

Glandular Odontogenic Cyst of Anterior Maxilla - A Rare Case Report

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Abstract

Rationale: Glandular odontogenic cyst (GOC) is a rare type of cyst of the jaws, which shares histological features with that of lateral periodontal cyst, botryoid cyst, radicular cyst with mucous metaplasia, or low-grade mucoepidermoid carcinoma, making it difficult to come to a definitive diagnosis. Not many cases of GOC have been reported in the literature. **Patient Concerns:** A 40-year-old male patient presented with pain in the upper right front tooth region. Mild extraoral swelling, obliterating the right nasolabial fold, was present. Tenderness on percussion was present in involved teeth. **Diagnosis:** On histopathological examination, GOC was confirmed featuring pseudostratified columnar cells with cilia, goblet cells, and mucous cells. **Treatment:** Complete enucleation of the cyst was done. **Outcomes:** No recurrence was noted on 1-year follow-up. **Take-away Lessons:** Due to high recurrence rate and aggressive nature of the cyst, it is important to plan proper management and long-term follow-up.

Keywords: Aggressive, anterior maxilla, enucleation, glandular odontogenic cyst, odontogenic cyst

INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare but now well-known type of developmental odontogenic cyst. It was originally reported by Padayachee and Van Wyk in 1987 as “sialo-odontogenic cyst.”^[1] Later Gardner in his paper titled, “The GOC: An apparent entity,” in 1988, suggested that GOC originates from odontogenic tissue and named it “GOC.”^[2] In 1992, the World Health Organization (WHO) included GOC in the classification as developmental odontogenic cysts. The WHO describes GOC as “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 crypts or cyst-like spaces within the thickness of the epithelium.”^[3]

GOC has a frequency rate of 0.012%–1.3% among all cysts of the jaws and its prevalence is nearly 0.2%. Clinically, it appears as an asymptomatic slow-growing swelling. The age range reported is 14–75 years, with slight male predilection. GOC is predominantly seen in the mandible (70%), mainly the anterior region. Radiographically, it can be both a unilocular (53.8%) or a multilocular (46.2%) radiolucent lesion, with well-defined margins. Histologically, GOC

shows a non-keratinized stratified squamous epithelial lining, focal plaque like thickening within the lining, microcysts or intraepithelial crypts containing mucin, mucous cells and hyaline bodies, eosinophilic cuboidal or columnar cells that may be ciliated, papillary projections of epithelium and absence of inflammation in the subepithelial connective tissue.^[4] Treatment methods vary from conservative to aggressive resection. A recurrence rate of 30% has been reported, and multicystic lesions have shown a recurrence rate as high as 55% when treated conservatively.^[5]

We report a case of GOC in the anterior maxilla because of its comparative rare nature, emphasizing its clinical, radiographical, and histopathological features as well as successful surgical management.

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CASE REPORT

A 40-year-old male patient reported to the outpatient department with the chief complaint of pain in the right front tooth region for 3 months. The pain was moderate, intermittent, and dull in nature, aggravated on chewing hard food items and relieved on medication. Medical and family history was nonsignificant. On extraoral examination, there was mild swelling noted in the middle third of the face obliterating the nasolabial fold, and approximately 1 cm × 1.5 cm, extending from the right ala of the nose medially to the vertical line drawn from the pupil of the eye laterally, superiorly from 2 cm above the vermilion border of the upper lip to the vermilion border of the upper lip inferiorly [Figure 1]. Intraoral examination revealed deep caries with respect to 17 and tenderness on percussion with respect to 17, 14, 13, 12, 11, and 21. Mild diffuse intraoral swelling was noted extending from 12 to 14; the gingiva and the overlying mucosa were normal. Fixed partial prosthesis was present with respect to 21, 22, 23, 24, and 25. Based on the patient's history and clinical examination, a



Figure 1: Frontal view showing mild swelling in the right middle third of the face obliterating the right nasolabial fold

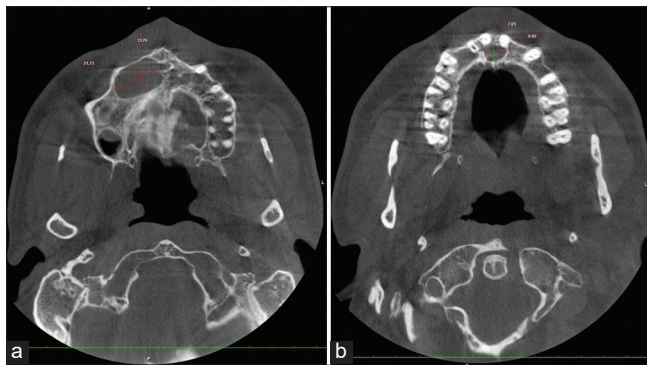


Figure 3: Cone-beam computed tomography (axial cut). (a) An oval-shaped, unilocular, homogeneously hypodense region measuring 13.29 mm (labiopalatally) × 21.73 mm (mediolaterally) in size extending from 21 to 14 region. (b) An oval-shaped, unilocular homogeneous hyperdense region seen in the palatal aspect of 11 and 21 measuring 7.05 mm (labiopalatally) × 9 mm (mediolaterally) in size

provisional diagnosis of chronic apical periodontitis was given. The presence of periapical abscess, radicular cyst, or a benign tumour of the jaws was added as differential diagnoses. All haematological parameters were within normal limits.

Radiological investigations included intraoral periapical radiograph, orthopantomogram (OPG), and cone-beam computed tomography (CBCT) scan. OPG revealed a well-defined, unilocular, radiolucent lesion with corticated margins, involving the right maxilla extending from the left central incisor, crossing the midline, till the right first bicuspid region (14–21 region) with no teeth displacement or root resorption [Figure 2].

The CBCT scan showed an oval-shaped, unilocular, homogeneously hypodense region, extending mediolaterally from the mesial aspect of 11 till the region of 15 and palatally to the periapical region of 11 and 21. The lesion was approximately (in mm): 13.29 (labiopalatally) × 21.73 (mediolaterally) × 15.56 (superoinferiorly) in size [Figures 3 and 4]. Periphery of the lesion was well demarcated. Expansion of the maxilla secondary to the lesion was present. There was loss of lamina dura with respect to all of the above-mentioned teeth. No extension to the ipsilateral maxillary antrum was noted. No calcifications were noted within the cavity of the lesion.



Figure 2: Preoperative orthopantomogram showing a unilocular, radiolucent lesion in the right maxilla extending from 14 to 21 region with no tooth displacement or root resorption

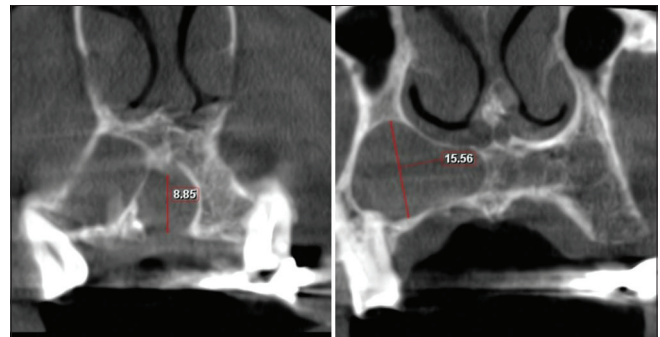


Figure 4: Cone-beam computed tomography (coronal cut). Superior-inferior extension of the cyst measuring 8.85 mm in 21 and 11 region and 15.56 mm in the premolar region

Endodontic treatment was done with respect to 11, 12, 13, 14, and 21. Under local anaesthesia, a crevicular incision was made and the lesion was surgically enucleated, followed by apicoectomy with respect to 11, 12, and 21 [Figures 5 and 6]. Closure was completed using resorbable, 3-0 vicryl suture. No intra- or postoperative adverse events were noted. The patient was followed up regularly at the interval of 2 weeks; no recurrence has been noted after 1 year of management [Figure 7].

Hematoxylin and eosin-stained tissue section revealed a cystic lumen lined by epithelium and a connective tissue capsule. The cystic lining was nonkeratinized stratified squamous type with variable thickness. It also exhibited pseudostratified columnar cells with cilia and goblet cells in a few areas. Mucous cells were also seen in certain areas. Underlying connective tissue showed bundles of collagen fibers, dense inflammatory cell infiltrate, blood vessels, and macrophages [Figure 8].

DISCUSSION

Padayachee and Van Wyk in 1987 reported two cases of GOC, which they termed as “sialo-odontogenic cyst,” because of the microscopic familiarity to the salivary gland tissue and

proposed that this lesion has three characteristic features: (1) it is radiologically multilocular and intrabony, (2) it can recur, and (3) it is multicystic.^[1]

Gurler *et al.* in 2017 stated that the prevalence of GOC varies from 0.012% to 1.3% of all jaw cysts, with the mean being 0.17%. Clinically, GOC most commonly involves mandibular anterior region and is usually an asymptomatic, slow-growing swelling,^[6] but in our case report, the cyst was noted in the anterior maxillary region.

In the literature, a predilection for the mandible was noted, mainly the anterior tooth region and gender predilection was nearly equal. The age range of 14–85 years was reported.^[5]

Radiographically, GOC is a localized, intraosseous radiolucent lesion with well-defined borders, which can be either multilocular or unilocular. Cortical bone thinning, perforation,

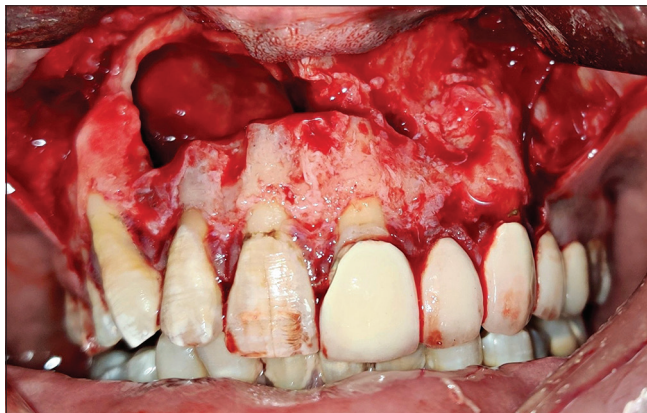


Figure 5: Intraoperative image showing cystic cavity after enucleation in the right anterior maxilla



Figure 7: Postoperative orthopantomogram after 1 year showing adequate bone formation in the region of cystic cavity with no recurrence



Figure 6: Enucleated cyst specimen with intact lining, as measured against a 2-ml disposable syringe

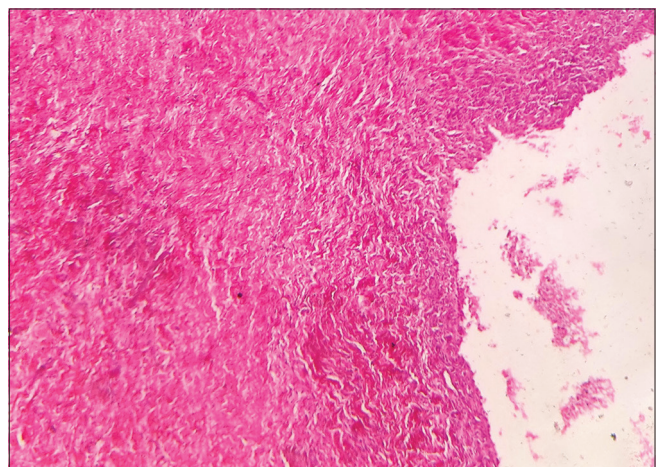


Figure 8: Hematoxylin- and eosin-stained tissue section revealed a cystic lumen lined by nonkeratinized stratified squamous epithelium with pseudostratified columnar cells with cilia and goblet cells in few areas and a connective tissue capsule showing bundles of collagen fibers, dense inflammatory cell infiltrate, blood vessels, and macrophages

Table 1: A list of major and minor microscopic criteria for histopathological diagnosis of glandular odontogenic cyst

Major criteria	Minor criteria
Squamous epithelial lining, with a flat interface with the connective tissue wall, lacking basal palisading	Papillary proliferation of the lining epithelium
Epithelium exhibiting variations in thickness along the cystic lining with or without epithelial “spheres” or “whorls” or focal luminal proliferation	Ciliated cells
Cuboidal eosinophilic cells or “hobnail” cells	Multicystic or multiluminal architecture
Mucous (goblet) cells with intraepithelial mucous pools, with or without crypts lined by mucous-producing cells	Clear or vacuolated cells in the basal or spinous layers
Intraepithelial glandular, microcystic, or duct-like structures.	-

root resorption, and displacement of the involved teeth have also been reported.^[5,7] In the present case report, a well-defined unilocular radiolucent lesion with corticated margins, involving the right maxilla, with no tooth displacement or root resorption, was noted on OPG.

GOC is frequently misdiagnosed as it shares its histopathological features with that of lateral periodontal cyst, radicular cyst, dentigerous cyst, and central mucoepidermoid carcinoma.^[6] Kaplan *et al.*, in 2008, proposed a list of major and minor microscopic criteria for histopathological diagnosis of GOC [Table 1].^[5]

There is no consensus on treatment of GOC in the literature; it totally depends upon surgeon’s preference. Enucleation and curettage is a commonly opted procedure. Marsupialization, resection of the jaw, and procedures such as application of Carnoy’s solution, use of bone grafts, and cryosurgery have also been used successfully.^[8] In the present case, complete enucleation of the cyst was performed under local anaesthesia followed by primary closure. Postoperative recovery was uneventful.

The range for recurrence rate for GOC lies between 21% and 55%, as reported in the literature. The high recurrence rate of GOC might suggest its aggressive nature. Recurrence is not only associated with the biology of the lesion but also with the management.^[9,10]

CONCLUSION

Careful and thorough histopathological examination is the most important pillar in the diagnosis of GOC as the clinical as well as the radiological findings tend to overlap with that of other odontogenic lesions. A high recurrence rate and aggressive nature of GOC demands proper management and long-term follow-up.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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