

# Glandular Odontogenic Cyst: A Case Report

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Received 2015 January 05; Revised 2015 January 26; Accepted 2015 February 18.

## Abstract

**Introduction:** The glandular odontogenic cyst (GOC) is a rare, locally aggressive and potentially recurrent cyst of jaws which lead in diagnostic and therapeutic challenges. Since 1992, 111 cases of GOC have been reported, with 0.2% incidence among odontogenic cysts.

**Case Presentation:** The patient was a 33-year-old woman who came to the department of oral medicine with a complaint of facial swelling. After taking her history, and conducting clinical, radiological, and histopathologic examinations, the diagnosis of GOC was made.

**Conclusions:** Glandular odontogenic cyst is an uncommon and relatively aggressive cyst with a high recurrence rate. Clinical and radiological evaluations are needed together with careful histopathologic evaluations for diagnosis. Oral medicine specialists should be aware of its characteristic features and make an accurate diagnosis so appropriate management can follow.

**Keywords:** Odontogenic Cysts, Mandible, Case Report

## 1. Introduction

Several classifications for odontogenic cysts have been published. Odontogenic cysts can be best categorized into inflammatory and developmental types, based on their origin and pathogenesis. Development of inflammatory cysts is due to inflammation, but the initiating factors for their formation are still unknown. The different types cysts could distinguish with either clinical signs and symptoms or biologic behavior (recurrence, aggressive or expansive growth, and malignant potential). Diagnosis is on the basis of histopathologic findings, but clinical and radiographic assessments are also necessary. Reports of carcinomas growing in the cystic wall increase the need for biopsy (1).

GOC is an uncommon cyst of the jaws, locally aggressive and with potentially recurrent, which poses a diagnostic and therapeutic challenge (2). Padayachee and Van Wyk described two cystic lesions with unknown histologic features in 1987 and called it "sialo-odontogenic cyst" (3). One year later, Gardner et al. described eight cases with similar histological appearance and suggested the term "glandular odontogenic cyst" (4). In 1992, the WHO named the GOC and categorized it as a developmental odontogenic cyst (5).

The most common site of occurrence is the mandible, especially in the anterior region, presenting as an asymptomatic swelling. This lesion appears as an unilocular

or multilocular radiolucency, usually with well-defined in some cases scalloped borders. However, there is no pathognomonic radiological feature for GOC. Lack of universally accepted exact microscopic criteria necessary for diagnosis, leads to the issue that this lesion may not be as rare as it has been considered. It has been suggested that many cases diagnosed before as central mucoepidermoid carcinoma (CMEC) could be cases of GOC, and some low-grade CMECs may have originated from GOCs. The reason is that a considerable overlap between histological features of GOC and CMEC is present, and some authors suggest the hypothesis that GOC and CMEC of the jaws represent a spectrum of one disease. Thus, several attempts to use molecular markers to facilitate the diagnosis were made. Some hopeful markers were cytokeratins (CK), mostly CKs 18 and 19, whose expression profile was found to be different in these two lesions (6). Since 1992, 111 cases of GOC have been reported, with 0.2% incidence among odontogenic cysts (7). In this study, we report a case of GOC.

## 2. Case Presentation

A 33-year-old woman came to the Department of Oral Medicine, Hamadan University of Medical Sciences, with a complaint of anterior mandibular swelling for a month. According to the patient, the lesion size was not changed

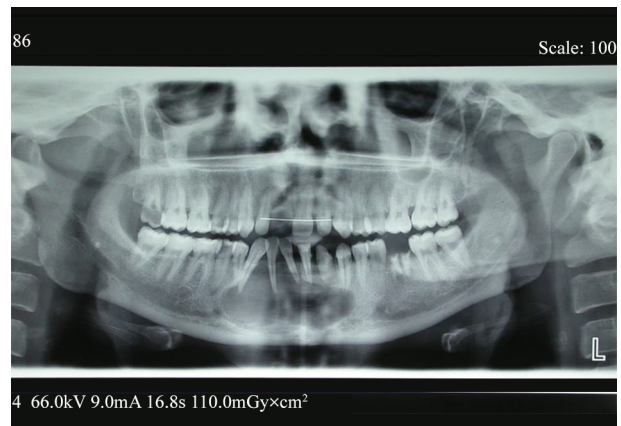
much and there was no pain, pus discharge or bleeding. In her medical history, the patient was healthy and free of any disease. In the extra oral examination, a swelling in the anterior mandible and mental area with a slight tendency to the right was observed. There was no cervical lymphadenopathy. In an intraoral examination, a buccal expansion was found in the anterior mandible with some tendency to the right, from the left central incisor to the right canine. Consistency in most areas was bony hard, but some blue tint areas were soft. There was no pain, paresthesias, bleeding, or pus and just a little tenderness. The patient had poor oral hygiene, and calculus and abundant plaque masses were observed. Involved teeth were vital in the examination and no mobility was observed. Left to right maxillary lateral incisors were splinted together, so the patient was questioned about it and cited a history of trauma two months earlier (Figure 1).



**Figure 1.** Intraoral View of the Lesion

After examination, panoramic and occlusal radiographs were requested. In the radiography, a multilocular radiolucent lesion was observed in the anterior of mandible with a well-defined border from the mesial of the left first premolar to the mesial of the right second premolar and in vertical dimension from the crest to near the lower border of the mandible. The lesion scalloped between the roots of the central, lateral and canine and displaced them. Blunting of the left central incisor and right first premolar root tips were observed. There was buccal and lingual expansion and perforation was seen in some areas of the buccal cortex. Some fine internal septations were seen (Figures 2 and 3).

A provisional diagnosis of central giant cell granuloma and keratinized odontogenic tumor were made. Aspiration was performed from the area with soft consistency,



**Figure 2.** Panoramic View of the Lesion



**Figure 3.** Occlusal View of the Lesion

for assessing content, and 1 cc cystic fluid found and the differential diagnosis limited to cystic lesions (Figure 4). After performing the above steps and for proper management, the patient was referred to the department of maxillofacial surgery. For pathologic assessment, elastic tissue with a cystlike wall was prepared and stained with Hematoxylin and Eosin (H & E), Microscopic evaluation revealed an odontogenic cyst with a different epithelial lining consisted of ulcerated non-keratinized stratified squamous epithelial cell in most areas and ciliated cylindrical and cubic lining in some others, Some goblet cells were detectable. The relationship between epithelium and stroma was flat, and in the connective tissue stroma, middle to abundant chronic inflammatory infiltration was

present(Figure 5).



Figure 4. Aspirated Fluid

### 3. Discussion

The glandular odontogenic cyst is an uncommon lesion. Its frequency rate ranges from 0.012% to 1.3% of all jaw cysts, and its prevalence is 0.17% (6). It has a slight male predilection with a male to female ratio of 1.3:1, and the most commonly reported site is the anterior mandible. It usually occurs over a wide age range of 10-90 years old (mean age of 49.5 years old). Radiographic features consist of a well-defined cyst-like unilocular or multilocular radiolucency, often with scalloped sclerotic borders. Additionally, root resorption and tooth displacement with cortical perforation may exist, leading to invasion of the cyst into the adjacent soft tissues. These data highlight the absence of a pathognomonic clinical or radiographic presentation of GOC, considering that there are similarities in other lesions, such as odontogenic keratocysts, unicystic or multicystic ameloblastoma, CMEC, lateral epithelial cysts and botryoid odontogenic cysts (BOC). The consequence was that the diagnosis of this cyst on the basis of physical and radiological examination was virtually impossible.

The histogenesis of GOC remains uncertain; in fact, it was initially suggested to develop from intraosseous salivary gland tissue (7). Now most authors believe that these cysts originate from odontogenic epithelium (6). Histologic characteristics of GOC consist of a cyst wall lining of non-keratinized epithelium with papillary projections, nodular thickenings, mucous-filled clefts, and "mucous lakes." In addition, there are cuboidal basal cells, which are sometimes vacuolated. However, central mucoepidermoid carcinoma (CMEC), with its significant histopatho-

logical similarities, is the most important diagnosis to be considered (7).

Histopathologically, some authors suggested that GOC may show variable histologic features ranging from cystic lesions similar to BOC to lesions resembling low-grade CMEC (6). Kaplan et al. divided the microscopic characteristics of GOC into major and minor criteria, with the purpose of facilitating the diagnosis (2). However, the practical applicability of the above suggested criteria may include some difficulties. Due to the strong similarities between GOC and CMEC, immunohistochemistry may help with a diagnosis (6). Pires et al. demonstrated that there are differences in the expression of cytokeratins (CKs) in GOC and CMEC, and suggested that CKs 18 and 19 could be used for distinguishing between the two (8).

In this study, the presented case did not match in terms of gender and age. Men were reported slightly superior. However, Fowler and colleagues in 2011 reported that there is no gender preference (9). Although, an age range of 10 to 90 years old with a mean of 49.5 is reported, the current case was a 33-year-old who was younger than the reported mean age. The site of involvement was the anterior mandible, which is compatible with characteristic glandular odontogenic cysts. Some major and minor histopathologic criteria were observed, including a non-keratinized squamous epithelial lining having a flat interface with connective tissue, and goblet, cubic and ciliated columnar cells.

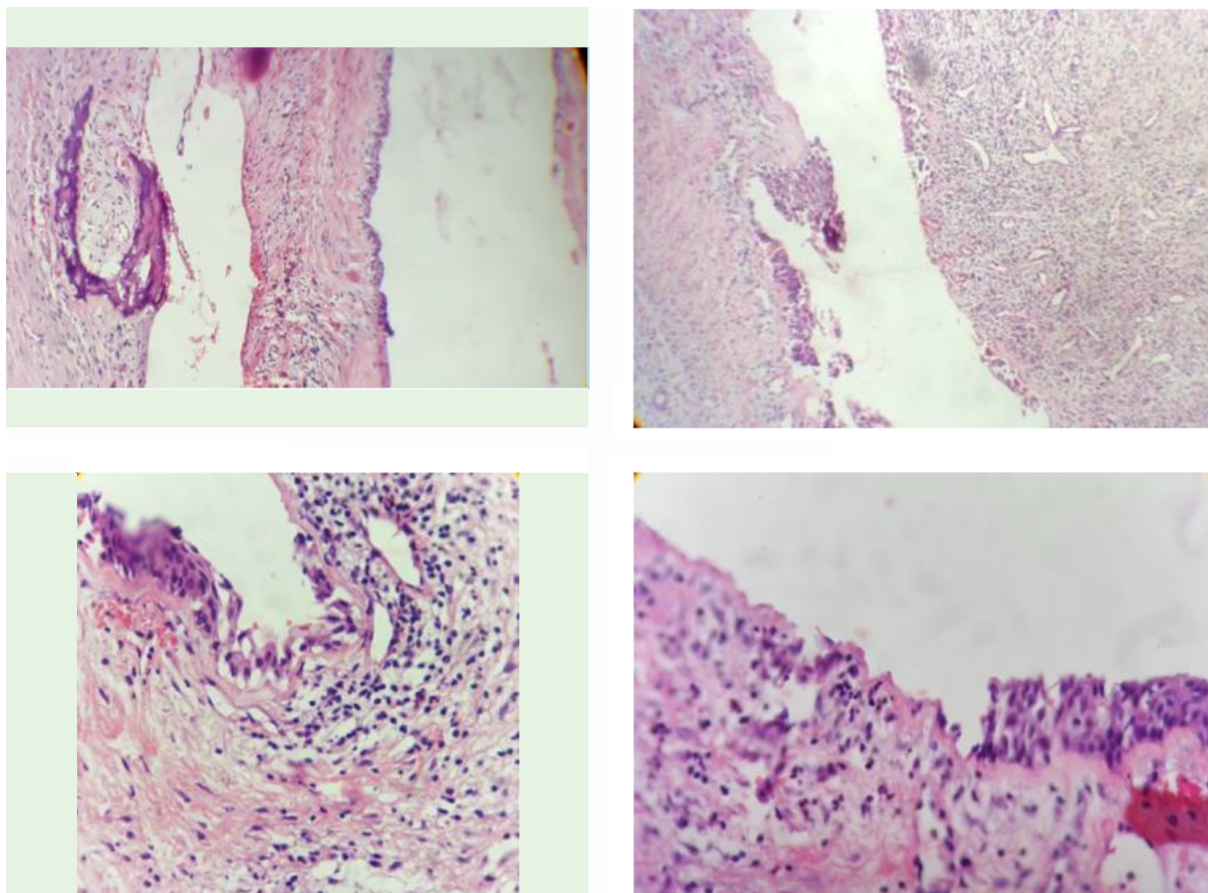
The treatment of GOC varies from conservative approaches (like enucleation, marsupialization, curettage with or without peripheral ostectomy, curettage with adjuvant Carnoy's solution, or cryotherapy) to marginal resection and segmental resection. Because of the cyst's recurrence tendency after conservative treatment, a few authors preferred marginal and segmental resection (7).

Mascitti and his colleagues in 2014 reported a 19.8% recurrence rate of glandular odontogenic cyst, but this was probably an underestimation, because about 50% of cases had a very short follow-up, up to 2 years, while the average time for recurrence was 3 years (6). Fowler and colleagues reported a recurrence rate of 50% and stated that in a sufficient length of follow up, multiple recurrences would be common (9). It was suggested that a longer follow up period of 8 years for patients is important (6). In our study, the cyst was treated with enucleation and curettage and would be under long term follow up.

#### 3.1. Conclusions

Glandular odontogenic cyst (GOC) is an uncommon cyst with a high recurrence rate and is relatively aggressive. Clinical and radiological evaluations are needed together with careful histopathologic evaluations. Oral medicine



**Figure 5.** Histopathologic Views of the Lesion

It demonstrates non-keratinized squamous epithelial lining in most areas and ciliated cylindrical and cubic lining in other areas. Goblet cells are seen in some areas. Epithelium-connective tissue interface is flat and chronic inflammatory infiltration is observed.

specialists should be aware of its characteristic features and make an effort toward an accurate diagnosis that can be followed by appropriate management.

### Acknowledgments

We would like to thank the patient who kindly participated in the study.

### Footnote

**Authors' Contribution:** Study concept and design: Hamidreza Abdolsamadi, Parastoo Sadr, and Yasamin Barakian; drafting of the manuscript: Parastoo Sadr, and Yasamin Barakian; critical revision of the manuscript for important intellectual content: Hamidreza Abdolsamadi, Parastoo Sadr, and Yasamin Barakian; study su-

pervision: Hamidreza Abdolsamadi, Parastoo Sadr, and Yasamin Barakian.

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