dentinogenic ghost cell tumor in the left sinus of a 32-year-old male patient. It is temporarily diagnosed through clinical findings and radiographic scan, and later on, confirmed via histology.

keywords: COC, DCOT, Maxillary sinus, Neoplasia

Introduction

GCOTs are a group of transient tumors characterized by the presence of ghost cells, which include calcifying cystic odontogenic tumor (CCOT), dentinogenic ghost cell tumor (DGCT), and ghost cell odontogenic carcinoma (GCOC), all made of calcifying odontogenic cysts (COCs) [1]. (DGCT) dentinogenic ghost cell tumors are extremely rare tumors that are considered solid variants of calcifying epithelial odontogenic cysts (CEOC) and their main characteristics are aberrant keratinization in the form of ghost cells and dysplastic dentin [2]. This lesion only makes up 2-14% of cases of all COCs [3].

There are two types of DGCTs, extraosseous (peripheral) and intraosseous (central). The intraosseous DGCT is more invasive, with invading growth pattern and a high recurrence rate after removal. Thus, in comparison with the extraosseous type, the intraosseous DGCT is treated by extensive surgical resection together with a sufficient safety margin [4].

These lesions are mostly prevalent in the lower jaw, anterior mandible area. They are distributed equally between the male and female population in the second, sixth, and eighth decades of life. Almost 20% of all lesions show signs of bone resorption in the anterior region, which occurs more in DGCT [5]. Since the recurrence rate is very low, it seems that a conservative approach is sufficient for most cases [6].

Case Report

A 32-year-old man suffering from nasal congestion and swelling of the sinus area was referred to our department by his dentist. His CT scan showed a filled-up left sinus area with a defined thin cortical boundary (Figure 1). In the Waters' view of the radiographic scan, a space-occupying lesion was observed in the left sinus area, which has destroyed the lateral nasal wall and has caused erosion (Figure 2).

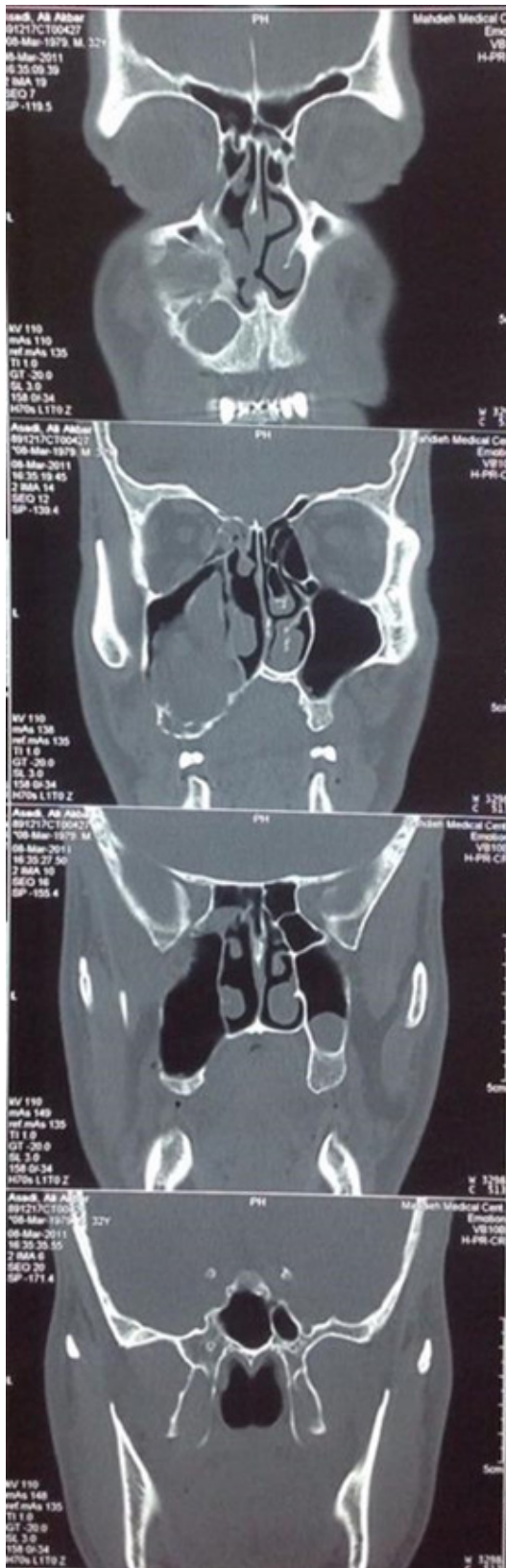


Figure 1: CT scan showing maxillary sinus



Figure 2: waters radiograph showing maxillary sinus filled-up

In order to diagnose if the lesion was originated from the base of the sinus or the alveolus, the sample was sent for histopathological testing. The sample taken from the patient showed a cystic lesion, and in some area, the evidence of ghost cells was detected. These accumulations of ghost cells in the connective tissue are observed along with the areas of satellite cystic and tumor islands; thus, according to the mentioned issues, this lesion diagnosed as dentinogenic ghost cell tumor (Figure 3 and 4).

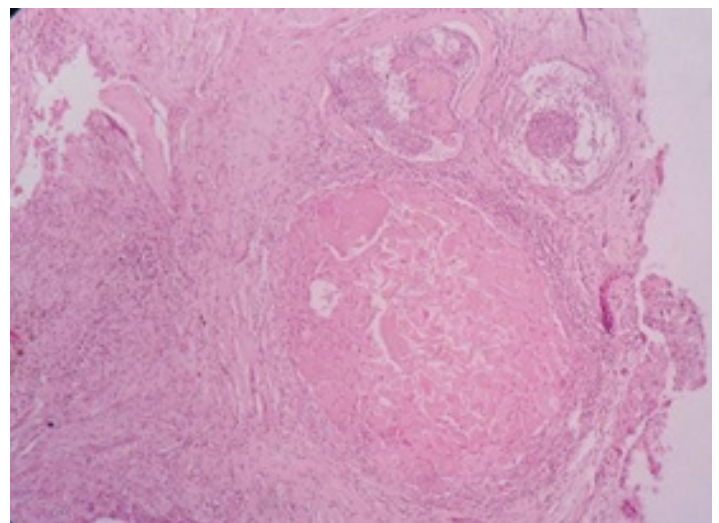


Figure 3: Low-power microscopic Microscopic view shows tumor proliferation in dentinogenic ghost cell tumor

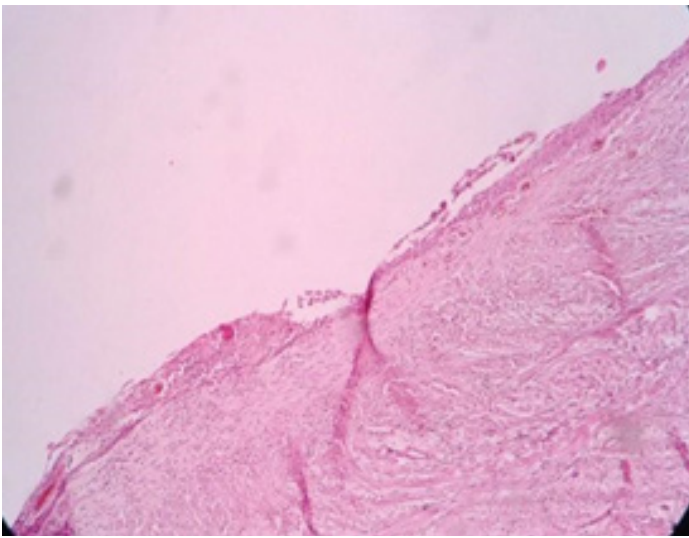


Figure 4: High-power Microscopic view of cystic lining in dentinogenic ghost cell tumor

Discussion

Calcifying odontogenic cyst (COC) comprises 1%–2% of all odontogenic cyst and tumors [7]. and DGCTs, a type of solid variant of COC, are 11.5% among all COC [8].

DGCT affects both jaws with higher ratio in the anterior region of mandible. Clinically, this lesion does not have any symptoms; however, it creates considerable swelling and cause asymmetry of the face, which depends on the size of the lesion [6,9].

DGCTs are histologically identified by odontogenic epithelium islands, which show pseudo-ameloblastoma characteristics in the fibrous tissue, where dentin dysplasia and ghost cells are mostly observed. The neoplastic epithelia of DGCT are considered an aggressive area. A formation of microscopic dentin containing ghost cells might make the final diagnosis of DGCT possible [1].

It is classified into 2 types: intraosseous (central, 83%) and extraosseous (peripheral, 17%) [10]. Central DGCTs are often seen in the mandible and maxillary bones and mostly detected in the canine to first molar teeth region. They appear as swelling without any symptoms, but in some cases, it might cause slight pain [11,12]. Many of the peripheral DGCTs originate from the gingiva in the dentate patients and alveolar mucosa in edentulous patients. It usually appears as a sessile or pedunculated exophytic mass [5,13].

The recurrence rate for this rare tumor is reported to be 1 to 20-year period with a 33-73% invasive behavior [14].

Early diagnosis of DGCT is necessary for better prognosis and due to the difference in the recurrence rate and the potential for malignancy, the treatment plan for both types of DGCT is different. Considering the size and extent of the anatomic spread of the intraosseous lesions, block excision or segmental resection with sufficient safety margin is usually required, and they have a high local recurrence rate after limited local resection or conservative local treatment, while the extraosseous lesions are generally treated by conservative local resection [4]. In the case studied here, the patient underwent surgical enucleation and was put under a yearlong follow-up program. No tumor recurrence was observed and the patient was asymptomatic.

Table 1

Type	Area	Age	Gender	Follow up	Treatment	Recurrence
peripheral COC	Left central incisor	9-year-old	male	one year	surgical removal	no recurrence
COC	right posterior mandible	42 years old	male	6 months	enucleated	no recurrence
GCOC	right mandibular corpus	12-year-old	male	1-year	surgical resection	no recurrence
COC and Ameloblastic Fibroma	left posterior mandible	14-year-old	female	20 months	surgical excision	no recurrence
COC	right side body of the mandible	11-year-old	male	more than 10 years	Marsupialization before enucleation	no recurrence
intraosseous COC	anterior mandible	82-year-old	male	six months	surgical enucleation, a peripheral osteotomy	no recurrence
GCOC	right submandibular area	53-year-old	male	Six month	resection with partial mandibulectomy	no recurrence
CCOT	in the lower right jaw	13-year-old	male	2-year	surgical enucleation	no recurrence
CCOT	left maxillary sinus	16-year-old	male	4-month	Transnasal endoscopic resection	no recurrence
recurrent CCOT	maxillary sinus		male		marsupialization & surgical enucleation	no recurrence

We did a research (mini-Review) for COC case report in last 6 years and we found out that among all the COC that have been reported with age range of 9 to 81 years none of the cases had recurrence and from 3 cases of CCOT all had been free of the tumor and one case of sinus CCOT had been also free of recurrence even with the same treatment procedure as ours [15-24].

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