

Glandular Odontogenic Cyst Mimicking Dentigerous Cyst: A Rare Case Report

Meghashyama Kulkarni, Sahana N.S., Satish T Yadav, Hajira Khatoon, Renuga S, Srinath SK

ABSTRACT

Background: Glandular odontogenic cyst is an uncommon developmental cyst of odontogenic origin. Till now, around 200 cases have been reported in the English literature, out of which approximately 25 of them are associated with an unerupted tooth. Herein we present a case report of a 7-year-old boy with swelling in the anterior maxillary region which was later diagnosed as a Glandular odontogenic cyst that mimicked a Dentigerous cyst.

Case presentation: A painless swelling was noted in the anterior palatal region. Orthopantomogram was advised which revealed a unilocular radiolucency associated with an impacted supernumerary tooth. Provisional diagnosis of Dentigerous cyst was made. The cyst was enucleated in conjunction with peripheral osteotomy.

The histopathological examination revealed a cystic capsule with non-keratinized stratified squamous epithelium with 2-4 cell layer thickness, with some areas showing epithelial plaque, microcysts, hobnail cells and few clear cells. Contemplating all the histological features, final diagnosis of Glandular odontogenic cyst was given.

Conclusions: The presented case emphasizes the importance of histopathological examination of the unusual and rarely observed Glandular odontogenic cyst which can be missed due to similarities with other entities. Recurrence rates being very high, follow up of the cases is imperative.

Keywords: Dentigerous cyst; Epithelial plaque; Glandular odontogenic cyst; Hobnail cells; Microcyst

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INTRODUCTION

Glandular odontogenic cyst (GOC) is an uncommon developmental odontogenic cyst with rare occurrence and histopathologically distinct features.¹ It was initially described as "sialo-odontogenic cyst" by Padayachee and Van Wyk in 1987 but later Gardner et al. in 1988 delineated this cyst as "glandular odontogenic cyst".^{2,3} In 1992, WHO classified GOC as an odontogenic cyst.⁴

Clinically they appear as an asymptomatic swelling, usually small in size. In few cases, pain and paresthesia has been documented. Anterior mandible is the most affected site.⁵ Age of occurrence is between 45 and 50 years. However, pediatric patients have also been affected. The male to female ratio seems to be nearly equal with slight male predilection.⁶ GOC has a recurrence rate of 35.9%, especially when conservative surgical treatment is done.⁷

Radiographic appearance of GOC is quite ambiguous and not pathognomonic. The lesion may appear as a unilocular or multilocular radiolucency, usually with well-defined margins and sometimes with scalloped borders.⁸

Histologically, GOC shows a non-keratinized stratified squamous epithelial lining, focal plaque like thickenings within the epithelium, microcysts, mucous cells, eosinophilic cuboidal or columnar cells (hobnail cells) with minimal or complete absence of inflammation in the subepithelial connective tissue.⁹

Due to rarity of the cyst, the histopathological diagnosis re-

Department of Oral Pathology and Microbiology, Government Dental College and Research Institute, Rajiv Gandhi University of Health Sciences, Bengaluru, India.

Corresponding author: Meghashyama Kulkarni, Department of Oral Pathology and Microbiology, Government Dental College and Research Institute, Rajiv Gandhi University of Health Sciences, Bengaluru, India. email id: meghashyamakulkarni@gmail.com

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mains a challenge and there exists no universally accepted microscopic criteria which makes the diagnosis even more strenuous. The aim of this report is to present an unusual case of GOC that clinically and radiologically mimicked a dentigerous cyst which arose from an impacted supernumerary tooth in the anterior maxilla.

CASE DESCRIPTION AND RESULTS

A 7-year-old male, with no medical history, was referred from a private clinic to our Institute, for evaluation of a painless swelling in the anterior palatal region. The swelling was present

since 3 and half months which was not associated with pain and subsided on medication. Intraorally, a diffuse swelling was seen on the left side of the anterior palate (Fig. 1). Orthopantomogram (OPG) revealed presence of a supernumerary tooth which was palatal to 21 (Fig. 2).

Table 1: Kaplan's criteria for diagnosing GOC

MAJOR CRITERIA	MINOR CRITERIA
<ul style="list-style-type: none"> • Squamous epithelial lining with a flat interface • Epithelium exhibiting variations in thickness along the cystic lining • Cuboidal eosinophilic cells or "hobnail" cells • Mucous cells with intraepithelial mucous pools, with or without crypts • Intraepithelial glandular, microcystic or duct-like structures 	<ul style="list-style-type: none"> • Papillary proliferation of the lining epithelium • Ciliated cells • Multicystic or multiluminal architecture • Clear cells in the basal or spinous layers

Table 1: Histopathological criteria given by Kaplan et al. in 2008 for diagnosis of Glandular odontogenic cyst. It comprises of 5 major and 4 minor criteria.

tal to 21 (Fig. 2). Cone-beam computed tomography (CBCT) scan showed an expansile unilocular radiolucency measuring about 17.5mmx27.5mm on the left side of the anterior maxilla involving the roots of 21,63, developing tooth bud to 22 and a supernumerary tooth located palatal to 21 (Fig. 3A,3B). Provisional diagnosis of dentigerous cyst was given. Since the lesion was quite small in size, the treatment of choice was enucleation and peripheral osteotomy along with extraction of the supernumerary tooth. The surgery was done under general anesthesia (Fig. 4).

On gross examination, the specimen received was greyish white in colour measuring about 2.4x1.6x0.4cm (Fig. 5). The stereo-microscopic image in Figure 6 shows a thick cystic capsule that is attached to the neck of the tooth.

The microscopic examination of Hematoxylin and Eosin stained section revealed thin, non-keratinized stratified squamous epithelium made up of flat to cuboidal cells varying from 2-4 cell layers in thickness with flat connective tissue interface (Fig. 7). Another bit showed epithelial plaque like thickenings in some areas along with numerous clear cells and few microcysts (Figs. 8A, 8B and 8C). The superficial layer of the epithelium showed eosinophilic cuboidal cells, also called "hobnail cells" (Fig. 8D)

Mucous cells and glycogen rich cells were evident in mucicar-



Fig. 1: Diffuse swelling noted on left anterior maxilla

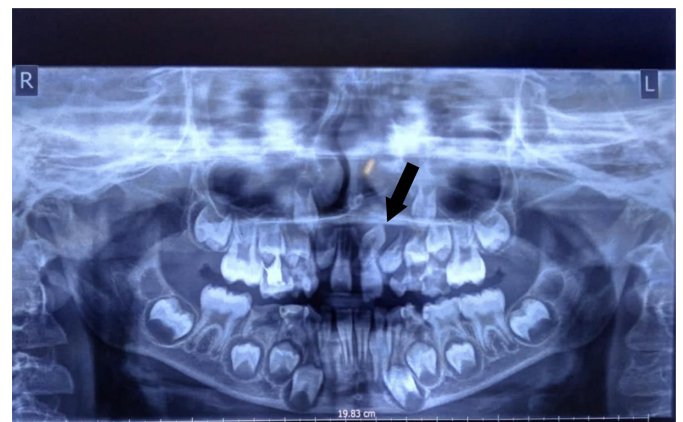


Fig. 2: Orthopantomogram(OPG) reveals impacted supernumerary tooth present palatal to 21

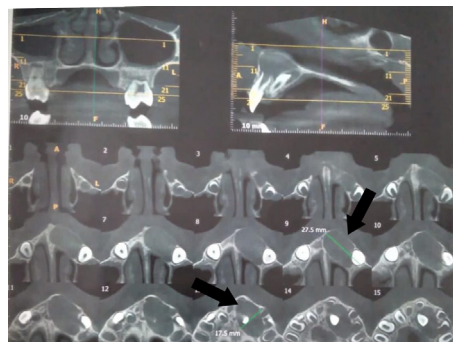


Fig. 3A: Cone beam Computed Tomography(CBCT) shows a well defined radiolucency measuring about 17.5*27.5mm(arrows) with corticated periphery associated with bicortical expansion



Fig. 3B: 3D reconstructed image of maxilla. Arrow indicating a well defined unilocular radiolucency associated with the impacted tooth



Fig. 4: Intraoperative view. A mucoperiosteal flap was raised with a labial approach

mine and Periodic Acid Schiff (PAS) staining respectively (Fig. 9A and 9B). A final diagnosis of GOC was made following the criteria established by Kaplan et al in 2008⁷. Patient showed no sign of recurrence 3 months post-surgery and is scheduled for regular follow up every 6 months.

DISCUSSION

Our case report describes a rare case of GOC associated with

an impacted supernumerary tooth mimicking a dentigerous cyst. This attribute was reported as "dentigerous relationship" (GOC-DR) by Fowler et al. in the year 2011.¹⁰

Since GOC has no pathognomonic radiographic feature, it can be confused with various other lesions associated with unilocular and multilocular radiolucencies. Clinical and radiographic differential diagnoses include dentigerous cyst, odontogenic keratocyst, ameloblastoma, radicular cyst, lateral periodontal cyst, myxoma,



Fig. 5: Grossing picture shows a greyish white mass along with tooth measuring about 2.4×1.6×0.4cm



Fig. 6: Stereomicroscopic image showing a thick cystic capsule that is attached to the neck of the tooth.

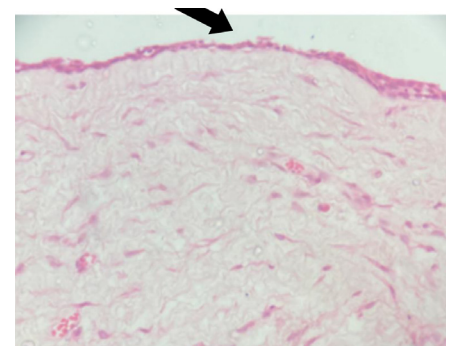


Fig. 7: Non keratinized stratified squamous epithelium made up of flat to cuboidal cells varying from 2-4 cell layers in thickness (hematoxylin and eosin stain, original magnification x400)

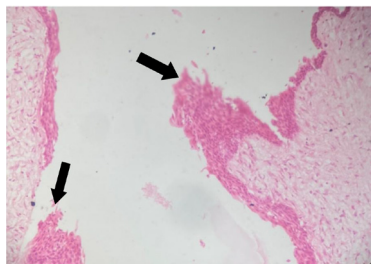


Fig. 8A: Epithelial plaque like thickenings (black arrows) (hematoxylin and eosin stain, original magnification x200)

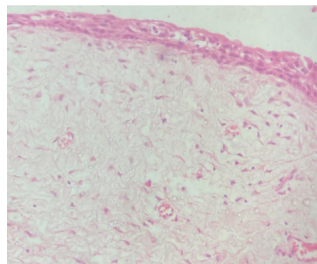


Fig. 8B: Numerous clear cells in the suprabasal and spinous layers of epithelium (black arrows) (hematoxylin and eosin stain, original magnification x400)

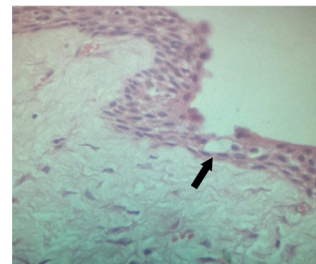


Fig. 8C: Intraepithelial microcyst (black arrow) (hematoxylin and eosin stain, original magnification x400)

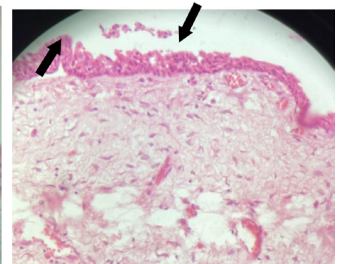


Fig. 8D: The superficial layer of the epithelium showed eosinophilic cuboidal cells, also called "hobnail cells" (black arrows) (hematoxylin and eosin stain, original magnification x400)

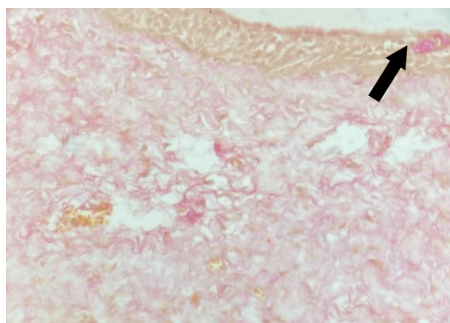


Fig. 9A: Mucicarmophilic mucous cell (black arrow) (mucicarmine stain, original magnification x400)

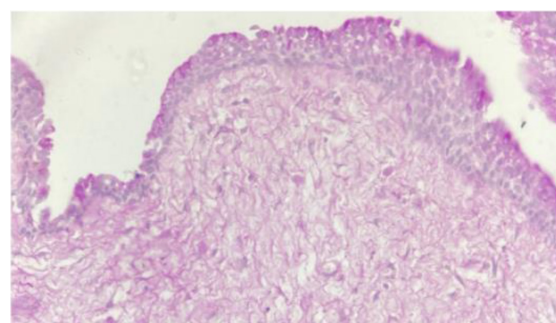


Fig. 9B: Glycogen rich cells showing wide Periodic Acid Schiff stain positivity in the superficial cell layer (magenta pink in colour) (PAS stain, original magnification x400)

central giant cell granuloma and fibrous dysplasia.¹¹ Hence identifying GOC clinically and radiographically is nearly impossible.

Histological diagnosis remains a challenge as well due to the rarity of the lesion and lack of any universally accepted criteria.

However, Kaplan et al., in 2008 have given major and minor diagnostic criteria (Table 1) which has been accommodating.

It is important to differentiate GOC histologically from lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC), central low grade mucoepidermoid carcinoma (MEC) and dentigerous cyst with mucous metaplasia.¹²

The lack of mucin producing cells and ciliated cells rules out the possibility of LPC and BOC. Although, dentigerous cyst with mucous metaplasia can be perplexing, absence of plaque like thickenings with intra-epithelial microcysts and other features helps to differentiate between the two cysts.

Central MEC is one of the main differential diagnoses of GOC and some authors including

Magnusson et al., have considered GOC as the benign variant of central MEC and that GOC has a very high potential to turn into a low grade mucoepidermoid carcinoma.^{13,14} However, absence of epidermoid and intermediate cells in the GOC lining helps in differentiating between the two lesions. Cytokeratin 7, 14, and 19 positivity in GOC has proved its odontogenic origin along with a negative reaction for epithelial membrane antigen (EMA) marker which shows positivity in MEC and other salivary gland lesions.¹²

Due to aggressive and unpredictable behavior of GOC, curettage and enucleation will not suffice, especially in large-sized lesions. Kaplan et al., in 2005 have deduced that the rate of recurrence is directly related to the size of the cyst. Small lesions had a recurrence rate of 14.4% whereas 85.6% of the larger lesions recurred. Hence large lesions should be biopsied to establish a definitive diagnosis before taking the treatment decision. Enucleation along with peripheral ostectomy reduces the chance of recurrence. If the lesion approaches adjacent vital structures, marsupialization can be done followed by enucleation.¹⁵

Around 50% of cases in the literature had a very short follow-up period, up to 2 years, while the average time for recurrence was 3 years.¹⁶

Therefore, follow-up is advocated for at least 3 years, and up to 7 years in aggressive cases.¹⁵

CONCLUSION

It is of utmost importance to place emphasis on the rarity of GOC and that it should not be missed due to its ambiguous clinical and radiographic features. Careful histological examination is essential along with necessary immunostaining to differentiate from central mucoepidermoid carcinoma. Extended follow-up of the cases is crucial due to high recurrence rates especially in aggressive cysts.

This report describes an unusual case of GOC associated with an unerupted tooth which clinically, radiographically mimicked a

dentigerous cyst. Nonetheless, diligent histopathological evaluation helped us to arrive at the accurate diagnosis.

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