

## Case Report

## Glandular odontogenic cyst: A diagnostic challenge

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Glandular Odontogenic Cyst (GOC) is an uncommon developmental cyst of the jaw with distinct histopathological features, but significant diagnostic overlap exists with both benign and aggressive odontogenic lesions. Its resemblance to neoplasms such as central mucoepidermoid carcinoma (CMEC) complicates diagnosis, demanding meticulous evaluation. This article presents a rare case of GOC in a 17-year-old female, emphasizing its clinical, radiological, and microscopic hallmarks and highlighting the critical role of comprehensive histopathological and ancillary diagnostic techniques for precise identification and management.

**Keywords:** Glandular odontogenic cyst, Developmental jaw cyst, Hobnail cells, Mucoepidermoid carcinoma mimicker, Odontogenic lesions, Histopathological diagnosis.

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## 1. Introduction

Glandular odontogenic cyst (GOC) was first described by Padayachee and Van Wyk in 1987 and later termed by Gardner in 1988 as a distinct developmental odontogenic cyst with glandular features. Although it accounts for less than 0.5% of odontogenic cysts, its aggressive potential, recurrence risk, and diagnostic challenges necessitate careful evaluation. The latest World Health Organization (WHO) classification (2023) defines GOC as "a developmental cyst wherein the epithelial lining demonstrates glandular differentiation".<sup>1,2</sup>

## 2. Case Report

A 17-year-old female presented with pain and swelling in the right posterior maxillary region for two months. The patient reported undergoing root canal treatment (RCT) for tooth 16, followed by persistent pus discharge from the buccal vestibule.

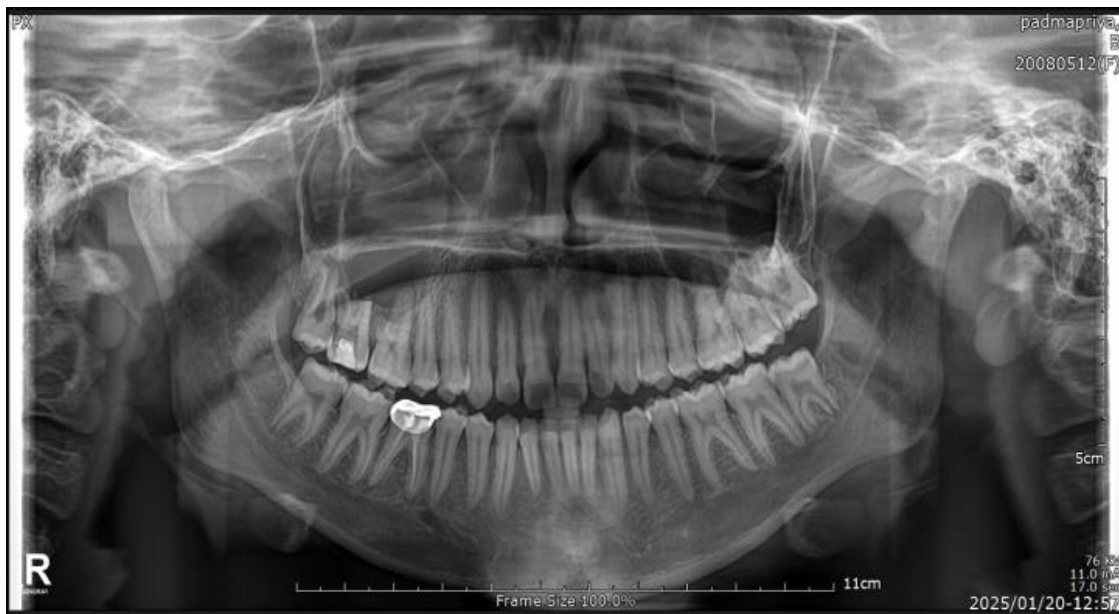
Extraoral clinical examination revealed mild facial asymmetry without palpable lymphadenopathy. Intraorally, obliteration of the buccal vestibule near the upper right molars with pus discharge was observed.

Radiological evaluation with an intraoral periapical radiograph and OPG revealed a poorly defined radiolucency involving the periapical regions of teeth 16 and 17, extending toward tooth 18, with evident root resorption of teeth 16 and 17 (**Figure 1**).

## 2.1. Procedure

Enucleation and curettage under local anesthesia were performed, and the specimen was sent for histopathological examination.

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**Figure 1:** Intraoral periapical radiograph demonstrating ill-defined radiolucency with root resorption of 16 and 17, extending toward 18



**Figure 2:** Gross examination revealed multiple irregular, soft, greyish-white tissue fragments: **a)** Before; and **b)** After grossing

## 2.2. Provisional diagnosis

A diagnosis of an odontogenic cyst was considered with differentials including radicular cyst, odontogenic keratocyst (OKC), and unicystic ameloblastoma.<sup>3,4</sup>

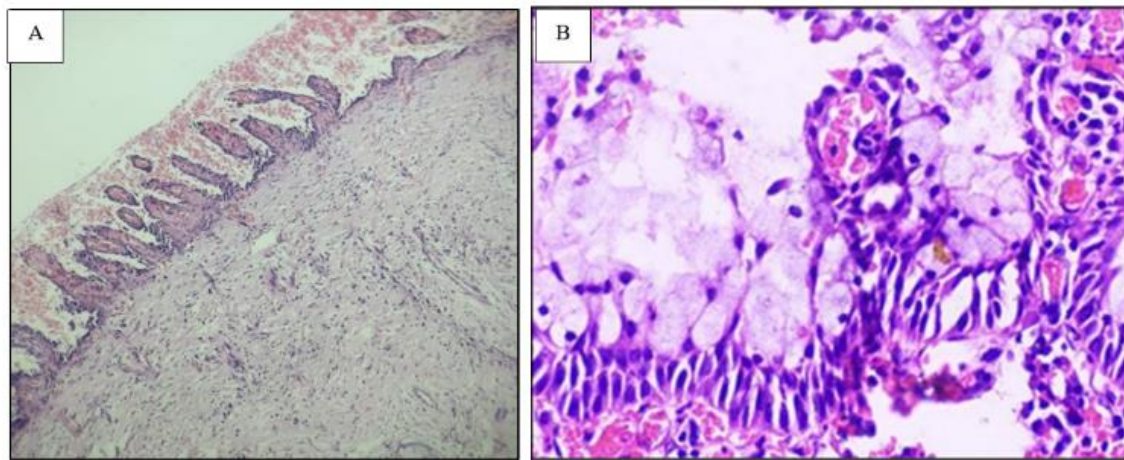
Gross examination revealed multiple irregular, soft, greyish-white tissue fragments (**Figure 2**). Histopathological findings and microscopic examination demonstrated non-keratinised stratified squamous epithelium with papillary projections, numerous mucous (goblet) cells within the epithelial lining, cholesterol clefts and hemorrhagic areas with mixed chronic inflammatory cell infiltrate (**Figure 3**). Characteristic hobnail cells with apical nuclear protrusions, palisaded basal cell layer were observed. These findings confirmed the diagnosis of GOC.

## 3. Discussion

GOC most commonly occurs in the anterior mandible; however, maxillary cases, especially near the

globulomaxillary region, have been reported.<sup>5,6</sup> While the lesion predominantly affects middle-aged adults, occurrence in adolescents, such as in our case, though rare, has been documented.<sup>2,5</sup>

The lesion originates from remnants of the dental lamina with gland-like differentiation. The absence of adjacent salivary gland tissue in such cases supports its odontogenic origin.<sup>1</sup> Histopathological features of GOC can overlap with those of lateral periodontal cyst, botryoid cyst, and CMEC, making diagnosis challenging.<sup>7</sup> Key distinguishing features include abundant mucous (goblet) cells, Hobnail cells with apical nuclear protrusion, and intraepithelial microcyst formation. Immunohistochemistry and Molecular Diagnostics with Mucicarmine, Alcian Blue, and PAS-Diastase highlight mucous elements in the epithelium.<sup>6,7</sup> Ki-67 Proliferative Index is Lower in GOC, aiding differentiation from CMEC.<sup>6</sup>



**Figure 3: A):** Low-power photomicrograph showing papillary projections, mucous cells, and hobnail cells in the cystic epithelium (H&E stain, 10x); **B):** High-power view highlighting hobnail cells with apical nuclear protrusions (H&E stain, 40x)

The most critical distinction is between GOC and low-grade MEC, as both can exhibit cystic structures lined by epithelium of varying thickness, predominantly composed of mucous cells, with some epidermoid and intermediate cells (Kaplan et al.,). However, a key differentiating feature is that in MEC, the epidermoid cells typically demonstrate exophytic proliferation toward the periphery of the cystic spaces, whereas in GOC, epithelial plaques or whorled thickenings protruding into the lumen are more characteristic (Kaplan et al., 2008). Additionally, the presence of hobnail-shaped eosinophilic cuboidal cells along the surface of the cystic lining and the occurrence of intraepithelial microcysts further support a diagnosis of GOC. Notably, the detection of MAML2 gene rearrangements has emerged as a significant molecular diagnostic tool to help differentiate GOC from MEC.<sup>9</sup>

Conservative surgical techniques such as enucleation and curettage are associated with recurrence rates of 21.6% to 50%.<sup>4,5</sup> Larger or recurrent lesions may necessitate marsupialisation followed by enucleation or peripheral osteotomy to minimise recurrence risk.<sup>5</sup> Long-term follow-up, extending beyond seven years, is recommended for recurrence detection.<sup>1,4,5</sup>

The recurrence potential of Glandular Odontogenic Cyst (GOC) is influenced by its clinical features, biological behavior, and the treatment approach used. Recurrences are more commonly seen in multilocular lesions, those associated with cortical bone thinning or perforation, and in larger cysts—particularly those occupying a bone area greater than the space of two adjacent teeth.<sup>9</sup>

#### Factors associated with higher recurrence

1. Multilocular radiolucency: More commonly associated with recurrence compared to unilocular lesions.
2. Large lesion size—lesions occupying an area larger than two adjacent teeth are more prone to recur.

3. Cortical bone involvement: Cases with thinning or perforation of cortical bone show higher recurrence risk.
4. Inadequate surgical treatment: Simple enucleation or curettage often leads to recurrence. Aggressive management (e.g., en bloc resection) is recommended for large or multilocular cases.
5. Histological complexity: The cyst lining is fragile and variable, making complete removal difficult. Presence of epithelial plaques, mucous cells, hobnail cells, and microcysts may be linked to aggressive behaviour.<sup>10</sup>

#### 4. Conclusion

Glandular Odontogenic Cyst, though rare, presents significant diagnostic and therapeutic challenges due to its overlap with other odontogenic and glandular lesions. A definitive diagnosis requires correlation of clinical, radiological, histopathological, and when necessary, immunohistochemical and molecular data. Early recognition, comprehensive surgical management, and vigilant long-term follow-up are essential to reduce morbidity and recurrence rates.

#### 5. Source of Funding

None.

#### 6. Conflict of Interest

None.

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