**CASE REPORT**

**Glandular odontogenic cyst associated with an impacted maxillary canine: A rare case report**

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**Abstract**

Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst of the oral cavity. The most common site of occurrence for GOC is a mandibular anterior region, presenting typically in the middle age and has a minor male predilection. This article presented a case of GOC and focused on the review of clinico-pathologic features of the lesions considered in the differential diagnosis. A 17-year-old female patient reported with pain and swelling in the right upper front tooth region since 8 months. Radiographically, a well-defined unilocular radiolucency is seen in relation to impacted right maxillary canine. A provisional diagnosis of the dentigerous cyst was given. The excised tissue revealed a stratified squamous epithelial lining exhibiting cuboidal cells with goblet cells which are numerous and foci of epithelial cells presenting with eosinophilic material resembling mucin. The present case was associated with an impacted maxillary canine, which is found to be a very rare location for GOC. Thus, this case report of GOC will add to the existing knowledge of rare cysts.

**Keywords:** Canine, glandular odontogenic cyst, impacted tooth, mucous cells

**Introduction**

Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst. Padayachee and Van Wyk reported the first case of the cyst as a “sialo-odontogenic cyst” in 1987. This cyst was further recognized as a distinctive entity by Gardner et al., in 1988, as “GOC.”

Clinically, the most common site of occurrence for GOC is a mandibular anterior region (88%), seen most commonly as an asymptomatic slow growing swelling. GOC occurs typically in the middle age (40-60 years) and has a slight male predilection. Radiologically, they might show unilocular or multilocular radiolucency with a distinct border.

Histologically, GOC is characterized by a cystic lining of non-keratinized epithelium, mucous filled clefts, with papillary projections, nodular thickenings, and “mucous lakes.” It also includes vacuolated cuboidal basal cells.

Treatment of GOC includes enucleation with curettage and although a number of authors consider marginal resection to be added dependable treatment, due to the propensity of the cyst to recur following curettage and enucleation. Recurrences were seen in 30% of the cases.

This article presented a case of GOC and focused on the review of clinico-pathologic features of the lesions considered in the differential diagnosis.

**Case Report**

A 17-year-old female patient complained of pain and swelling in the right upper front tooth region since 8 months. On extra-oral examination, facial swelling of the size of 4 cm × 6 cm × 3 cm was present on the right anterior region and was non-tender with normal overlying mucosa. On intra-oral examination, a solitary swelling is present on the right side of the hard palate extending from 12 to 16 [Figure 1]. Comparing with the history and clinical examination, the panoramic radiograph was recommended. This discovered a well-defined unilocular radiolucency in relative to impacted right maxillary canine [Figure 2]. Aspiration of the lesion was carried out to rule out vascular and cystic lesions. The straw-colored aspirate fluid was obtained. A provisional diagnosis of the dentigerous cyst was given. A clinical differential diagnosis of an adenomatoid odontogenic tumor, calcifying epithelial odontogenic cyst, ameloblastoma, and odontogenic keratocyst was made.

The patient underwent surgical exploration under local anesthesia, and the whole lesion was excised all along with the impacted canine. The excisional tissue was sent for histopathological examination. In a histopathological analysis stratified squamous epithelial lining exhibiting superficial cuboidal cells with numerous goblet cells [Figure 3] were seen. In few areas, the superficial layers of epithelium showed
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eosinophilic cuboidal and ciliated columnar cells [Figure 4]. Foci of epithelial cells were also showing eosinophilic material resembling mucin [Figure 5] with clear or vacuolated cells seen in the spinous cell layer [Figure 6]. In the underlying connective tissue stroma, few chronic inflammatory cell infiltrate with predominantly lymphocytes was seen. Features were suggestive of GOC [Figure 6]. Follow-up for 1 year showed no evidence of recurrence.

Figure 1: A solitary swelling present on right side of the hard palate extending from 12 to 16

Figure 2: OPG reveals well-defined radiolucent lesion extending from 22 to mesial surface of 18 with well-corticated border and impacted 13

Figure 3: Stratified squamous epithelial lining exhibiting superficial cuboidal cells with numerous goblet cells and underlying connective tissue showing chronic inflammatory cells predominantly lymphocytes (H and E, original magnification ×200)

Figure 4: The epithelial superficial layer showed eosinophilic cuboidal and ciliated columnar cells (H and E, original magnification ×200)

Figure 5: Foci of epithelial cells showing eosinophilic material resembling mucin (H and E, original magnification ×400)

Figure 6: Clear or vacuolated cells seen in spinous cell layer (H and E, original magnification ×100)
Discussion

GOC is a rare cyst of odontogenic origin. According to Shah et al., only 111 cases of GOC were reported in the literature, and only 0.012% of the cysts were seen in the oral cavity have fulfilled the criteria of GOC histopathologically. GOC has a broad range of clinico-pathologic spectrum ranging from the lateral periodontal cyst (LPC) to a critical malignant neoplasm such as intraosseous mucoepidermoid carcinoma.[5]

The origin of GOC is very diverse. However, Mark and Stern et al. have proposed one of three possibilities:
1. A true cyst of glandular origin can arise either from entrapped salivary gland primordia or undifferentiated primitive epithelial rests that develop into the glandular epithelium.
2. An odontogenic primordial cyst in which the epithelial lining undergoes proplasia (metaplasia from a smaller specific differentiation to an added specific differentiation) into the glandular epithelium.
3. Low-grade mucoepidermoid carcinoma that forms an initial single cystic space as an alternative to the usual multicystic spaces.[6]

GOC does not exhibit exact or pathognomonic radiographical features. It may be seen as a multilocular or unilocular radiolucency with well-defined border, or as perifollicular radiolucency, simulating a hyperplastic follicle or dentigerous cyst.[7] On clinical and radiological examination, a differential diagnosis of other lesions such as dentigerous cyst odontogenic keratocyst, adenomatoid odontogenic tumor, and ameloblastoma can be made. In the present case, the dentigerous cyst was made as a provisional diagnosis; whereas, histopathologic examination revealed GOC.

Similar to our case, Qin et al.[8] reported five maxillary cases associated with unerupted teeth. Shimoyama and Horie[9] also reported a cyst in which clinical diagnosis was the dentigerous cyst, but histopathological features were consistent with GOC.

Histopathological features of the present case are correlated with the major and minor criteria given by Kalpan et al.[10] which includes squamous epithelial lining, flat interface, variation in thickness of the lining presence of epithelial spheres or whirls, no palisading, cuboidal eosinophilic cells or “hob-nail” cells, mucous goblet cells with intraepithelial mucous pools with or without crypts lined by mucous-producing cells and interepithelial glandular microcysts or duct-like structures as major criteria and papillary projections, ciliated cells, and clear or vacuolated cells in basal or spinous layer as minor criteria.

Histopathologically, GOC must be differentiated from LPC, botryoid odontogenic cyst (BOC), central mucoepidermoid carcinoma (CMEC), and a radicular or dentigerous cyst with mucous metaplasia as they display substantial overlie of histological features.

1. LPC is a developmental cyst and odontogenic in origin lined by thin non-keratinized squamous epithelium and also exhibits focal epithelial thickenings and glycogen-rich epithelial cells, comparable to those seen in GOC’s.[5]

2. BOC is a locally destructive polycystic variant of LPC, shows alike histomorphologic features with those of GOC, such as areas of glycogen-rich clear cells and epithelial plaques. On the other hand, the identification of duct-like spaces with mucous cells and ciliated epithelium specifically differentiated from LPC and BOC favors the diagnosis of GOC’s.[5]

3. The differentiation of low-grade central mucoepidermoid carcinoma from GOC particularly its multicystic variant is further important and complicated. Significant histopathological overlap exists among GOC and CMEC. However, superficial cuboidal cells, ciliated cells, epithelial whorls, and duct-like structures or intraepithelial microcyst are not characteristic for CMEC, and their occurrence or non-existence can help in establishing an ultimate diagnosis.

In addition, immuno-staining with cytokeratin-18 and 19 and their positivity in GOC might assist in differentiating GOC from CMEC.[10]

4. Radicular or dentigerous cysts with mucous metaplasia have also to be differentiated by means of GOC. The incidence of clear cells, microcysts, and epithelial spheres appears to be the majority helpful in the distinctive diagnosis of GOCs associated with an unerupted tooth from dentigerous cysts by means of metaplastic changes.[11] Mucous metaplasia of the epithelial lining of radicular cyst can be differentiated by the findings that radicular cyst is associated with the periapical area, non-vital tooth.[11]

The treatment of these cysts is a controversial issue. The most of the cases reported in the literature were treated with enucleation and curettage (83.5%). In addition, marsupialization and curettage in conjunction with Carnoy’s solution and cryotherapy have been used. However, some authors prefer marginal or segmental resection because of the high potential for recurrence and the aggressive nature of these lesions.[12]

Conclusion

This report of GOC’s can be added to the accessible knowledge of these uncommon cysts. The present case was associated with an impacted maxillary canine, which is found to be a very rare location for GOC. GOC may be missed because its histopathology is similar to that of many other conditions. Hence, all the tissue sections with proper criteria should be evaluated for the diagnosis of GOC.

Clinical significance

GOC’s are widespread in middle age collection, having an elevated predilection for mandible and trauma might be an impulsive factor for its incidence. The augmented reappearance rates can be owing to its inherent biological activities, multilocularity of the cyst and imperfect elimination of the lining subsequent to conservative treatment. Thus, the treatment should be added aggressive elimination over conservative approach, and a cautious extended term follow-up is also essential.
References


