



GLANDULAR ODONTOGENIC CYST OF THE MANDIBLE ASSOCIATED WITH IMPACTED TOOTH-REPORT OF A RARE CASE

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ABSTRACT

A glandular odontogenic cyst (GOC) is a rare odontogenic lesion with a distinct entity. It exhibits morphological similarities to other lesions both radiologically and histopathologically, making its diagnosis challenging. We report a rare case of GOC associated with the crown of an impacted mandibular third molar presenting as a unilocular osteodestructive lesion in a 49-year male. The diagnosis was made based on characteristic histopathological findings.

KEYWORDS: Glandular odontogenic cyst. Impacted tooth, Mandible, Mucous cells.

INTRODUCTION

Studies show the prevalence of cystic and neoplastic changes associated with impacted teeth ranging from 3 to 16%. Common pathological entities in intimate association with impacted tooth include dentigerous cyst, odontogenic keratocyst, calcifying odontogenic cyst, central mucoepidermoid carcinoma and unicystic ameloblastoma.^[1] Clinical diagnoses of such cases can be challenging and are to be substantiated by histopathology. Here we report an uncommon case of GOC associated with an unerupted third molar mimicking a dentigerous cyst.

Padayachee and Van Wyk described uncommon jawbone cysts of odontogenic origin with glandular components as "sialo-odontogenic cysts".^[2] Gardner et al. in 1988 established this cyst as a distinct entity terming it as glandular odontogenic cyst, which was classified as an odontogenic cyst by the WHO in 1992. Synonyms include "mucoepidermoid cyst" by Sadeghi et al and "polymorphous odontogenic cyst" by High et al.^[3] Though rare its aggressive behaviour and tendency to recur underlines its importance for an accurate diagnosis to decide on the treatment modality.

Clinically, GOCs are small and usually appear as an asymptomatic swelling,^[4] though a few cases have presented with pain and paresthesia.^[5] The most common site is the mandible, particularly the anterior region.^[6] Case reports have shown a general male predilection in the age group of 40-502 but a case series by Urs AB et al

has proved contrarily showing a strong female predilection and slightly more cases found in the maxilla compared to the mandible.^[7] Reported pediatric case of GOC in a 12-year-old proves GOC is an unpredictable cyst.^[8]

Radiographically GOC reveals a well-defined unilocular or multilocular radiolucency, often exhibiting scalloped margins and sclerotic borders. In addition, root resorption and tooth displacement with cortical perforation, leading to extension of the cyst into the adjacent soft tissues are also seen.^[2, 9, 10]

Histologically, it bears a resemblance to lateral periodontal cyst (LPC), botryoid odontogenic cysts (BOCs), radicular and residual cysts with mucous metaplasia, and low-grade mucoepidermoid carcinoma (MEC) making the diagnosis a challenge.^[11] Classically according to Fowler^[12] et al microscopic criteria of GOC include eosinophilic cuboidal (hobnail) cells, a feature not specific for GOC, but necessary for diagnosis. When considered individually, the presence of microcysts, clear cells, epithelial spheres, variable thicknesses, and multiple compartments appears to be most helpful in distinguishing GOCs from GOC mimickers. These features help distinguish GOCs associated with an unerupted tooth from dentigerous cysts with metaplastic changes. Fowler et al suggest the presence of 7 or more microscopic parameters as highly predictive of a diagnosis of "GOC".^[13]

The present paper aims to discuss the clinical, radiological, histopathological aspects, and differential diagnoses of GOC for correct diagnosis.

CASE REPORT

A 49-year-old male, with no medical history, reported to our institution with a complaint of pain in the lower back tooth region for a few days. On clinical examination, mandibular third molars were absent suspecting

impaction of the same. Panoramic radiographic evaluation revealed a radiolucent lesion in the posterior mandible region associated with an impacted right third molar, extending to the distal root of the second molar, measuring 10 x 18.8 mm and showing obliteration of the inferior orbital canal. (Fig 1) Clinically a provisional diagnosis of the dentigerous cyst was made. Informed consent was obtained and the lesion was carefully enucleated under local anaesthesia.



Fig 1: Panoramic radiograph showing impacted 38 with radiolucency.

Histopathological examination showed a large cystic cavity lined by the non-keratinized stratified squamous epithelium of varying thickness and areas of epithelial plaque formation. Superficial epithelial cells of the cystic lining were cuboidal to columnar referred to as “hobnail” cells with few of them showing cilia extensions. Epithelium showed numerous PAS-positive goblet cells (Fig 2) and frequent microcyst formation with few areas of papillary projection. (Fig 3) Several areas show

thinned-out epithelium resembling reduced enamel epithelium. Inductive changes were noticed in subepithelial areas. (Fig 4) Epithelium and connective tissue junction were flat with supporting connective tissue consisting of bundles of collagen fibres, numerous stellate and spindle-shaped fibroblasts with few blood capillaries. Correlating the above histopathological findings a final diagnosis of the glandular odontogenic cyst was made.

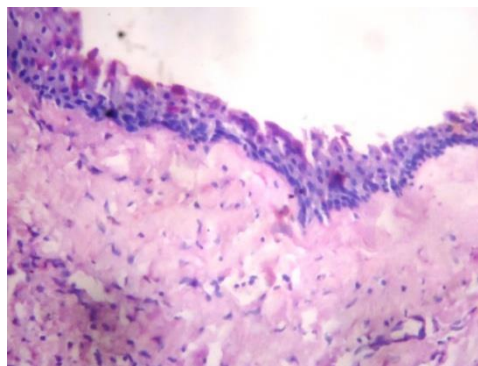


Fig 2: PAS staining showing PAS positive PAS-positive goblet cells (PAS staining, original magnification ×400)

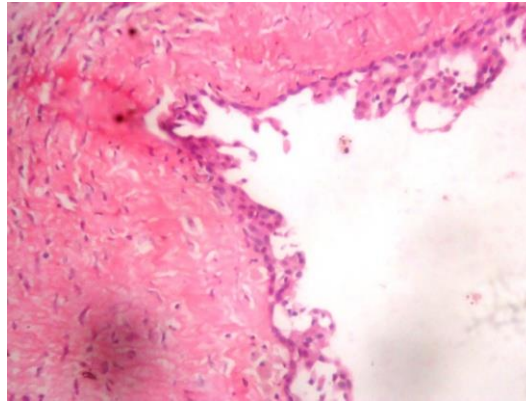


Fig 3: Cystic lining showing “hobnail” cells, microcyst formation and papillary projections. (H and E, original magnification ×200)

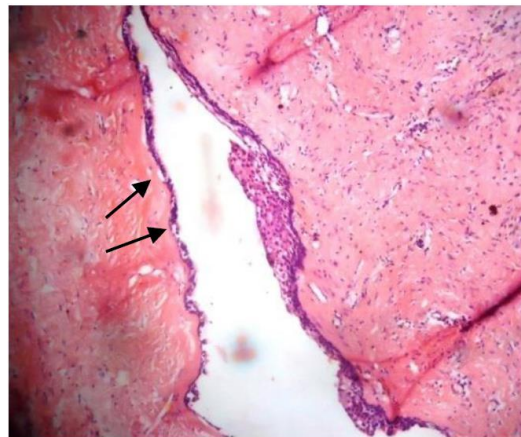


Fig 4: Cystic epithelium resembling reduced enamel epithelium and areas showing subepithelial inductive changes (Arrow in black). (H and E, original magnification ×200)

DISCUSSION

A glandular odontogenic cyst (GOC) is an extremely rare developmental odontogenic cyst of the jawbones exhibiting aggressive behaviour. The prevalence of the lesion ranges from 0.012% to 1.3% and accounts for 0.17% of all jaw cysts.^[13] The age at diagnosis of the cyst showed a range from the 5th to 7th decade. GOC has no gender predilection, and the mandible is the most common site for development, especially in the anterior region.

The histogenesis of GOC was initially suggested to develop from intraosseous salivary gland tissue.^[9] Recent studies evidence its origin from odontogenic epithelium rather than sialogenic tissue.^[14]

Our study reports an uncommon case of GOC associated with an unerupted mandibular third molar mimicking a dentigerous cyst. Very few cases have been documented and the characteristics are defined by Fowler *et al.*^[6] as a “dentigerous relationship”.

Most of the cases have been identified incidentally whereas our present case reported a complaint of pain and swelling. Unilocular radiolucency was seen in 76.9% of the cases^[2] as was in our case.

Studies suggest that a preoperative aspiration biopsy may help diagnose GOC. Aspiration fluids reported in the literature, include clear with low viscosity, creamy high-viscosity, and brownish-red liquids.^[4,15] Studies have also shown negative aspiration.^[2] However, in the present case aspiration was not done.

The treatment of choice is controversial ranging from enucleation, and curettage to En-bloc resection.^[16] Most of the GOC cases were treated conservatively and a recurrence rate of 30% has been reported.^[14] Our case was also treated conservatively with cyst enucleation.

The histopathological diagnosis of GOC has been a challenge because of the diversified features reported. Microscopic features include focal epithelial thickening, epithelial plaques, and glycogen-rich epithelial cells,^[17] which are also observed in botryoid and lateral periodontal cysts. The presence of ciliated epithelium and duct-like spaces with mucous cells and eosinophilic cuboidal cells located in the epithelial surface support the diagnosis of GOC.^[17,18] According to Fowler *et al.*^[13] the presence of microcysts, clear cells, and epithelial spheres may help distinguish GOC-DR from the dentigerous cyst with metaplastic changes. The same was confirmed in our histopathological examination.

To differentiate and distinguish low-grade MEC from GOC is mystifying due to strong histopathological similarities, especially the multicystic variant. Ciliated cells, superficial cuboidal cells, and intraepithelial microcyst or duct-like structures are not typical for low-grade MEC and help in differentiation.^[2] In addition Immunomarkers like MASPIN, Ki-67, and CKs 18 and 19 may be helpful to distinguish GOC from lowgrade MEC.^[14] Our case was as per the microscopic criteria suggested by Fowler et al.^[13] for diagnosing GOC.

The complexity of the induction effects within the odontogenic apparatus and the involvement of both ectodermal and mesodermal tissues are probably responsible for the bewildering variety of forms in which the odontogenic lesions exhibit. Studies by Hirshberg et al. have focused on the importance of epithelial-mesenchymal interactions in odontogenic lesions and the role of stroma in inductive and degenerative changes.^[19] Furthermore; it also suggests the biological behaviour of these lesions and reflects aggressiveness.

Follow-up is essential because of its aggressive biologic behaviour and propensity for recurrence.^[20] The aggressive pattern and high recurrence rate of GOC can be explained by the multilocular nature, presence of microcysts and the easy separation of thin epithelium from underlying connective tissue. The present case reported no incidence of recurrence in a periodic follow-up of 2 years.

CONCLUSION

In conclusion, GOC in association with an impacted tooth is a rare occurrence. It is important to consider both radiological and histopathological features, for accurate diagnosis of GOC to guide appropriate treatment.

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