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Case Report

Multiple Calcifying Odontogenic Cysts Involving the Maxillary Sinus

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Introduction: Calcifying odontogenic cyst (COC) is an uncommon odontogenic lesion, classified in two variants: the cystic variant and neoplastic (solid) variant.

Case Presentation: This case report presents multiple COC, which involved the maxillary bone and sinus in a 30-year-old man. Several of these lesions were cystic, while the others were neoplastic in type, and the lesions were removed surgically.

Discussion: Based on a literature review available on this topic in English, our case study was found to be the first one with multiple COC, showing both the cystic and neoplastic histopathological variant. Considering the high rate of recurrence of neoplastic COC, the patients should benefit from a long follow-up after treatment.

Keywords: Cysts; Odontogenic Cyst; Calcifying; Maxillary Sinus

1. Introduction

The calcifying odontogenic cyst (COC) was first introduced in 1962 by Gorlin et al. as a potential analog of pilomatricoma of the mouth (1). About 15-21% of the cases of COC are peripheral (2, 3), while 79-85% are central (intraosseous). This lesion does not have a propensity toward any specific gender and involves the maxilla and the mandible, equally. It mainly occurs at the canineincisor region (65%). From a histopathological perspective, COC often consists of a fibrous capsule, covered by odontogenic epithelial cells. With a loose arrangement, these cells are surrounded by a basal (peripheral) layer, ranging from cuboidal to columnar shapes. This lesion is similar in its microscopic view to ameloblastoma. However, unlike ameloblastoma, this lesion undergoes the ghost cell change process in the suprabasilar areas (4). Further, ghost cell changes undergo dystrophic calcification. They sometimes form a histology image similar to the dysplastic dentin adjacent to epithelial elements (4). A spectrum of histopathological patterns are proposed in relation to this lesion, encompassing cystic and solid lesions (5). The COC can be treated through enucleation and curettage. After the treatment, prognosis is considered to be good and recurrences are uncommon (4), and malignant changes have also rarely been reported (6,7).

2. Case Presentation

The patient was a 30-year-old man, admitted to the

Maxillofacial Surgery Department of Besat Hospital of Hamadan, Hamadan, Iran, with complaints of a swelling on the right side of the face. The patient did not have any clinical symptoms, such as pain, paresthesia or lymphadenopathy. The consistency of lesion was hard on palpation. The intraoral examination revealed that the right upper canine was absent and teeth 4C21 were mobile to various degrees. The panoramic view revealed several radiolucent-radiopaque well defined lesions from the distal maxillary left central incisor to the right tuberosity. The lesion had caused root resorption of teeth 4C21 and had pushed the impacted canine toward the orbit and the medial nasal wall. There were no perforations observed in the patient's panoramic view and CT scan (Figure 1). On the basis of the clinical and radiological findings, the differential diagnoses were dentigerous cyst, desmoplastic ameloblastoma, central giant-cell granuloma and COC. Following, an incisional biopsy was performed from the lesion. In histopathological examination of the incisional sample, odontogenic cyst was diagnosed. After the initial diagnosis, the patient underwent enucleation surgery under general anesthesia and multiple lesions were removed from the maxilla along with the impacted canine. Lesions included two solid masses from around the apex of teeth 4C21, each with the approximate dimensions of 20×10 × 6 mm (Figure 2), three separate cystic lesions with the dimensions of $2 \times 3 \times 20$ mm and $17 \times 5 \times 5$ mm, and another cystic lesion with a diameter of 30 mm in the maxillary sinus (Figure 3).

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Moreover, five apparently solid lesions, with the overall dimensions of $40 \times 10 \times 10$ mm were removed from the maxilla, with the largest measuring $15 \times 20 \times 10$ mm in dimension (Figure 4). The histopathological examination of the lesions around the apices of teeth 4C21 showed islands and cords of odontogenic epithelium infiltrating into mature connective tissue, composed of two types of cells, a row of peripheral cells, ranging in shape from cuboidal to columnar, and a nucleus away from the basement membrane, with reverse polarity, that surrounded central cells loosely arranged (similar to the stellate reticulum). Single and multiple ghost cells were observed in the center of these islands and sheets, and were also calcified in several areas. All these islands and sheets were located in a fibrous connective tissue. including fibroblasts and collagen bundles. Ghost cells were observed in the center of these islands, and dentinoid were in contact with odontogenic epithelium. Several parts of the background also contained remains of the malassez epithelium and cholesterol clefts (Figure 2). As for the cystic lesions removed from the maxillary sinus, a connective tissue wall of low thickness, with epithelial lining, was observed, containing two types of cells, namely, a row of columnar cells in the basal layer, with a nucleus away from the basement membrane, and the upper cells, looking like the stellate reticulum and loosely arranged. The surface of the epithelium was also partly lined with ghost cells. No daughter cysts or odontogenic epithelium were observed on the connective tissue wall of these cysts (Figure 3). The microscopic view of the five lesions removed from the maxillary bone also looked cystic. Multiple islands of odontogenic epithelium were also observed in the supportive connective tissue of the cysts. Ghost cells had also entirely filled in the cyst lumen. Several of these ghost cells, however, had fallen into the connective tissue of the cyst, and therefore caused foreign body reaction and giant cell accumulation; in a small number of areas, these ghost cells were calcified (Figure 4). The COC diagnosis was therefore confirmed for these lesions. The researcher followed the patient up for 6 months after the treatment and did not find any recurrence.

3. Discussion

Based on our reviews, multiple COC is a rare case reported to date (8). An analysis of 215 cases showed that COC occur among younger Asians in their second and third decades of life, at a rate of 70%, while in Caucasian people, they occur during the later decades of life and at an approximate rate of 53%. Moreover, the lesion tends to involve the maxilla in Asians (65%) and the mandible in white Caucasians (62%) (8), while in our case, the lesion occurred in the maxilla and toward the end of the third decade of the patient's life. In COC, involvement of the maxillary sinus has only rarely been reported to date (1, 9, 10); one of these reports was on a recurrent case

(9). Another one has been a COC with malignant transformation (10) and, among the other cases reported for maxillary sinus involvement, several lesions have been accompanied by resorption of the orbital and zygomatic bones (9). However, no perforation or bone resorption were observed in the present case. The COC can resorb the roots of adjacent teeth and, in approximately one third of cases, is associated with an unerupted tooth (9). The patient's radiograph also revealed root resorption at teeth 4C21 and an impacted canine. Different histopathological classifications have been made for COC. One classification contains four subcategories (4): 1. Nonproliferative cystic: the epithelial lining of the cystic lesion might consist of only a limited number of cell layers, and might hold scattered dentinoids, which is observed in 45% of the cases; 2. Proliferative cystic (ameloblastomatous): the cystic component is often accompanied by multiple daughter cysts in the connective tissue wall; the proliferation of the odontogenic epithelium is expanded to the lumen, imitating a view similar to ameloblasts; 3. Associated with odontoma: tissues similar to odontoma can be observed in the lesion wall (11, 12); and 4. Epithelial dentinogenic ghost cell tumor: this pattern is identified with the presence of cords and islands, similar to the ameloblastoma in the connective tissue, has neoplastic features and also contains various amounts of eosinophilic calcified material (dentinoid) - the origin of the name it has been given (13). Out of the four variants, three histopathologic subtypes were seen in the microscopic evaluation of the case: 1. Non-proliferative cysts in maxillary sinus lesions; 2. Proliferative cysts in maxillary bone lesion; and 3. Dentinogenic ghost cell tumor, in lesions around the apex of teeth 4C21. Gnepp et al. believe that similar to ameloblastoma, there are no differences between the histopathological varieties of COC and their clinical behavior/course (4). However, others believe that the neoplastic COC has a different clinical behavior, with increased recurrence rate compared to the cystic type (9). In this case report, both the cystic and neoplastic variants of COC were observed, and no recurrence was seen after a 6-month follow-up period, although long-term follow-up is recommended.



Figure 1. Panoramic View of the Patient

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Figure 2. Macroscopic and Microscopic View (Hematoxylin and Eosin Stain; Magnification × 400) of Lesions Around the Apices of Teeth



Figure 3. Macroscopic and Microscopic View (Hematoxylin and Eosin Stain; Magnification: ×100) of the Maxillary Sinus Lesions

Figure 4. Macroscopic and Microscopic View (Hematoxylin and Eosin Stain; Magnification: × 100) of the Maxillary Bone Lesions

Based on the literature review available on this topic in English, our case study was found to be the first one with multiple COC showing both the cystic and neoplastic histopathological variant. Considering the high rate of recurrence of neoplastic COC, the patients should be followed-up long after treatment.

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Administrative, technical, and material support: Mohammadreza Jamalpour; Study concept and design: Atefeh Hashemi; Acquisition of data: Masoumeh Zargaran.

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