

GLANDULAR ODONTOGENIC CYST OF THE MAXILLA: A UNIQUE PRESENTATION OF AN UNCOMMON ENTITY

Panda Abikshyeet, Annaluru Sri Sasank Tejaswee, Mondal Chitrita, Mahapatra Niva, Thangadurai Maheswaran, Harikrishnan Prasad

ABSTRACT

Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst of the jaws that usually presents as a slow-growing asymptomatic swelling, but it is locally aggressive and shows a tendency for recurrence. Clinico-radiographically, GOC can mimic numerous odontogenic cysts and tumors, making preoperative diagnosis challenging. Radiographically, it commonly appears as a well-defined unilocular or multilocular radiolucency with scalloped margins. Histopathology is characteristic but may still be confusing because the lining shows glandular features with mucin-producing goblet cells, which may suggest salivary gland origin or lead to misdiagnosis as mucoepidermoid carcinoma. However, absence of salivary gland markers and the presence of odontogenic epithelial features support its odontogenic lineage. Presentation in the maxilla is distinctly uncommon. We report a case of GOC with a rare maxillary presentation in a 34-year-old female patient, highlighting the clinical, radiographic, and histopathological diagnostic challenges and the importance of accurate diagnosis for comprehensive treatment planning and reduction of recurrence risk.

Keywords

Glandular odontogenic cyst, odontogenic cysts, maxilla, mucoepidermoid carcinoma, case report

INTRODUCTION

Glandular odontogenic cyst (GOC) is an uncommon but biologically intriguing odontogenic cyst arising from the remnants of dental lamina and showing glandular differentiation with potential for locally aggressive behavior.[1] It was first described in 1987 as a sialo-odontogenic cyst and later redefined by Gardner et al. in 1988 as glandular odontogenic cyst because of its distinctive glandular-like histopathological architecture.[2] In 1992, the World Health

1. Panda Abikshyeet, M.D.S, Professor, Oral and Maxillofacial Pathology and Microbiology, Kalinga Institute of Dental Sciences (KIDS), Kalinga Institute of Industrial Technology (KIIT) Deemed to be University, Bhubaneswar-751024, Odisha, India.
2. Annaluru Sri Sasank Tejaswee, M.D.S, Lecturer, Oral and Maxillofacial Surgery, Kalinga Institute of Dental Sciences (KIDS), Kalinga Institute of Industrial Technology (KIIT) Deemed to be University, Bhubaneswar, Odisha, India.
3. Mondal Chitrita, M.D.S, Lecturer, Oral and Maxillofacial Surgery, Kalinga Institute of Dental Sciences (KIDS), Kalinga Institute of Industrial Technology (KIIT) Deemed to be University, Bhubaneswar, Odisha, India.
4. Mahapatra Niva, M.D.S, Lecturer, Oral and Maxillofacial Pathology and Microbiology, Kalinga Institute of Dental Sciences (KIDS), Kalinga Institute of Industrial Technology (KIIT) Deemed to be University, Bhubaneswar, Odisha, India.
5. Thangadurai Maheswaran, Professor, Adhiparasakthi Dental College and Hospital, Melmaruvathur, Tamil Nadu, India.
6. Harikrishnan Prasad, Professor, Department of Oral and Maxillofacial Pathology and Oral Microbiology, KSR Institute of Dental Science and Research, Tiruchengode, Tamil Nadu, India.

Correspondence:

Thangadurai Maheswaran, Professor, Adhiparasakthi Dental College and Hospital, Melmaruvathur, Tamil Nadu, India.
Email: maheswaranmnds@gmail.com

Organization recognized GOC as a separate developmental odontogenic cyst and emphasized its characteristic epithelial lining with cuboidal or columnar cells, microcysts, and gland-like structures.[3] The 2022 WHO classification further highlighted hobnail cells as one of its most characteristic features.[4]

Despite this recognition, GOC remains one of the most diagnostically challenging odontogenic cysts because of marked radiological overlap with lateral periodontal cyst, botryoid odontogenic cyst, odontogenic keratocyst, and low-grade central mucoepidermoid carcinoma. Consequently, imaging alone is usually insufficient and histopathological evaluation remains the gold standard for definitive diagnosis.[1][5]

GOC accounts for a very small proportion of odontogenic cysts, with reported prevalence around 0.17%, usually affecting middle-aged individuals and more frequently occurring in the anterior mandible. Maxillary presentation is rare, making such cases especially important in the literature.[6] Because recurrence after conservative treatment has been reported in 21%–55% of cases, timely recognition and appropriate management are essential.[7] The present case highlights a rare maxillary GOC in a 34-year-old woman and underlines the importance of clinicopathologic correlation for accurate diagnosis and long-term follow-up.

Case report

A 34-year-old female patient reported with a persistent painless swelling in the left maxillary region for three months. The lesion initially appeared as a small mass and gradually enlarged, resulting in restricted mouth opening and functional discomfort.

Intraoral examination revealed a well-defined swelling measuring approximately 3 × 2 cm in the left gingivobuccal sulcus, extending from the 22 to 25 region with complete obliteration of the buccal vestibule. The overlying mucosa was intact and pale pink. The swelling was firm, non-tender,

and non-pulsatile, and there was no evidence of purulent discharge. All associated teeth were vital.

Panoramic radiography demonstrated a well-circumscribed unilocular radiolucency in the left nasolabial region extending from 22 to 25 without maxillary sinus involvement, with inferior extension between the roots of the canine and incisors. Fine-needle aspiration yielded thick grayish-white cheesy material. Based on the clinical, radiological, and aspirational findings, a provisional diagnosis of odontogenic cyst, possibly odontogenic keratocyst, was considered.

Incisional biopsy showed a cystic lining composed of odontogenic epithelium with superficial cuboidal cells, focal goblet cell differentiation, and glandular duct-like structures. Some areas showed eosinophilic cuboidal surface cells consistent with hobnail cells. The underlying connective tissue stroma exhibited inflammatory cell infiltration and osseous tissue components. These features were diagnostic of glandular odontogenic cyst.

Because of the lesion's size and location, segmental resection of the maxilla with adequate surgical margins was performed under general anesthesia. Histopathological examination of the excised specimen confirmed GOC with clear margins. Periodic acid-Schiff staining further supported the diagnosis. The patient remained disease-free during two years of postoperative follow-up.

DISCUSSION

GOC is a rare intraosseous odontogenic lesion, usually affecting middle-aged patients with a slight male predilection. Although most reported cases occur in the anterior mandible, maxillary lesions are distinctly unusual and may be overlooked in the initial differential diagnosis.[6][7] In the maxilla, close proximity to the maxillary sinus, nasal cavity, and orbit may facilitate early cortical perforation and broader local spread, thereby complicating treatment planning and rehabilitation.[8]

The histogenesis of GOC remains uncertain. Earlier hypotheses favored intraosseous salivary gland tissue origin, whereas current evidence

supports an odontogenic epithelial origin. Proposed explanations include glandular differentiation of odontogenic epithelium by prosoplasia or possible overlap with low-grade central mucoepidermoid carcinoma in some lesions.[9] This diagnostic overlap is clinically important because management and prognosis differ substantially.

Clinically, GOC may be asymptomatic or may present with swelling, pain, tooth mobility, displacement, and cortical expansion. Radiographically, it usually appears as a unilocular or multilocular radiolucency with well-defined or scalloped borders, sometimes associated with root resorption or tooth displacement. Larger lesions, especially multilocular ones, may simulate odontogenic keratocyst or ameloblastoma.[1] [5] In the present case, the clinicoradiographic picture initially favored odontogenic keratocyst, illustrating the diagnostic challenge.

Histologically, GOC is characterized by a lining of variable thickness showing cuboidal to columnar cells, hobnail cells, mucous or goblet cells, intraepithelial microcysts, and gland-like structures. These features distinguish it from other odontogenic lesions but may still overlap with central mucoepidermoid carcinoma. Special stains such as PAS and mucicarmine are useful adjuncts, and molecular assessment for MAML2 rearrangement may help exclude mucoepidermoid carcinoma in difficult cases.[10][11][12][13]

Management remains controversial because of the lesion's aggressive potential and recurrence tendency. Conservative approaches have been associated with substantial recurrence, particularly for larger or multicystic lesions, whereas resection with adequate margins is often considered more

reliable in extensive disease. [14-19] In the present case, surgical resection was selected because of the lesion's extent and maxillary location, and no recurrence was observed during follow-up.

CONCLUSION

Glandular odontogenic cyst involving the maxilla is a rare and clinically important lesion because it lacks pathognomonic clinical and radiographic features and can mimic several odontogenic cysts and tumors. Accurate diagnosis depends on careful histopathological evaluation supported by special stains and clinicoradiographic correlation. Early recognition, appropriate surgical management, and long-term surveillance are essential because of the risk of local aggressiveness and recurrence. This case adds to the limited literature on maxillary GOC and supports the need to include it in the differential diagnosis of locally aggressive radiolucent lesions of the maxilla.



FIGURE 1

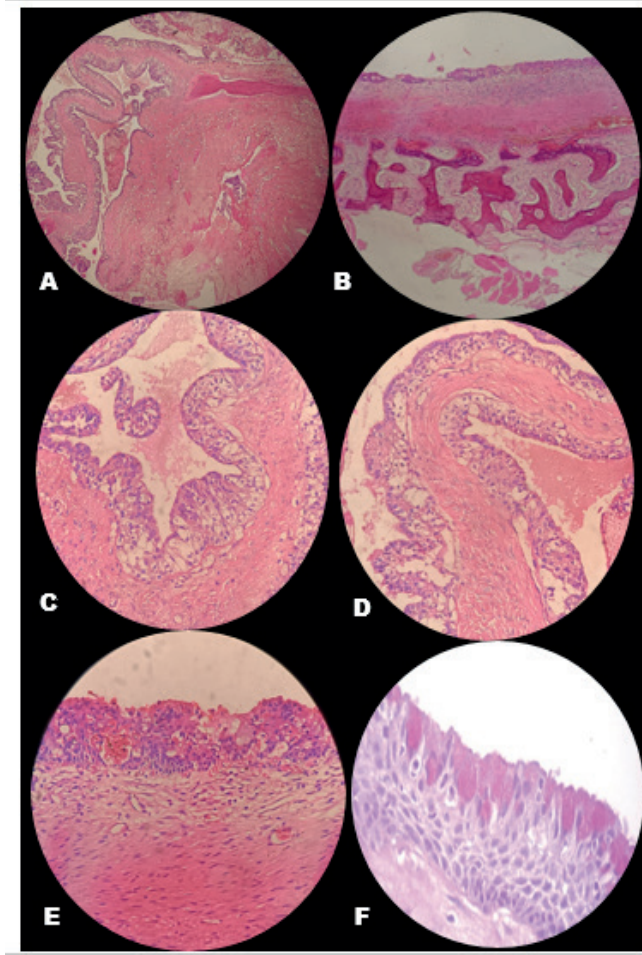


FIGURE 2

REFERENCES

- Noffke C, Raubenheimer EJ. The glandular odontogenic cyst: clinical and radiological features; review of the literature and report of nine cases. *Dentomaxillofac Radiol.* 2002;31:333-8.
- Gardner DG, Kessler HP, Morency R, et al. The glandular odontogenic cyst: an apparent entity. *J Oral Pathol.* 1988;17:359-66.
- Kramer IRH, Pindborg JJ, Shear M. *Histological typing of odontogenic tumours.* 2nd ed. Berlin: Springer; 1992.
- Suluk-Tekkesin M, Wright JM. The World Health Organization classification of odontogenic lesions: a summary of the changes of the 2022 (5th) edition. *Turk J Pathol.* 2022;38(2):168.
- Nel C, Robinson L, Roza ALOC, Ker-Fox J, Gomes NR, Fonseca FP, et al. Clinical and radiologic spectrum of glandular odontogenic cysts: a multicenter study of 92 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2022;133(5):593-603.
- Ajay A, Vasanthi V, Ramadoss R, Kumar AR. Glandular odontogenic cyst: review of literature and report of two cases. *J Cancer Res Ther.* 2024;20(1):488-92.
- de Campos WG, Araújo R, Martin V, et al. Glandular odontogenic cyst in the anterior mandible: a case report of a conservative approach and a recurrence detection. *Diagnostics (Basel).* 2023;13(8):1452.
- Sreekumar VN, Salim S, Cherian SK, John R. A vacillating swelling in maxillary anterior region: diagnostic challenges and management. *Natl J Maxillofac Surg.* 2020;11(2):292-7.
- Heiliczor S, Shmuly T, Avishai G, et al. Histopathological and histomorphometric analysis of glandular odontogenic cyst: a diagnostic aid. *Oral Dis.* 2023;29(8):3306-12.
- Demetriades N, D'Innocenzo R, Kahn MA. A clinico-pathologic correlation. Glandular odontogenic cyst. *J Mass Dent Soc.* 2005;54(3):34-7.
- Kaplan I, Anavi Y, Hirshberg A. Glandular odontogenic cyst: a challenge in diagnosis and treatment. *Oral Dis.* 2008;14:575-81.
- Kaplan I, Anavi Y, Manor R, Sulkes J, Calderon S. The use of molecular markers as an aid in the diagnosis of glandular odontogenic cyst. *Oral Oncol.* 2005;41:895-902.
- M S, Periasamy S, Kumar SP, Thota R. Glandular odontogenic cyst: a diagnostic and management dilemma. *Cureus.* 2021;13(12):e20701.
- Momeni Roochi M, Tavakoli I, Ghazi FM, Tavakoli A. Case series and review of glandular odontogenic cyst with emphasis on treatment modalities. *J Craniomaxillofac Surg.* 2015;43(6):746-50.
- Kwon YE, Choi SY, Kim MS, An CH, An SY. Glandular odontogenic cyst: a ten-case series and literature review with emphasis on radiological diagnostic challenges. *BMC Oral Health.* 2025;25(1):1989.
- Sikder MA, Khan AA, Ferdousi F, Pradhan L, Tareq BH. Reconstruction of oral mucosal defect with oven dried human amniotic membrane graft: a case report. *Bangladesh J Med Sci.* 2010;9(3):170-173. doi:10.3329/bjms.v9i3.6480.
- Das D, Bhuyan L, Panda A, Mishra P, Mohanty A. Hybrid odontogenic tumor of calcifying epithelial odontogenic tumor and ameloblastoma: a rare case report. *Bangladesh J Med Sci.* 2026;25(10):S174-S177. doi:10.3329/bjms.v25i10.86643.
- Alam MK, Ayub AAM, Taib H. Interdisciplinary care of the periodontally compromised elderly patient. *Bangladesh J Med Sci.* 2018;17(1):161-168. doi:10.3329/bjms.v17i1.35298.
- Alam MK, Ganji KK, Alzarea BK, Patil SR, Sghaireen MG, Basri R, et al. 3D CBCT assessment of the mandibular canal in a Saudi Arabian subpopulation. *J Hard Tissue Biol.* 2019;28(1):87-92. doi:10.2485/jhtb.28.87.