Case Report

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A Calcifying Odontogenic Cyst Associated with Complex Odontoma: A Case Report

A. Shiva (DDS)[1], K. Nosrati (DDS)[*2]

1. *Department of Pathology, Oral and Maxillofacial Pathology, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, I.R. Iran*
2. *Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Babol University of Medical Sciences, Babol, I.R. Iran*

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ABSTRACT

BACKGROUND AND OBJECTIVE: Calcifying odontogenic cyst is an uncommon developmental cyst. It accounts for only 1% of jaw cysts and uncommonly occurs with odontoma. Herein, we report a case of calcifying odontogenic cyst, associated with complex odontoma.

CASE REPORT: The patient was a 12-year-old girl with complaints of painless swelling in the mandibular first premolar. In radiographic examination, a radiolucent-radiopaque lesion was reported. The lesion, as well as the impacted tooth, was surgically removed via enucleation. In pathological analysis, an odontogenic cyst with ghost cells and complex odontoma was observed. Enucleation was performed for treatment and no recurrence was reported during the one-year follow-up.

CONCLUSION: Clinical and radiographic findings of odontogenic lesions are not exclusive. Therefore, more attention should be paid to the fact that the type of lesion associated with Gorlin cyst determines the clinical course, treatment, and prognosis of complex lesions.

KEY WORDS: Calcifying Odontogenic Cyst, Ghost Cells, Gorlin Cyst, Complex Odontoma.

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Introduction

Calcifying odontogenic cyst (COC) was first described by Gorlin et al. in 1962 and Gold in 1963 (1). COC is an uncommon odontogenic lesion accounting for about 3% of oral lesions and 1% of jaw cysts (3). Although COC is generally considered a cyst, some researchers and even World Health Organization (WHO, 1992) classified all its diversities within the group of neoplasms, which may be either infiltrative or malignant (3). In addition, the cystic type of this lesion may be associated with other odontogenic tumors such as odontoma, ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibroma, and ameloblastic fibro-odontoma (4).In a study on 349 cases of odontogenic tumors performed by Mosqueda-Taylor in Mexico, only 6.8% of the evaluated cases were COC (5). Also,

[*] **Corresponding Author; A. Shiva (DDS)**
Address: Faculty of Dentistry, Mazandaran University of Medical Sciences, Khazar Boulevard, Sari, I.R.Iran .
Tel: +98 11 33244894
E-mail: atenashiva@yahoo.com

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in two studies by Knezevic and Buchner, the prevalence of Gorlin cyst associated with odontoma was 20% and 35%, respectively (6, 7). COC is mainly an intraosseous lesion with **equal** frequency in the maxilla and mandible. About 65% of these lesions are found near incisors and canine teeth, and they are associated with impacted teeth (mostly canines) in 10-32% of cases (8). In a study by Seyed Majidi et al. on 21 COCs, most cases were reported in the posterior mandible, female subjects, and during the second decade of life; also, 100% of cases had a cystic appearance (9).

COS often occurs in the second or third decade of life, although the neoplastic type has been reported in older patients. COCs associated with odontoma are prevalent among younger patients with the mean age of 17 years. Clinically, COC with odontoma is often associated with slow growth and painless swelling (8). Sometimes, it occurs without swelling and only delayed tooth eruption is observed. In some cases, COC may be accidently seen during routine radiography (10).

Clinical and radiographic maifestations of COCs, associated with odontogenic tumors, are not exclusive; thus, histopathologic examination is required for detection (11). Histopathologically, Gorlin cyst contains cystic epithelium with a cubic or cylindrical-shaped basal layer, a hypochromic nucleus, and reverse polarity. This cyst is seen in the anterior layers of eosinophilic ghost cells with a distinct cellular outline (without a nucleus). The connective tissues of COC are usually fibrovascular.

Microscopically, ghost cells are the most distinctive feature of these lesions. Moreover, in some cases, radiopaque structures may be seen as irregular calcification or tooth-like masses inside the lesion, which is similar to complex or compound odontoma (8, 12).

The clinical, radiological, pathological, and pathogenic importance of COC associated with complex odontoma has not been recognized yet; moreover, the simultaneous occurrence of these two lesions is uncommon. Therefore, we decided to report a case of COC and compare the properties of this cyst with other previously reported features.

Case report

The patient was a 12-year-old girl with complaints of painless swelling in the left mandibular first premolar, referring to an oral and maxillofacial surgeon in Sari, Iran. As the patient stated, the swelling had started approximately two to three months ago. As the intraoral examination indicated, the lesion occurred as a swelling with firm consistency in the buccal cortex on the left side of the mandibular first premolar (1.5×2 cm). The mucosal surface was normal, and the patient's medical history indicated no diseases or medication use. The panoramic image of the patient indicated a unilocular radiolucent (along with adiopaque masses within it) with a distinct outline in the first impacted premolar. Considering the radiographic findings, the differential diagnosis included odontoma and ameloblastic fibro-odontoma (fig 1).



Figure 1. The radiographic view of multifocal unilocular radiolucent lesion in the left impacted first premolar in the mandible

The lesion, as well as the impacted tooth, was surgically removed via enucleation and was sent to the oral pathology laboratory (kept in 10% formalin). According to the macroscopic examination, the lesion was composed of soft and hard tissues, which were in two separate parts. The soft tissue was creamish brown (1.1×8.0×9.0 cm), whereas the hard tissue had firm

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consistency (0.5×6.0×6.0 cm in total). The hard tissue was placed in 10% formic acid for decalcification for two days and was then cut. In the histopathologic examination of the lesion, using hematoxylin and eosin (H&E) staining, an ontogenic epithelium, similar to a cystic tissue, with cubic-to-cylindrical basal layer, a hyperchromic nucleus, reverse polarity, and surface ghost cells, was observed. The connective tissue of the cyst was fibrovascular, containing scattered chronic inflammatory cells (fig 2). Also, irregular calcified structures of enamel, dentinal tubules, and connective tissues associated with complex odontoma were observed (fig 3). Finally, COC (Gorlin cyst) with complex odontoma was diagnosed. During the one-year follow-up of the patient after surgery, no recurrence was reported.

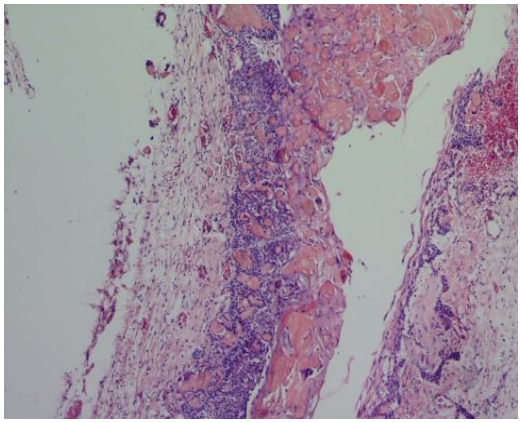


Figure 2. H&E staining of Gorlin cyst; the epithelial surface of Gorlin cyst and ghost cells (10x magnification)

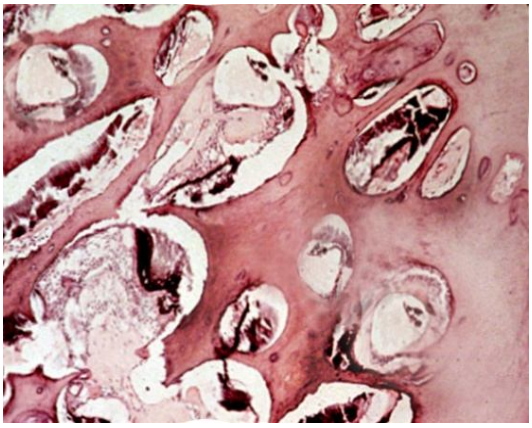


Figure 3. H&E staining of the Gorlin cyst with complex odontoma (40x magnification)

Discussion

The most important feature of COC (Gorlin cyst), especially if associated with odontoma, is its uncommon nature and variable manifestations and clinical behaviors (13). Some COCs appear as non-neoplastic cysts, while some are known as epithelial odontogenic ghost cell tumors; these cases do not have cystic properties and may be infiltrative or even malignant (neoplasms) (8). In 1981, Praetorius et al. classified Gorlin cysts as cystic neoplasms and categorized them into three groups:

1. Simple unicystic Gorlin cysts with or without calcified particles;
2. Unicystic odontoma-producing type with all the characteristics of type one, except that the calcified tissue is similar to odontoma; and
3. The unicystic ameloblastomatous type, which is characterized by the growth of ameloblastic epithelium in the cyst wall and inside the lumen (an odontogenic tumor) (14).

COC is often reported in the second or third decade of life in people younger than 40 years of age. If accompanied by odontoma, the neoplastic type may occur in younger patients with the mean age of 17 years (6). Hirshberg et al. clinically and histologically examined 52 cases of Gorlin cyst, associated with odontoma. The prevalence of these complex lesions was higher in the upper jaw, female subjects, and at the age of 16 years (15); this was similar to the findings in the present report (the patient was 12 years old). Also, Nevile et al. indicated that this complex variant is often unicystic and is more often seen with compound odontoma than the complex type (8); this was not in agreement with the present case. Majidi et al. in 2010 reported a case of Gorlin cyst, associated with odontoma in a 15-year-old boy. The patient's chief complaint was painless swelling in the upper jaw, similar to our case (2). Additionally, Nakayama et al. showed that two lesions simultaneously occur in the first or second decade of life, which is similar to the present findings (16).

In our study, odontoma was outside the Gorlin cyst, which was in agreement with a case reported by Seifi et al. (17). Gallana-Alverz et al. reported a case of

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Gorlin cyst associated with odontoma in a 19-year-old boy. In radiography, a unilocular radiolucentat lesion was seen in the canine, which was surrounded by radiopaque masses (10). Moreover, Pisota at al. reported complex lesions in a 36-year-old man with complaints of painless swelling in the lower premolar and the presence of primary canine in the area (18). Considering the pathogenesis of Gorlin cysts with complex odontoma, the epithelium of Gorlin cyst may be proliferative (19); in fact, due to the effect of epithelium on connective tissues, odontoma is secondarily formed. Another possibility is that these two lesions appear together in one place since other odontogenic tumors such as ameloblastoma have been also seen with Gorlin cysts (2).

COC (Gorlin cyst) is an odontogenic developmental lesion, with the potential for recurrence (10). In our case, treatment was performed with enucleation and removal of the involved tooth. However, if the cyst is accompanied by other tomoral lesions with aggressive behaviors, a more invasive treatment should be applied (20).

Prognosis of complex COC is dependent on the associated tumor; also, the histopatological examination of all removed lesions is required. Finally, it should be noted that the type of lesion accompanying Gorlin cyst determines the clinical course, treatment method, and ultimately the prognosis of compound lesions. In fact, detailed histopathlogical examination and definitive diagnosis can lead to successful treatment.

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