

# Glandular Odontogenic Cyst of the Anterior Mandible

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## Abstract

**Context:** Glandular odontogenic cyst (GOC) is a rare cyst occurring in the middle-age people with mandibular anterior as the common site of occurrence. **Case Report:** We report a case of massive GOC in a 65-year-old female with an emphasis on its clinical course, histological features, and treatment modalities. **Conclusion:** The aggressiveness and recurrences of GOC warrants clinicians for the careful examination, treatment, and long-term follow-up.

**Keywords:** Aggressive, Anterior, Glandular, Mandibular, Odontogenic

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## Introduction

Glandular odontogenic cyst (GOC) is an uncommon developmental cyst.<sup>[1]</sup> Padayachee and Van Wykin 1987,<sup>[2]</sup> reported the first two cases of this cyst as a "sialo-odontogenic cyst". Later Gardner *et al.*,<sup>[3]</sup> in 1988 established this cyst as a distinct entity under the term "glandular odontogenic cyst". The GOC is included in the World Health Organization (WHO) histology typing of odontogenic tumors under the terms "glandular odontogenic cyst or sialo-odontogenic cyst". The term "polymorphous odontogenic cyst" was introduced by High *et al.*, in 1996 because of its varied histological appearances.<sup>[4]</sup>

GOCs are rare, with just over 111 cases reported in the English language literature.<sup>[5]</sup> Between 1977 and 1995, Magnusson *et al.*, analyzed 5,800 biopsies of jaw cysts and observed that only seven cases fulfilled the criteria for GOC, which comprised 0.012% of the total.<sup>[6,7]</sup> Later,

Shen *et al.*, (2006) found a similar frequency of 12 cases in a series of 7,023 jaw cysts (0.17%) over a 37-year period and Jones *et al.*, (2006) listed 11 cases (0.2%) in their sample of 7,121 odontogenic cysts over a 30-year period.<sup>[8]</sup>

GOC predominantly occurs in the anterior mandible of middle-aged patients with slight male predilection. It can be asymptomatic or may cause pain, slow-growing swelling leading to tooth displacement.<sup>[9,10]</sup> Radiographic appearance shows a well-defined unilocular or multilocular radiolucency, often with scalloped margins.<sup>[11]</sup> The microscopic features of GOC, particularly the morphology of the epithelium, strongly suggest an origin from the remains of dental lamina which shows papillary projections and focal thickenings (plaques) within it along with mucous cells, interepithelial gland-like structures, and lack of inflammation in the connective tissue.<sup>[2,3,11]</sup> Treatment of GOC is still controversial and varies from curettage, enucleation to wide surgical resection; but due to the tendency to recur after curettage or enucleation, most of the authors prefer marginal resection.<sup>[12]</sup>

In the present study, a case of GOC is reported, emphasizing the origin, epidemiology, clinical, radiographic, and histopathological aspects along with a note on its treatment, potential for recurrence, and the importance of long-term follow-up.

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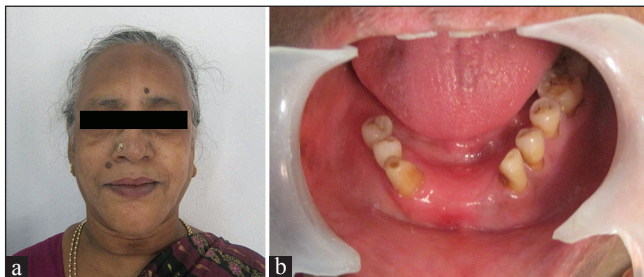
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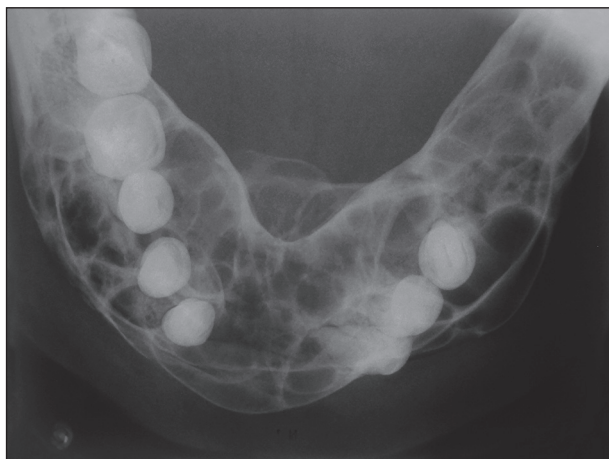
## Case Presentation

A 65-year-old female reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of pain due to decayed tooth in right lower back region since 2 months. The intraoral examination revealed partial edentulous lower jaw with 33, 34, 35, 36, 37, 43, 44, and 45 teeth present. Careful examination revealed swelling in the anterior mandible, which was not obvious for the patient to take treatment. The swelling was present from past 9 years and was asymptomatic with slow growth. There was no associated history of trauma. The anterior teeth 31, 32, 41, and 42 were extracted 4 years back by the dental surgeon due to loosening [Figure 1a and b]. Patient was not advised for the intraoral radiographs before extraction. Patient was wearing removable partial denture in the lower anterior mandible, which impinged the underlying alveolar mucosa and appeared erythematous.

The medical history of the patient revealed her to be hypertensive and diabetic from past 2 years and is under medication for the same. The swelling appeared hard without any change in the color of overlying mucosa except in denture region. The extraoral examination showed mild swelling of the lower anterior mandible. The submandibular glands were



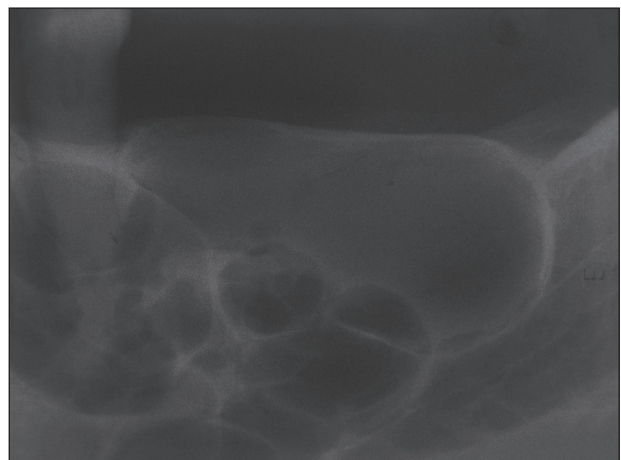
**Figure 1:** (a) Frontal profile of the patient. (b) Intraoral photograph



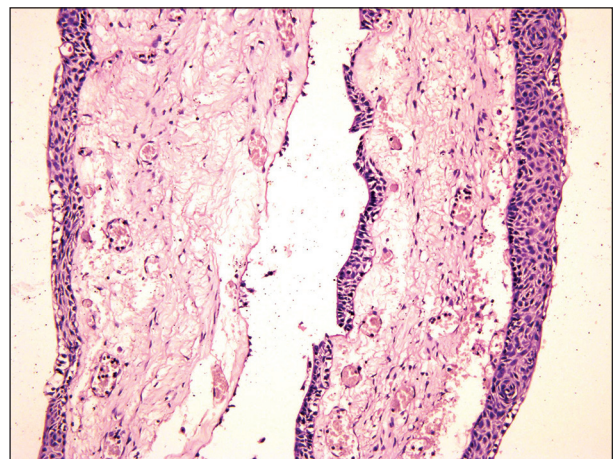
**Figure 3:** Occlusal radiograph showing multilocular lesion involving the body of the mandible

palpable, nontender but hard, and appeared fixed to the underlying structures.

Intraoral periapical radiograph and panoramic radiographs were advised, which revealed well-defined lytic multilocular radiolucency involving the whole of the anterior mandible with its extension in the anteroposterior direction involving right and left body of the mandible [Figures 2 and 3]. Based on the clinical and radiographic finding, a differential diagnosis of odontogenic keratocyst and ameloblastoma were made. Fine-needle aspiration cytology (FNAC) was advised, which revealed only blood cells. Incisional biopsy was carried out, which on gross examination appeared as blood clots; but on histopathology, it showed ciliated cells with intraepithelial microcyst formation and mucous cells with periodic acid-Schiff (PAS) positive material. The connective tissue was filled with blood cells and few inflammatory cells [Figure 4]. The diagnosis of GOC was made.



**Figure 2:** Intraoral periapical radiograph showing honey-combed appearance



**Figure 4:** Hematoxylin and eosin (H and E) stained section showing ciliated cells with intraepithelial microcyst and mucous cells



The patient was explained about the lesion, the surgical treatment plan, and informed consent was taken. Segmental resection of the mandible was advised, which was carried out under general anesthesia. A bilateral extended submandibular incision was made and the mandible was exposed [Figure 5]. The osteotomy cuts were performed from right antegonial to left antegonial region including the surgical margin of 1cm. The submandibular lymph nodes were also removed. The angle of the mandible was spared along with the ramus and condyle. Reconstruction plate was adapted along with the curvature of mandible across the midline and fixed to the remaining mandible [Figure 6]. Closure of the surgical site was done in layers. Postoperative healing was good with no complications except for the paresthesia. The patient is kept under long-term follow-up and no recurrence was observed in 2 years of duration.

The excisional biopsy of the mandible was sent for the confirmatory histopathological diagnosis and margin clearance. The final diagnosis was in correlation with the incisional diagnosis and the margins were also free of the lesion.

## Discussion

GOC is a very rare pathology and has so far been documented just over 111 cases. The clinical aspects of the present case are consistent with those reported for GOC in the literature.<sup>[5,6]</sup> There is a slight predilection for men and the condition occurs mostly in middle-aged patients, but the present case reported is in female patient of elderly age. The anterior mandible is the most common site of occurrence; however in the current case report, the lesion extended to the right and left molars. The pain reported by the patient was probably due to the inflammation because of impingement and pressure by the removable partial denture in anterior region.

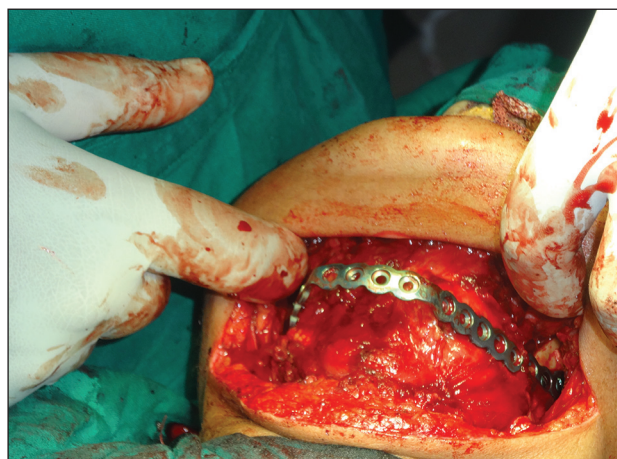
GOC does not display specific or pathognomonic radiographical features. It may present as a multilocular or unilocular radiolucency with well-defined borders. The recognition of this cyst on the base of clinical and radiological examination is practically impossible. Only the histopathological examination allow for certain diagnosis of the cyst.<sup>[5]</sup> GOC is an intraosseous developmental cyst which may occur in the apical region of the tooth and may show interdental extension. On the basis of clinical and radiographic examination, the diagnosis of the dentigerous cyst, odontogenic keratocyst, radicular cyst, myxoma, ameloblastoma, central giant cell lesion, fibrous dysplasia, and central mucoepidermoid carcinoma (MEC) can be made.

The aspirated fluid in the present case showed compatibility with the peripheral blood, perhaps due to inflammation. Literature suggests the low viscous, colorless fluid content may indicate clinically GOC.<sup>[6]</sup> The characteristic feature of GOC histopathologically strongly suggests its origin from the dental lamina. The study reported by Koppang and Matthews and a case report by Ide *et al.*, suggested the odontogenic nature of GOC.<sup>[13,14]</sup>

The microscopic features of the GOC shares its similarity with the lateral periodontal cyst (LPC), botroid odontogenic cyst (BOC), and the central MEC. The plaque-like epithelial thickening in LPC made authors to believe that the GOC could be the clinical microscopic variant of LPC.<sup>[15]</sup> But the low aggressive nature, limited growth potential and low recurrence nature of LPC has annulled GOC. The multilocularity, multicystic, and GOC was considered to be the variant of BOC. Some authors contradict this statement due to the presence of mucous and ciliated epithelial cells and mucous pooled cystic spaces in GOC and not in BOC, thereby considering them as different entities.<sup>[16]</sup> It is mandatory to differentiate the GOC from the central



**Figure 5:** Submandibular incision



**Figure 6:** Reconstruction plate

variant (low grade cystic type) of MEC as the later does not always support its intrabony salivary gland origin, but can originate from pluripotent cells of oral epithelium or due to metaplastic change in the odontogenic epithelium. Magnusson *et al.*,<sup>[6]</sup> considered GOC the most benign end of the spectrum of central MEC. Many authors follow the major and minor criteria suggested by Kaplan and Salehinejad *et al.*, for the diagnosis of GOC [Table 1]. The distinction between GOC and MEC were established based on immunohistochemical studies using cytokeratin (Ck). Ck 7, 13, 14, and 19 positivity and negative reaction for epithelial membrane antigen (EMA) supported odontogenic origin. Some authors suggested expression of Ck 8, 18, and 19 might be useful tool in differentiating these lesions.<sup>[6,16]</sup>

The unpredictable, aggressive behavior and high recurrence rate of GOC has led different treatment modalities such as conservative approach including enucleation, marsupialization, curettage with or without peripheral ostectomy, curettage with adjuvant Carnoy's solution, or cryotherapy, to marginal resection and segmental resection. Small unilocular lesions can be treated by enucleation whereas the large lesions should include enucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw resection in multilocular cases. Marsupialization followed by second phase surgery is an option for lesions approaching vital structures.<sup>[5,17,18]</sup> Kaplan *et al.*, also demonstrated that the rate of recurrence also increases with the radiographic complexity of the cyst. Hussain *et al.*, found a high recurrence rate of 55% and a long recurrence interval, with an average of 4.9 years; whereas, Koppang *et al.*, observed a recurrence rate of 21%.<sup>[19-21]</sup> Considering the age, recurrence rate and pooreconomic status microvascular reconstruction was ruled out. Long-term follow-up is recommended ranging with a minimum of 3-5 years after treatment and the prognosis of GOC remains good.<sup>[6,13]</sup>

**Table 1: Major and minor criteria for diagnosing GOC**

Major criteria	Minor criteria
Variations in thickness of the lining with or without epithelial "spheres" or "whorl", no palisades	Papillary projections
Squamous epithelial lining with flat interface	Ciliated cells
Cuboidal eosinophilic cells or "hob-nail" cells	Multicystic or multiluminal architecture
Mucous "goblet" cells with interepithelial mucous pools with or without crypts lined by mucous-producing cells	Clear or vacuolated cells in basal or spinous layer
Interepithelial glandular microcystic or duct-like structures	

The author concludes by stressing on the rarity of the GOC, with only 65 cases being reported in the English language literature so far. Careful clinical and radiological examination along with the confirmatory histopathological study is required to differentiate GOC from the differential diagnosis of central mucoepidermoid carcinoma.

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