Glandular Odontogenic Cyst: A Case Report
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ABSTRACT
Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst that is considered to be a distinct entity because of its atypical histopathological characteristics. It has some characteristics in common with the lateral periodontal cyst, botryoid odontogenic cyst and low grade mucoepidermoid carcinoma, which makes its diagnosis challenging for pathologists. This cyst occurs over a wide age range with average age being 50 years, involving either jaw, but the anterior region of the mandible is the most commonly affected area. It has a propensity to grow to a large size and to recur if treated conservatively. Here, we report a case of GOC in a 38 year old male patient in the posterior region of maxilla, which is very rare.

Key words: Botryoid odontogenic cyst, Glandular odontogenic cyst, lateral periodontal cyst, low grade mucoepidermoid carcinoma.

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INTRODUCTION
Glandular odontogenic cyst (GOC) was first documented as 'sialo-odontogenic cyst' by Padayachee and Van Wyk in 1987, who described two cases that resembled both the botryoid odontogenic cyst and the central mucoepidermoid tumour of the jaws. But, they concluded
that the lesions differed sufficiently to justify as different entities\(^1\). Gardner \textit{et al.}, in 1988 established it as a distinct entity and proposed the term 'GOC'\(^2\), which was later adopted by world health organization. Recently, High \textit{et al.}, (1996) proposed the term 'polymorphous odontogenic cyst' for this cyst because of its aggressive growth pattern\(^3\). The lesion was also described as mucoepidermoid odontogenic cyst by Sadeghib (1991).\(^4\) Although GOC is rare, it is a well-known clinical entity and is important to recognize and diagnose due to its aggressive behaviour and tendency to recur. GOC is as frequently as seen twice in males and the most-commonly affected site is the anterior mandible. It occurs over a wide age range of 10-90 years, with mean age of 49.5 years and peak frequency in the 6\(^{th}\) decade. The mandible seems to be affected more commonly (87.2\%) than the maxilla.\(^5\)

Radiographically, glandular odontogenic cysts show well-defined, unilocular or multilocular radiolucency without specific diagnostic characteristics.\(^6\)

Histologically, GOC shows a lining of non-keratinized stratified squamous epithelium of variable thickness, focal plaque like thickenings within the lining, with superficial layer of epithelium consisting of columnar or cuboidal cells, occasionally with cilia and numerous goblet cells. The epithelium has glandular or pseudo glandular structure, with intra-epithelial crypt or microcyst formation.\(^2,7\)

GOC is a rare entity, so here we present a unique case of a GOC with clinico-histopathological features.

### CASE REPORT

A 38 year old male patient reported with the chief complaint of pain and swelling in left upper back tooth region since one month. Extraoral examination showed slight asymmetry with fullness of the left side of the face. Skin over the swelling was normal with no secondary changes. Patient has undergone root canal treatment of 24 and 25, three years before. Intraoral examination revealed a single diffuse swelling in the left maxillary posterior region extending from 25 to 27, approximately 2.5 × 2.0cm in size (Figure 1). Vestibular obliteration was seen in the region of 25,26 and 27. Overlying mucosa

![FIG 1. Diffuse swelling in the maxillary posterior region extending from 25 to 27 region.](image-url)
was smooth and erythematous. On palpation, the swelling was firm, non-tender and non-mobile. No secondary changes in the mucosa were seen. Submandibular lymph nodes on left side were palpable and tender. A provisional diagnosis of radicular cyst and differential diagnosis of calcifying odontogenic cyst was given. Radiographic examination revealed a multilocular radiolucent area mixed with slight radio-opacities and having diffused margins extending from 25 to 27 region (Figure 2). Incisional biopsy was done and sent for histopathologic examination. Two tissue bits were received. On gross examination, the specimen was pinkish white in colour with smooth surface measuring about 1.2cm X 0.6 cm in dimensions and soft in consistency. Histopathologic examination revealed a cystic wall lined by non-keratinized stratified squamous epithelium with surface layer composed of ciliated columnar cells and mucous cells (Figure 3). The epithelium showed spherule arrangement at some areas with attempts to form microcysts. Few areas of epithelium exhibited plaque like thickenings resembling the epithelium of lateral periodontal cyst and botryoid odontogenic cyst. Numerous Periodic acid Schiff positive (Figure 4) and mucicarmine positive (Figure 5) goblet cells were present in the superficial.
FIG 5. Photomicrograph showing special stained mucicarmine positive mucous cells.

epithelium. The epithelial connective tissue interface was flat. Diffuse infiltrations of chronic inflammatory cells were seen in the connective tissue wall. Consistent with these histopathological features, a diagnosis of glandular odontogenic cyst was given.

DISCUSSION
GOC is a rare cyst of the jaws. Its characteristic histopathological features make this lesion a distinct entity. Most common site of occurrence is the anterior mandible. Incidence in the maxilla is rare, but cases were reported from the anterior region. However, in this case, the lesion was found in the posterior maxilla. About four cases have been reported till date and ours is probably the fifth case.

Padayachee and Van Wyk for the first time described the typical features of the cyst in comparison with two other similar lesions. They indicated that it was intrabony and radio logically gives multilocular apperance; that it could recur if not adequately excised and that it was multicystic, with the cystic spaces lined by a non-keratinised epithelium akin to that of reduced enamel epithelium. Epithelial thickenings or plaques were present in the cyst linings which resembled botryoid odontogenic cyst. Mucous and cylindrical cells formed an integral part of the epithelial component and mucinous material within the cystic spaces was a prominent feature giving the appearance of central mucoepidermoid tumour. But, after careful analysis, they concluded that the lesions differed sufficiently to warrant separation as an entity.

Gardner et al preferred the term Glandular odontogenic cyst than sialo-odontogenic cyst and believed that its histological features and biological behaviour were sufficiently distinct for it to be regarded as an entity.

Histopathologically, GOC should be differentiated from the lining of lateral periodontal cyst which exhibits focal thickenings and glycogen-rich epithelial cells, similar to those observed in GOC. However, the identification of ciliated epithelium and duct-like spaces with mucous cells specifically differentiates GOC from LPC and botryoid odontogenic cyst (BOC) and favors the diagnosis of GOC. The differentiation of low-grade CMEC from GOC is more important and
challenging. However, CMECs do not show superficial cuboidal cells, epithelial whorls, ciliated cells and intraepithelial microcyst or duct-like structures. Kaplan and co-authors recently suggested that a diagnosis of GOC has to be based on the mandatory presence of the five major features. These are squamous epithelium, varying thickness, cuboidal eosinophilic (‘‘hobnail’’) cells, mucous (goblet) cells and intraepithelial glandular or duct-like structures. The present case had all the characteristic features of GOC. Immunohistochemical studies suggested that the central MEC and the GOC were distinct entities with different cytokeratin (CK) profiles, and that expression of CKs 18 and 19 could be useful adjunctive tools in differentiating these two entities.

The treatment of GOC ranges from various forms of conservative approach like enucleation, marsupialization, and curettage with or without peripheral ostectomy to marginal resection and segmental resection. The prognosis of this cyst still remains unclear. However, the aggressive nature of the lesion has been reported and the recurrence rate is directly related to the size of the lesion. Therefore, many authors prefer marginal and segmental resection for large multilocular lesions due to their tendency to recur after conservative treatment.

CONCLUSION

In conclusion, GOC being a rare occurrence in maxilla, it is important to consider histopathological features for its diagnosis since it bears resemblance to lesions like lateral periodontal cyst and low grade mucoepidermoid carcinoma. Because of its aggressive biologic behaviour and propensity for recurrence adequate treatment and follow up of patient is required.

REFERENCES


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