Calcifying Odontogenic Cyst with Complex Odontoma: Histological and Immunohistochemical Features

Nooshin Mohtasham[1], Amin Rahpeyma[2], Saeedeh Khajeh Ahmadi[3], Mohsen Merati[4]

[1] Oral and Maxillofacial Diseases Research Center, Department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
[2] Dental Research Center, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
[3] Dental Research Center, Department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
[4] Department of Orthodontics and Dental Research Center, Faculty of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran

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Abstract

The calcifying odontogenic cyst (COC) is a rare odontogenic cyst. Only 2% of all odontogenic cysts and tumors are COC. COC associated with odontoma (COCaO) reported in 24% of COCs. COCaO presents a greater incidence in female, with a ratio of 2 to 1. The highest incidence of COCaO occurs during the second decade with a mean age of 16 years, most frequently occurring in the maxilla (61.5%). Here, we describe a classic case of COCaO of the maxillary incisor-canine region in 17-year-old girl, and discuss the clinicopathological features and immunohistochemical finding of this tumor.

Key Words: Calcifying odontogenic cyst, histopathologic feature, immunohistochemical straining, odontoma.

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Introduction

The calcifying odontogenic cyst (COC) was first described as a distinct entity by Gorlin et al. (1962) and Gold (1963) (1). They reported 15 cases of this entity that were called as an intraoral marlherbe's calcifying epithelium (pilomatricoma). Several variants of the cyst may be seen are cystic and neoplastic variants; peripheral and central types. Rare cases of malignant transformation have been reported (2). The basic features of COC consist of 1) cystic, nonproliferative 2) cystic, proliferative/ameloblastomatous 3) odontoma-associated 4) epithelial odontogenic ghost cell tumor (3).

Various terms have been used for description of this lesion such as COC (4), keratinizing calcifying odontogenic cyst (KCOC) (1), calcifying ghost cell odontogenic tumor (CGCOT) (5), calcifying cystic odontogenic tumor (CCOT) (6,7), dentinogenic ghost cell tumor (DGCT) (8), epithelial odontogenic ghost cell tumor (EOGCT) (9), odontogenic ghost cell tumor (OGCT) (10), odontocalcifying odontogenic cyst (11).

Cystic variant composed of 85% of the cases (12). In addition, occasionally dysplastic dentine and an area of dental hard tissue formation and resembling odontoma can be found.

Odontoma with COC present in 24% of COCs (13). Melanin pigmentation and clear cell also reported in epithelium (2). Radiographically, COC appear as a well -defined unilocular lesion, a few case of multilocular lesion also have been reported (14). The frequency of

association impacted teeth is approximate 10-32% (15). Multiple impacted teeth are a well known feature of COC (4). Radicular resorption is uncommon (2). COC is usually treated by enucleation and curettage. Recurrence is uncommon.

Case Report

A 17-year-old girl without any remarkable medical history, with two-month history of swelling in the left maxilla region was referred to Department of Oral and Maxillofacial surgery for definitive diagnosis. Intraoral examination showed a painless buccal expansion on the left maxilla from medial line to the canine region. The associated teeth were vital without pathologic mobility. A well-defined unilocular radiolucent area extending from medial line to the canine region, with radiopaque mass, was observed in the panoramic radiograph (OPG). Root divergence of lateral and canine teeth without root resorption was present (Fig. 1). A thick wall cyst was removed under local anesthesia. Histologic examination of biopsy tissue showed a cystic cavity lined by proliferation of odontogenic epithelial cells. Ghost cells were present within epithelial layer (Fig. 2). Odontoma-like structure were present in the wall of cyst (Fig. 3). Immunohistochemical staining for CK7 was negative (Fig. 4). Histopathologic diagnosis was calcifying odontogenic cyst with complex odontoma (COCaO). A 2-year follow-up after cyst excision showed no recurrence.

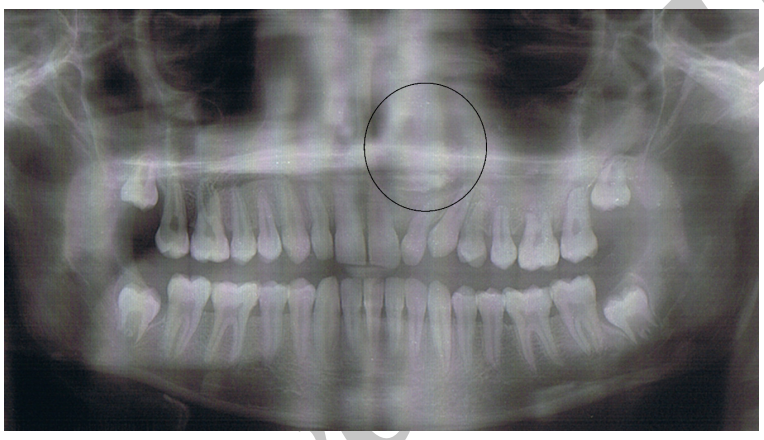

Figure 1. Panoramic radiograph: COC associated with odontoma (COCaO) with radiopaque mass

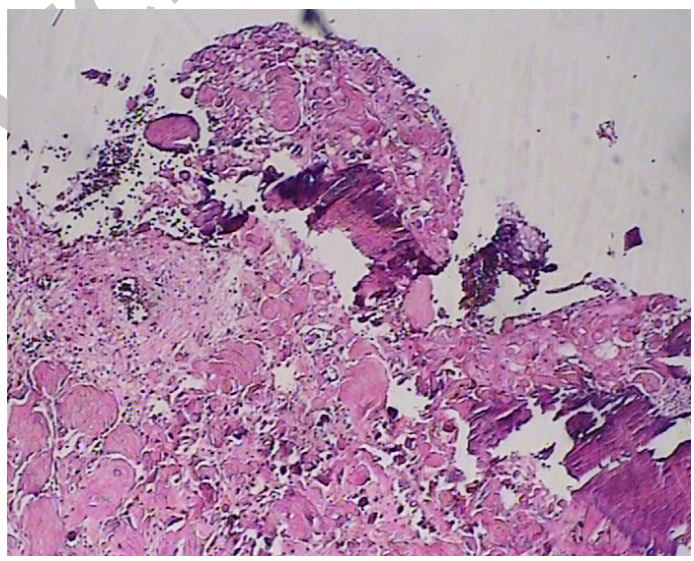

Figure 2. Histopathology feature and detail of the ghost cells (H&E staining, original magnification ×100)

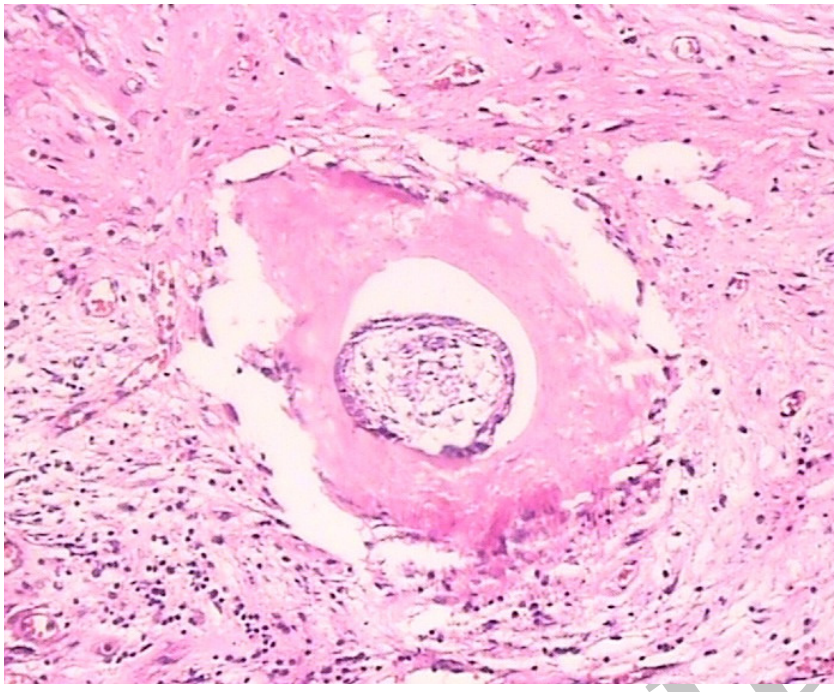

Figure 3. Histopathology feature and detail of the complex odontoma (H&E staining, original magnification ×400)

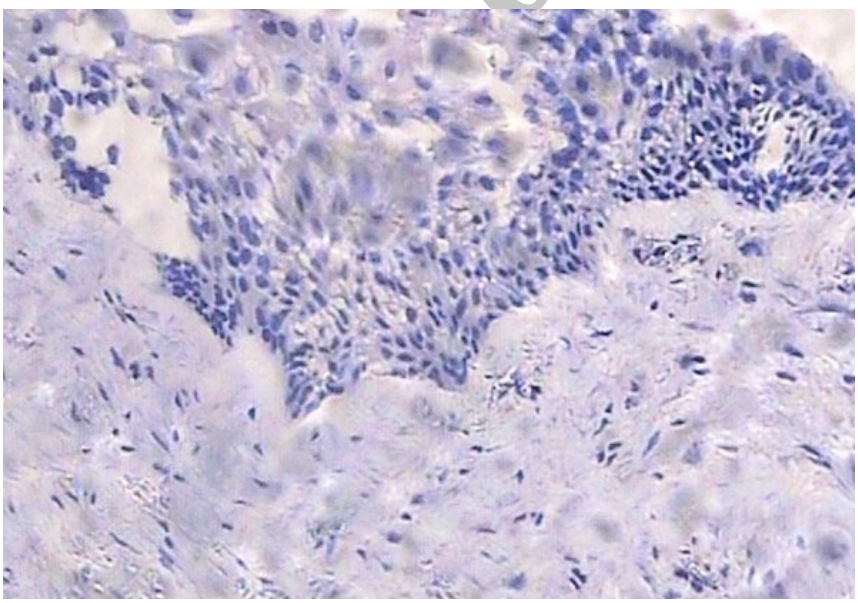

Figure 4. Immunohistochemical staining is negative for CK7 (IHC staining, original magnification ×100)

Discussion

COC may be arising from odontogenic epithelial remnants within the jaw or gingival. There is no gender predilection. It occurs in the maxilla and mandible with equal frequency (4). Rare peripheral variant of this lesion have been described.

COC may occur at any age with the prevalence peaks in the second and third decades (mean age, 33 years) but COC associated with odontoma (COCaO) occurs in younger persons, with a mean age of 17 years. In pathological feature, COC composed of a fibrous capsule that is lined with a proliferation of odontogenic epithelial cells and the ghost cell change that characterizes in these lesions (13).

COC rarely can occur in conjunction with other odontogenic tumors such as ameloblastoma, ameloblastic fibroma, amelobastic fibro-odontoma and adenomatoid odontogenic tumor (16). Radioluscency accompanying odontoma or presence of soft tissue with odontoma during biopsy or operation, guided clinician toward four differential diagnosis including: cystic odontoma, COCaO, amelobelastic fibro-odontoma, odontoameloblastoma. Unlike other lesions, surgical resection with safe bony margin is recommended for odontoameloblastoma. However, conservative treatment (enucleation) is required for another lesion. COCaO is similar to the cystic COC, it has tooth like structures in the connective tissue of the cyst (11).

COC may occur in association with odontoma; Buchner (2) shows this association in 35% of his cases, Nagao et al. (17) in 22% and Shamaskin et al. (18) in 47%. Radiographically COCaO appears as a mixed radiolucent-radiopaque lesion (80%) occasionally calcifications cannot be observed on OPG but can be visualized in CT scan (19). Treatment of COCaO consists of conservative enucleation and the prognosis is excellent. Recurrence after rather conservative therapy is uncommon (2).

Roudrigues-Fregnani et al. (20) showed that the epithelial cells of COC express antibodies directed against cytokeratins 7, 8, 14, and 19 but staining for CK7 was negative in present case. Finally, this finding denotes more research in this field.

Conclusion

Microscopic evaluation of soft tissue associated with odontoma is important; because this tissue can be dentigerous cyst, COC or ameloblastic fibroma. In this situation conservation complete removal of tissue recommended, but if soft tissue component to be ameloblastoma, more radical treatment and longer follow-up is needed.

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Corresponding Author:
Saeedeh Khajeh Ahmadi
Dental Research Center
Mashhad University of Medical Sciences
Vakilabad Blvd, Mashhad, Iran
P.O. Box: 91735-984
Tel: +98-511-8829501
Fax: +98-511-8829500
Email: Khajehahmadis@mums.ac.ir