

## Glandular Odontogenic Cyst Associated with Impacted Tooth: A Case Report

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### Abstract

Glandular odontogenic cyst (GOC) is an uncommon developmental cyst. It accounts for 0.012% to 1.3% of all the jaw cysts; its prevalence is 0.17%. It was described by Gardner et al. in 1988 as a distinct entity. GOC has a slight predilection for mandible and affect more commonly in the middle-aged patients. Radiographic and clinical features of this cyst are not pathognomonic or specific. Only the histopathological examinations allow for certain diagnosis of the cyst. The increased recurrence rate can be due to cell kinetics in the lining epithelium, multilocularity and incomplete removal of the lining following conservative treatment. In this report, we describe a case of GOC of the mandible associated with impacted tooth. The clinical diagnosis of dentigerous cyst was made, whereas histopathological examination results in a diagnosis of glandular odontogenic cyst. Based on clinical and radiological examination the diagnosis of other lesions such as radicular cyst, dentigerous cyst, and ameloblastoma may be made but histologically GOC shows certain characteristic features.

**Key Words:** Glandular odontogenic cyst, impacted tooth, mandible.

### Introduction

Glandular odontogenic cyst (GOC) is an uncommon developmental jaw cyst with special histopathological features and biological behavior (1,2). In 1987, Padyachee and Van Wyk (3) reported the first two cases of this cyst as a Sialo-Odontogenic Cyst.

This cyst was established as a distinct entity by Gardner et al. (4) in 1988, under the term of a glandular odontogenic cyst. In 1992, the GOC was included in the list of histologic type of odontogenic cysts by the world health organization (WHO) with the term of "Sialo-Odontogenic Cyst" or a "Glandular Odontogenic Cyst". The frequency of GOC has been reported to be 0.012% to 1.3% of all the jaw cysts and its prevalence is 0.17% (1). Clinically the most common site of occurrence is mandible, especially in the anterior region. GOC has a slight male predilection and occurs primarily in middle-aged patients (1,5). Radiographically, these cysts reveal a unilocular or multilocular radiolucency with well-defined borders (6).

On the base of clinical and radiological examination, other lesions such as radicular cyst, dentigerous cyst, and ameloblastoma might be considered as the other differential diagnoses (7), but histologically GOC shows certain characteristic features. These features include nonkeratinized stratified squamous lining epithelium, focal thickenings (plaques) within the lining, eosinophilic cuboidal or columnar cells that may be ciliated, mucous cells and mucin pools, and interepithelial gland-like structures (3,4,8,9).

Treatment of GOC includes curettage and enucleation, although some authors believe marginal resection due to a tendency of recurrence after curettage and enucleation (5). In this report, we present a case of GOC associated with impacted tooth. The provisional

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diagnosis of dentigerous cyst was made, whereas histopathologic examination diagnosed GOC.

### Case Report

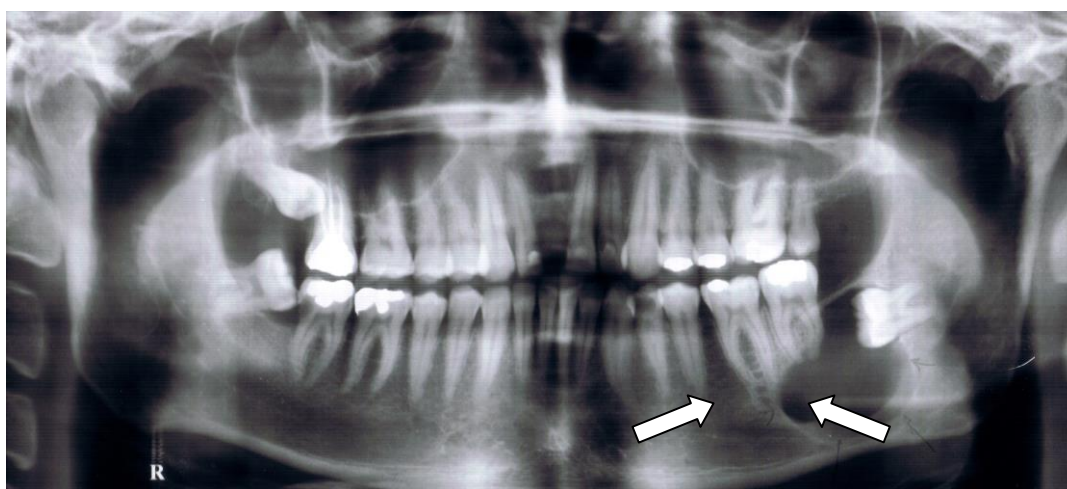
A 24-year old man was referred to Mashhad Faculty of Dentistry, Mashhad, Iran on November 17, 2009 for the evaluation and management of an asymptomatic, firm swelling of the left mandible.

Intraoral examination showed a swelling in the third molar region. A panoramic radiograph demonstrated a well-defined radiolucency surrounding the left mandibular impacted third molar (Fig. 1).

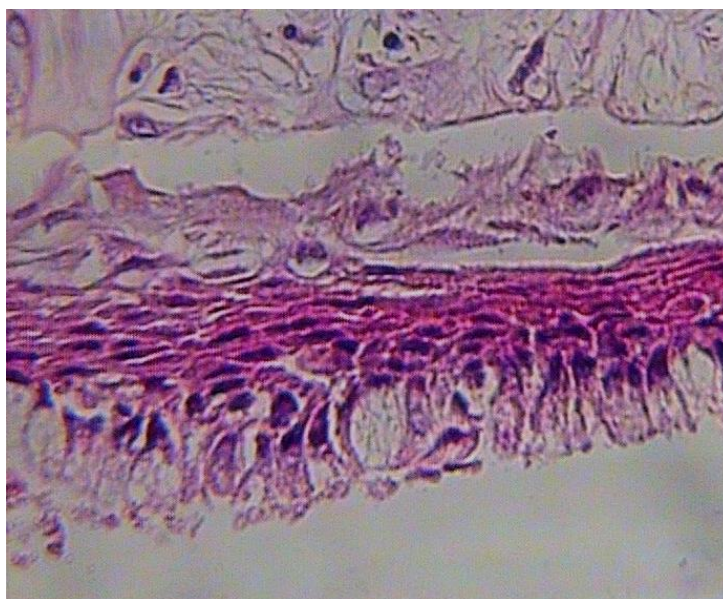
The patient was prepared for the surgical removal of the impacted tooth and the enucleation of the lesion with a provisional diagnosis of dentigerous cyst.

Grossly, the specimen consisted of a brownish-cream cystic mass which was elastic and measured  $30 \times 10 \times 2$  mm.

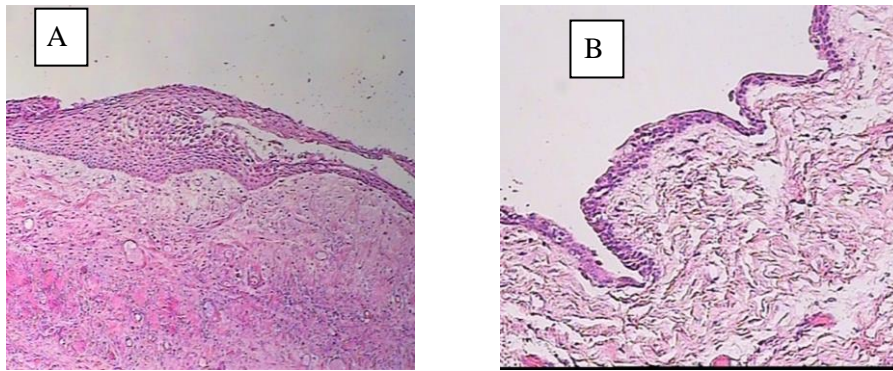
Histologic examination of the biopsy demonstrated a cystic lesion lined by nonkeratinized stratified squamous epithelium that exhibited the superficial layer of ciliated columnar cells in parts (Fig. 2). Papillary projections into the cyst's lumen and focal epithelial thickenings (plaques) were also observed (Fig 3). Mucous cells with interepithelial mucous pools and gland-like structures were identified throughout the lining epithelium (Figs. 4 and 5). The patient called for three months later and it was observed that the lesion had been healed altogether.



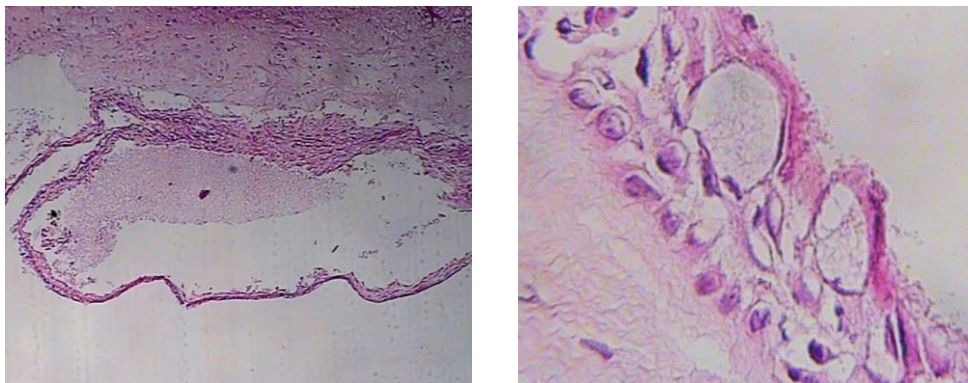
**Figure 1.** A panoramic radiograph shows a well-defined radiolucency, surrounding the left mandibular impacted third molar (arrows)



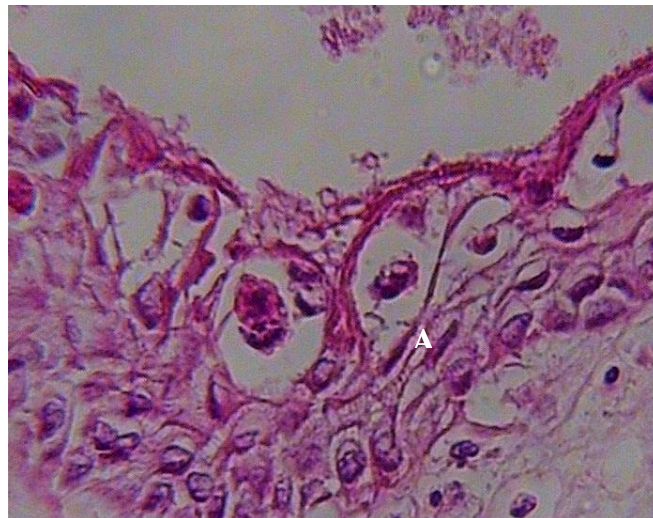
**Figure 2.** Cuboidal eosinophilic cells or "hob-nail" cells and superficial layer of the cyst with ciliated columnar cells (H&E Original magnification:  $\times 400$ )



**Figure 3.** Focal epithelial thickenings (plaque) (A), crypt formation and papillary projections are seen in this section (B) (H&E, Original magnification:  $\times 100$ )



**Figure 4.** Mucous cells and interepithelial mucous pools (H&E, Original magnification A:  $\times 400$  B:  $\times 40$ )



**Figure 5.** Gland-like structures throughout the lining epithelium (H&E, Original magnification:  $\times 400$ )

### Discussion

GOC may have a wide clinicopathologic spectrum ranging from a benign small pathology to a destructive neoplasm such as central muoepidermoid carcinoma.

Clinically, the most common site of occurrence is mandible (85%), especially in its anterior region (5).

GOC does not display specific or pathognomonic radiographical features. It may be present as a multilocular or unilocular radiolucency with well-

defined border, or as perifollicular radiolucency, simulating a hyperplastic follicle or dentigerous cyst (8). Therefore, regarding clinical radiological examination, the diagnosis of other lesions such as radicular cyst, odontogenic keratocyst (OKC), dentigerous cyst and ameloblastoma is made. Only the histopathological examination allows for certain diagnosis of the cyst (7).

In the present case, the clinical diagnosis of dentigerous cyst was made; whereas, histopathologic examination diagnosed a GOC.

Qin et al. (10) reported five maxillary cases contained unerupted teeth. Shimoyama and Horie (11) also reported a cyst in which clinical diagnosis was dentigerous cyst, but histopathological features of were consistent with glandular odontogenic cyst.

According to previous studies GOC is seldom related to unerupted teeth. Histopathological features of the present case satisfied major and minor Kalpan et al. (12) diagnostic criteria for GOC including squamous epithelial lining, flat interface, variation in thickness of the lining with or without epithelial spheres or whorls, no palisade, cuboidal eosinophilic cells or "hob-nail" cells, mucous goblet cells with interepithelial mucous pools with or without crypts lined by mucous-producing cells and interepithelial glandular microcysts or duct like structures as major criteria and papillary projections, ciliated cells, and clear or vacuolated cells in basal or spinous layer as minor criteria. These histological features were also described by other authors (3,4,6).

In GOC, mucous cells are remarkably abundant and the papillary fronds or projections are most exceptional (12). Low grade mucoepidermoid carcinoma (LGMEC) shares a number of histopathological similarities with GOC. In fact, it has been suggested that many cases formerly diagnosed as central MEC can be the examples of GOC, and also some LGMECs would have originated from GOCs (13-15).

Some authors suggested that the distinguishing feature in GOC is the typical thin epithelial lining without any solid epithelial proliferation as seen in MEC. Moreover, swirling spherical (epithelial plaque) that are often seen in GOC are not observed in MEC (16).

In cases where diagnosis of GOC or LGME cannot be made based on the morphological features alone, especially in small incisional biopsy samples, mammary serine protease inhibitor (maspin) immunolocalization can be used to distinguish these two lesions. The high levels of aggregates maspin in the epithelial-mucous cells (in both cytoplasm and nuclei) in LGMEC may serve as a tool to differentiate it from GOC (13).

Pires et al. (15) have demonstrated CK expression in GOC and central MEC. They found differences in CK18 and 19. CK18 is expressed by all MECs, but it is

only expressed by 30% of the GOCs, whereas CK19 is expressed by all GOCs and only 50% of the central MECs. They suggested that GOC and central MEC are distinct entities that expression CKs 18 and 19 can be useful adjunctive tools in differentiating these two lesions.

Immunohistochemical studies using cytokeratin 7, 13, 14, and 19 and their positivity strongly support the odontogenic nature (17).

The detection of osteodentin and negative reaction for EMA in the glandular structure show that these features are not glandular origin and support the concept of odontogenic differentiation in GOC (1).

The immunohistochemical expression of the apoptosis- inhibiting protein bcl-2, the cell cycle-related antigen ki-67 and the P53 which is involved both in cell cycle and apoptosis regulation was compared between GOC and dentigerous cyst by I. Tosis et al. (2). The percentage of Ki-67 and P53- positive cells was lower in GOC. They suggested that the biological behavior of GOCs may be associated with deregulation of cell death in the lining epithelium, indicated by increased expression of bcl2, while cell proliferation and P53 do not play a significant role.

The aggressive biologic behavior of the GOC and it's propensity for recurrence might be associated with, cell kinetics in the lining epithelium, as has been demonstrated in OKC (17). It has been also demonstrated that the rate of recurrence increases with the radiographic complexity of the cyst (11). Conservative treatment method is another factor which is responsible for recurrence rate (1). The treatment of choice for GOC has ranged from conservative approach (enucleation, marsupialization, curettage with or without peripheral ostectomy) to marginal resection and segmental resection. Some authors suggest more aggressive removal and long follow-up is necessary (1,5,18).

In conclusion, clinical and radiographic examination of the lesion may result in the diagnosis of other lesions such as radicular cyst, dentigerous cyst and ameloblastoma may be made, but histologically GOC shows certain characteristic features.

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