



## CASE REPORT

# Glandular Odontogenic Cyst Mimicker: A Novel Entity

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## ABSTRACT

Glandular odontogenic cyst (GOC) is an uncommon developmental odontogenic cyst of jaws with a frequency of approximately 0.2%. Because of its aggressive biological behavior including its malignant transformation potential, recently collaborative efforts by few researchers have laid down certain histopathological criteria segregating it from its mimickers which include dentigerous cyst, lateral periodontal cyst (LPC), or botryoid cyst, radicular cyst, and central low-grade mucoepidermoid carcinoma. Therefore, cautious histopathological evaluation is necessary of GOC mimickers in order to prevent its overdiagnosis. Here, we present a case of GOC mimicker in a 12-year-old male patient in left maxillary region.

**Keywords:** Glandular odontogenic cyst mimicker, Hobnail cell, Sialo-odontogenic cyst.

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## INTRODUCTION

Glandular odontogenic cyst (GOC) was first documented as “sialo-odontogenic cyst” by Padayachee and Van Wyk in 1987.<sup>1</sup> However, in 1988, Gardner et al<sup>2</sup> proposed it as a distinct entity and gave a term “GOC” whereas High et al<sup>3</sup> proposed the term “polymorphous odontogenic cyst” which is characterized by a potentially aggressive behavior with a high recurrence rate.<sup>4</sup> Glandular odontogenic cyst is a rare lesion comprising only 0.2% of all odontogenic cysts. The cyst is most frequently encountered in the age range of 14 to 75 years with male predilection (1.3:1), with predilection for mandible (70%) affecting both anterior and posterior areas. Radiographically, it is radiolucent, well-defined, either unilocular (53.8%) or multilocular (46.2%) with frequent perforation (61%) and thinning of cortical plates (24.4%). Furthermore, English literature revealed recurrence rate which varies from

21 to 55%<sup>5,6</sup> which indicates its aggressiveness.<sup>7</sup> Therefore, a confirmatory diagnosis is critical for treatment and follow-up. However, similar histopathological features are shared between GOC and other odontogenic cysts, such as botryoid odontogenic cyst, dentigerous cyst with metaplasia, radicular cyst, or central mucoepidermoid carcinoma (CMEC), thereby making the diagnosis more cumbersome.<sup>7</sup> However, even though the microscopic features of GOC have been described in detail, there is no consensus on its diagnostic features. It is sometimes difficult to discern whether a particular cyst having some but not all of the described features of GOC represents a true GOC or another cyst with GOC-like features imposing a need to introduce a new group “glandular odontogenic cyst mimickers” (GOC mimicker).

The most recent World Health Organization classification includes a definition of the GOC – “A cyst arising in the tooth bearing areas of the jaws and characterized by an epithelial lining with cuboidal or columnar cells, both at the surface and lining, with crypts or cyst-like spaces within the thickness of the epithelium.”<sup>8</sup> Few authors have selected parameters based on previously reported microscopic features for GOC from the literature.<sup>9,10</sup> Kaplan et al<sup>7</sup> proposed major and minor microscopic criteria for GOC based on the frequency of each feature in reported cases from the literature. Fowler has emphasized on certain criteria which are mandatory in differentiating a true GOC from GOC mimicker. The purpose of this paper is to present a case report depicting the clinical, radiographic, and histopathologic features of a GOC mimicker with particular emphasis on microscopic parameters necessary for diagnosis.

## CASE REPORT

A 12-year-old male patient reported in the Department of Oral Pathology, MGM Dental College and Hospital, Navi Mumbai with a chief complaint of painful swelling in upper left anterior region of the jaw since 3 months. Extraorally, a diffuse swelling was evident which resulted in facial asymmetry. Intraoral examination revealed a solitary diffuse buccal swelling in the left maxillary region extending from 21 to 25, approximately 3×2×1 cm in size (Figs 1A and B). The swelling extended superoinferiorly from attached gingiva obliterating the buccal vestibule. Displacement of 21 was also evident. Overlying mucosa was smooth and erythematous. On palpation, the swelling was firm, tender, nonmobile, and nonpulsatile. Radiographic examination

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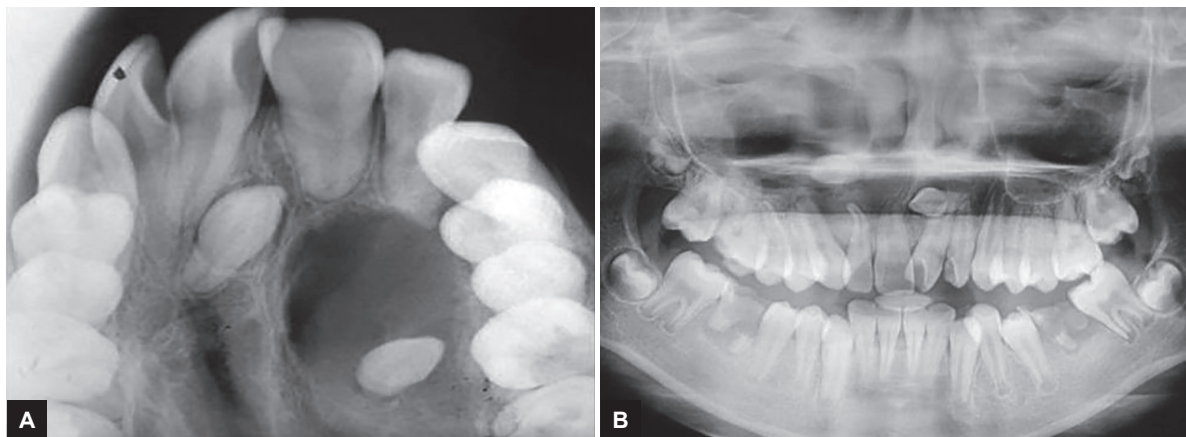


**Figs 1A and B:** Extraoral view: (A and B) shows diffuse swelling extending over left maxillary region

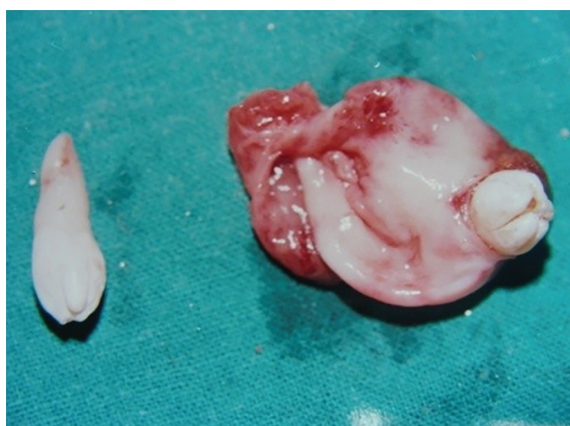
revealed a unilocular radiolucent area with well-defined sclerotic border extending from 21 to 25 region along with two impacted supernumerary teeth (Figs 2A and B). A provisional diagnosis of dentigerous cyst was given.

Incisional biopsy of the lesional tissue microscopically revealed nonkeratinized stratified squamous epithelial lining of 3 and 4 layers thick with features of pseudo-microcyst formation, superficial cuboidal eosinophilic (hobnail) cells. The connective capsule showed moderate

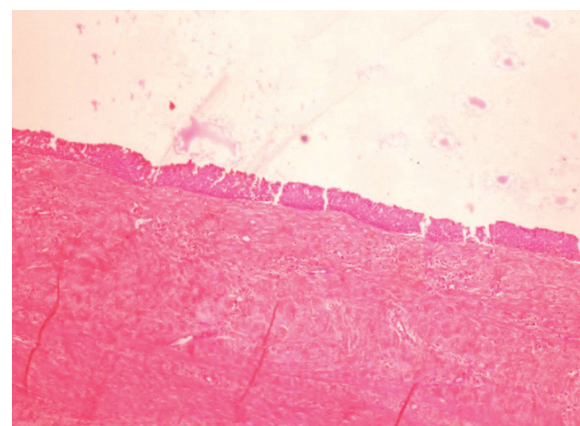
chronic inflammatory cells infiltrate, hemorrhage, and vascularity. A diagnosis of infected dentigerous cyst was made correlating clinical, radiological, and histopathological features. Based on the histopathological report, surgical enucleation was done along with extraction of supernumerary teeth. Subsequently, root canal treatment of teeth 11 and 21 were done. The gross excised soft tissue specimen received was greyish white in color measuring about  $4.0 \times 2.5 \times 0.3$  cm attached to one of the crown of the supernumerary teeth. The lesion resembled cystic lining with a smooth surface contour (Fig. 3). Microscopically, it is a nonkeratinized stratified squamous epithelial lining of 3 and 4 cells thickness and had a flat interface with the underlying connective tissue, lacking basal palisading. Some of the superficial cells showed an irregular surface and a hobnail appearance with eosinophilia and clear cells. The underlying connective tissue capsule showed moderate cellularity, with focal areas of chronic inflammatory cells and hemorrhage (Figs 4 and 5). The histopathological features confirmed the diagnosis of GOC mimicker. Postoperatively uneventful healing was noticed (Fig. 6).



**Figs 2A and B:** Occlusal view: (A) and orthopantomogram; and (B) reveals a well-defined unilocular radiolucency extending from 21 to 25 associated with the crown of one impacted supernumerary tooth

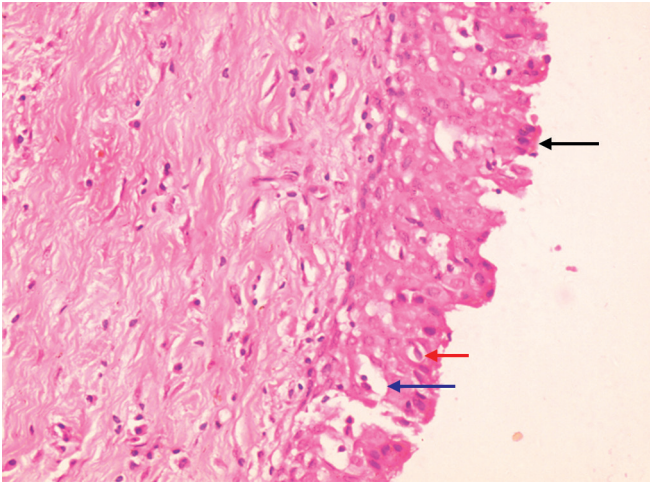


**Fig. 3:** Gross specimen shows soft tissue cystic lining with extracted impacted supernumerary tooth

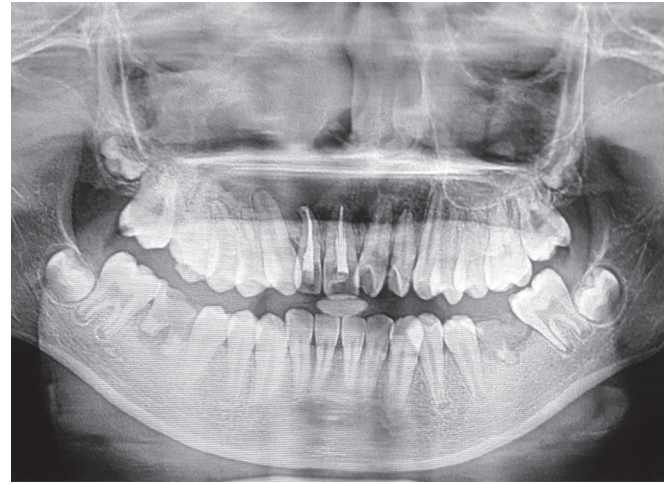


**Fig. 4:** Nonkeratinized stratified squamous epithelial lining of 3 and 4 cells thickness lacking basal palisading, hobnail, and clear cells (H&E, 10 $\times$ )





**Fig. 5:** Hobnail cells (black arrow), pseudomicrocysts (purple arrow), and clear cells (red arrow) (H&E 40×)



**Fig. 6:** Postoperative follow-up orthopantomogram shows postoperative follow-up with healing in the involved site (21–25) and with root canal treatment of 11 and 21

## DISCUSSION

Histopathological simulation with various odontogenic cysts makes the differential diagnosis of GOC taxing. Apart from their distinct clinicopathologic features, some overlapping histopathologic findings like nonkeratinized epithelial lining, focal thickenings, and glycogen-rich epithelial cells could pose problem in differential diagnosis of lateral periodontal cyst (LPC)/botryoid odontogenic cyst (BOC).<sup>11</sup> In radicular cyst, the metaplastic changes (ciliated cells and mucous cells) in epithelial lining share similarity with GOC. The clinical feature of dentigerous cyst being associated with an unerupted tooth could be shared by GOC. Histopathologically, when dentigerous cyst exhibits metaplastic changes like eosinophilic cuboidal cells (hobnail cell), mucous cells, and ciliated cells, the simulation makes the diagnosis challenging.<sup>12,13</sup> Additionally, GOCs showing the features of multicompartmentalization and presence of numerous small mucous cells islands in the cyst wall may complicate the

diagnosis as CMEC. All these lesions are clustered under the category of GOC mimickers.

Laying down of certain diagnostic features which are necessary for evaluating GOC and its mimickers needs to be considered. In this regards, continuous conscious efforts by researchers are put forward based on and adapted from previously reported microscopic features for GOC from the literature.<sup>9,10</sup> Kaplan et al<sup>4,7</sup> proposed a list of major and minor microscopic criteria for GOC based on the frequency of each feature in reported cases from the literature.

Later, Fowler et al<sup>14</sup> did modification in the earlier proposed criteria and enlisted common histological parameters—microcysts, epithelial spheres, clear cells, and variable thickness of the cyst lining which appear to be helpful in distinguishing GOCs from GOC mimickers. They also concluded that the presence of seven or more microscopic parameters was highly predictive of a diagnosis of GOC. Similarly, the presence of five or less microscopic parameters was highly predictive of a non-GOC or GOC mimicker<sup>14</sup> (Table 1).

**Table 1:** Histopathological criteria of GOC and its comparison with our present case

| Sl. no. | Microscopic parameters   | Kaplan's criteria   |       | Present case |
|---------|--|---|-------|--------------|
|         |  | Major   | Minor |              |
| 1       | Surface eosinophilic cuboidal cells, also called "hobnail cells"                                     | √   |       | √            |
| 2       | Intraepithelial microcysts or duct-like spaces lined by a single layer of cuboidal to columnar cells | √   |       |              |
| 3       | Apocrine snouting of hobnail cells   |   |       |              |
| 4       | Clear or vacuolated cells  |   | √     | √            |
| 5       | Variable thickness of the cyst lining  | √ Additionally, Squamous epithelial lining, with a flat interface with the connective tissue wall, lacking basal palisading |       | √            |
| 6       | Papillary projections or "tufting" into the cyst lumen   |   | √     |              |
| 7       | Mucous goblet cells  | √   |       | √            |
| 8       | Epithelial spheres or plaque-like thickenings  | √   |       |              |
| 9       | Cilia  |   | √     |              |
| 10      | Multiple compartments  |   | √     |              |

In our present case, clinical, radiological, and incisional biopsy report lead to the diagnosis of dentigerous cyst with metaplastic changes which include hobnail cells with eosinophilia, squamous epithelial lining with a flat interface with the connective tissue wall, lacking basal palisading and clear cells. These features are analogous with the mentioned criteria.<sup>4,7,14</sup> There was presence of pseudomicrocysts (microcyst lined by flattened cells) in contrast to the true microcysts (cuboidal to columnar cells lining) which are seen in GOC lining, thereby validating our histopathological diagnosis of GOC mimicker.

Glandular odontogenic cyst mimickers are an intermediate between conventional lesions and GOC, so persistent efforts should be carried out for their seclusion, so that their exact biological behavior could be traced out.

## CONCLUSION

This paper aims to depict a clear picture toward histopathological diagnosis so as to differentiate GOC mimickers from GOC to prevent overdiagnosis, as GOC has a higher recurrence rate. Further studies need to be undertaken to have a cumulative data to explore the exact biological behavior of this budding entity.

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