



## Glandular Odontogenic Cyst Rarely Seen in Maxilla- A Case Report and Review of Literature

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### Abstract

Glandular odontogenic cyst is rare developmental odontogenic cyst of jaws, that ranges from 0.012 to 1.3%. Generally, this cyst is seen in the anterior areas of the mandible and with the mean age being 49 years. GOC is often misdiagnosed because of its overlapping histopathological features with other odontogenic cysts such as lateral periodontal cyst (LPC) or Botryoid cyst and Central low-grade Mucoepidermoid carcinoma. Cyst have an unpredictable, potentially aggressive behavior, and has the propensity to grow in large size with relatively high recurrence rate. Here, we present a case of GOC in a 50 year old female patient in left anterior maxillary region.

**Keywords:** Cilia, glandular odontogenic cyst, mucous cell, hobnail cells, sialo-odontogenic cyst.

### Introduction

Glandular odontogenic cyst (GOC) is rare, uncommon jaw bone cyst of odontogenic origin described in 1988 by Gardener et. al as a distinct entity<sup>[1]</sup>. GOC was first documented as “Sialo-odontogenic cyst” by Padayachae and Van Wyk in 1987 who published two cases that resembles both Botryoid odontogenic cyst and Central mucoepidermoid tumor of jaw<sup>[2]</sup>. High et. al (1996) proposed the term, ‘polymorphous odontogenic cyst’ for this cyst, because of its aggressive growth pattern<sup>[2]</sup>. It was later in 1992, the WHO accepted GOC as a distinct pathological entity, and included

it in the classification of developmental odontogenic cysts<sup>[1]</sup>. Clinically, the most common site of occurrence is the mandibular anterior region, presenting as an asymptomatic slow growing swelling<sup>[4]</sup>. GOC occurs mostly in the middle age, and has a slight male predilection<sup>[4]</sup>. Radiologically, these cysts may be unilocular or multilocular with a well-defined border<sup>[4]</sup>. Histologically, it resembles to lateral periodontal cyst, botryoid odontogenic cysts, radicular and residual cysts with mucous metaplasia, and low grade mucoepidermoid carcinoma<sup>[1]</sup>. Although it has been noted, GOC has aggressive potential, a high incidence of cortical

perforation, and a relatively high rate of recurrence, especially in cases treated conservatively.

Therefore, the correct diagnosis is a major challenge and is of extreme clinical importance<sup>[1]</sup>. GOC in maxilla is very rare, so here we present a case of GOC and clinico-pathological features of this rare cyst

### CASE REPORT

A 50 year old female patient reported to dept. of OMFS with chief complain of pain in right upper front teeth region since 1 month and pain was dull, intermittent in nature and was not relieved on taking medication. She also complained of asymptomatic swelling in palate since 12 months which gradually increased in size till the present day. Intraorally, Gold onlay's were present in 11,12,21,22 teeth which were placed 35 years back. Discoloration of 11,12,21 teeth were present[Figure1]. The swelling was present palatally measuring approximately 2×3 cm in size, firm in nature with well-defined borders, which was not tender on palpation, extending from 11 to 13 tooth region[Figure 2]. Vitality test showed delayed response of 11,12,13,21,22. And 14,15 showed normal response Radiographically, OPG shows a radiolucent lesion with well-defined radiopaque sclerotic margin, of approximately 4×3 cm in size, extending from 11 to 15 tooth region[Figure 3].Based on the clinical and radiographic findings, a provisional diagnosis of radicular cyst or odontogenic keratocyst was made. Root canal treatment was done in 11 and 12 teeth. Enucleation of the cyst was done followed by apicectomy[Figure 4] and the defect was filled with egg shell derived hydroxyl appetite bone graft[Figure 5] and specimen was sent for histopathological examination [Figure6]. Histopathological examination showed odontogenic epithelial lining made up of cuboidal basal layer and stellate reticulum like cells arranged in the superficial part of the lining[Figure 7]. Numerous goblet cells and few mucous pulling area were seen with few areas showing cilia[Figure 8]. There was the presence of epithelial plaque like proliferation in some areas. Connective tissue capsule showed mild

to moderate amount of chronic inflammatory cell infiltrate and vascularity. Based on all the above findings, it was diagnosed as GOC. Follow up period was upto 1 year. OPG shows well-defined new bone formation in the defect.[Figure 9].



**Figure 1:** Gold onlays and discoloration of teeth.



**Figure 2:** Swelling present palatally in the anterior teeth region.



**Figure 3:** OPG shows a radiolucent lesion with well-defined radiopaque sclerotic margin, of approximately 4×3 cm in size, extending from 11 to 15 tooth region.



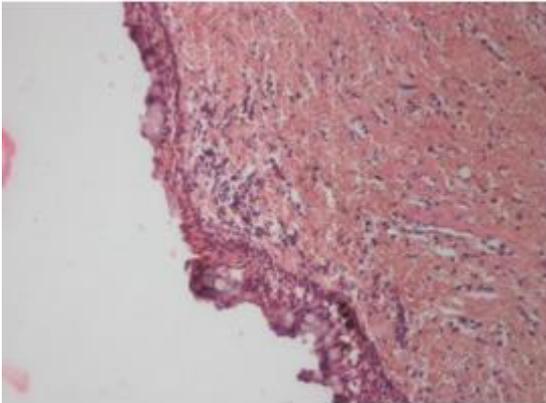
**Figure 4:** Enucleation of the cyst was done followed by apicectomy.



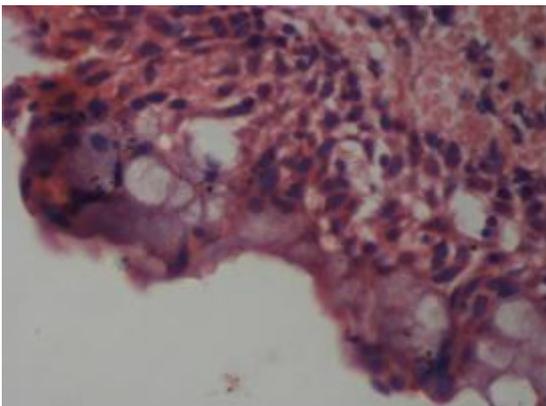
**Figure 5:** Egg shell derived hydroxyl appetite bone graft was placed.



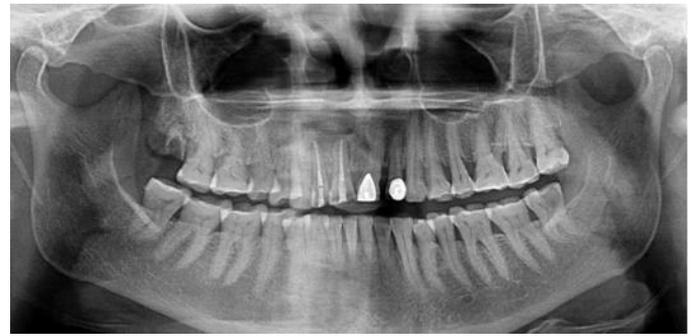
**Figure 6:** specimen measuring of 4×3 cm in size.



**Figure 7:** Photomicrograph shows odontogenic epithelial lining made up of cuboidal basal layer and stellate reticulum like cells (H&E stain, ×4).



**Figure 8:** Photomicrograph shows numerous goblet cells and few mucous pulling area (H&E stain, ×40).



**Figure 9:** Post operative OPG showing well defined radiopacity in the defect (at 1 year).

### Discussion

Because of intrinsic biological behavior and multilocularity it has high recurrence rate. Initially reported and coined by Padaychee and Van Wyk in 1987, it was later in 1992, GOC was included in the WHO typing of tumors under the term GOC or sialo-odontogenic cyst<sup>[1]</sup>.

The main clinical finding of this cyst is a painless local swelling, but it is non-specific. The lesion may cause pain due to compression of a neurovascular bundles, secondary infection or inflammation. There is a slight male predilection and lesions occur mostly in middle-aged patients and the anterior mandible is the most common site for occurrence of this cyst <sup>[2]</sup>. In contrarary to the literature, in our case, the lesion was seen in 50 year old female patient and in maxillary anterior teeth region, with gold onlays and discoloration of anterior teeth and there was asymptomatic, slow growing swelling which was firm, non-tender and was attached to the overlying tissue. On aspiration, clear and low viscosity fluid content may be a helpful clinical for the indication of GOC. The fluid may be brownish-red, which can attribute to blood, perhaps because of the previous surgery or secondary inflammation <sup>[1]</sup>.

Radiographically, the lesions typically present as a radiolucent unilocular or multilocular lesion. Same features can be found in odontogenic keratocyst, unicystic or multicystic ameloblastoma, CMEC, lateral epithelial cyst and botryoid odontogenic cyst (BOC) <sup>[3]</sup>.

According to Kaplan *et al.* histopathologically it exhibits:<sup>[1]</sup>

Major criteria includes Non-keratinized squamous epithelial lining with a flat interface., Presence of “spherules”/knobs or “whorls” or focal luminal proliferations., Epithelial lining exhibits surface cuboidal eosinophilic cells or “hob-nail” cells., Goblet cells with intraepithelial mucous pools which are with or without crypts that are lined by mucous producing cells., Intraepithelial glandular microcystic or duct-like (pseudoglandular) structures are present.

Minor criteria includes Papillary proliferation, Ciliated cells, Multicystic or multiluminal architecture, Clear or vacuolated cells in basal or spinous layer.

GOC should be distinguished from Lateral Periodontal Cyst, Botryoid Odontogenic Cyst, and Central Mucoepidermoid Carcinoma as they have similar histological features. LPC is a developmental odontogenic cyst lined by thin nonkeratinized epithelium and also exhibits focal epithelial thickenings and glycogen rich epithelial cells, similar to those observed in GOC's. BOC is a locally aggressive polycystic variant of LPC, that also shows similar histologic feature of GOC. However, the identification of ciliated epithelium and duct like spaces with mucous cells is differentiating factor of GOC from LPC and BOC. Low grade Central Mucoepidermoid Carcinoma and GOC also has the similar histological features and is more important and difficult to diagnose. However, superficial cuboidal cells, epithelial whorls, ciliated cells, and intraepithelial microcyst or duct like structures are not typical for CMEC and their presence or absence can help in establishing a definitive diagnosis. GOC can be differentiated from CMEC by immunostaining with CK-18 and 19. GOC exhibited decreased p-53 positivity and increased Ki-67 index when compared to CMEC.<sup>[2]</sup> Rarely GOC can be seen in association with Ameloblastoma, squamous odontogenic tumor like hyperplasia, solid epithelial down growths into the cyst wall, satellite microcysts, hyaline bodies and epithelial ghost cell calcification<sup>[6]</sup>.

GOC is treated by enucleation or curettage alone but is associated with a high recurrence rate. Small

unilocular lesions can be treated by enucleation like the case presented here. Surgical treatment of large lesions can be done by ienucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw resection in multilocular cases. Marsupialization followed by second phase surgery is option for lesions approaching vital structures. Follow up should be continued for at least 3 years (up to 7 years in cases with features associated with increased risk of recurrence rate due to its intrinsic biological behavior, multilocularity of the cyst and incomplete removal of the lining following conservative treatment)<sup>[2]</sup>.

### Conclusion

In terms of establishing a diagnosis of Glandular Odontogenic Cyst, it must be taken into consideration that this is a rare lesion, and diagnosis is difficult due to the strong similarities with Central low-grade Mucoepidermoid Carcinoma. Adequate treatment of GOC is required because of its aggressive biologic behavior and high recurrence rate.

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