Case Report

Glandular odontogenic cyst of the maxilla in a Nigerian: Case report and review of literature

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Abstract

Background: Glandular odontogenic cyst is a rare locally aggressive developmental odontogenic cyst that recurs with conservative management. It has been observed to commonly occur in the mandible with unilocular radiographic presentation.

Aim: To improve knowledge of glandular odontogenic cyst among pathologists and clinicians.

Methods: A case report and review of literature.

Results: We report the only case of recurrent maxillary glandular odontogenic cyst diagnosed in Lagos University Teaching Hospital over a period of 47 years. The progressively growing cyst occurred in a 33-year-old Nigerian male. The lesion recurred 10 months after initial surgical excision. A second surgical excision was performed, and the patient is being closely followed up.

Conclusion: Glandular odontogenic cyst is a rare developmental odontogenic cyst that has been reported to have a high recurrent rate. Detailed histopathologic assessment is required to arrive at a definitive diagnosis due to its similarities to other lesions. Segmental or marginal resection with long-term follow-up is advised to prevent recurrence.

Keywords: Cyst, glandular, Nigerian, odontogenic

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INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst that is associated with recurrence and termed aggressive.[1] The first two cases were reported by Padayachee and Van Wyk[2] in 1987. These lesions were observed to have features comparable with the botryoid odontogenic cyst (BOC) and therefore named sialo-odontogenic cyst due to suspected association with salivary glands. Gardner *et al.*[3] however, clearly identified GOC as a distinct clinicopathologic lesion in 1988. This was formally accepted by WHO in 1992 and classified as glandular odontogenic cyst/sialo-odontogenic cyst.[4]

GOC clinically presents as a painless, slow-growing swelling with a site predilection for the anterior mandible with a mandible–maxilla ratio of 3:1.[5,6] Although it presents among a wide age range, it commonly occurs in the middle age group and has a slight male gender predilection.[1,7] Radiographically, GOC usually presents

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as well-defined unilocular radiolucent lesion with scalloped margins;[5,6] although multilocular types have been reported.[8] It is believed to arise from remnants of the dental lamina[9] and typically presents as a cyst lined by nonkeratinising stratified squamous epithelium that consist of papillary structures (which project into the cystic lumen), nodular thickenings in form of plaques, mucous cells, intraepithelial gland-like structures, superficial eosinophilic columnar/cuboidal cells (Hobnail cells) flat epithelium-connective tissue interface and connective tissue without inflammation.[1,6,7] Treatment modalities that have been used for GOC include conservative treatment in the form of enucleation with curettage and surgical treatment in the form of surgical excision although more radical treatment in the form of marginal resection has been advocated due to propensity of recurrence after enucleation and curettage[1,8,9]

The rarity of the lesion requires more clinicopathologic information for its recognition by pathologists. Its observed aggressiveness and high rate of recurrence necessitate its prompt and accurate diagnosis for effective management. As far as we know no publications specifically on GOC among Nigerians exist in the English literature. We present a case of GOC with mineralisation in the maxilla in a 33-year-old Nigerian male which was initially surgically excised but recurred after 10 months, and in addition, we concisely review the scientific literature on the cyst, in an attempt to improve its knowledge among pathologists and clinicians.

CASE REPORT

A 33-year-old Nigerian male patient presented at the Dental Clinic of the Lagos University Teaching Hospital (LUTH) with a fluctuant painless swelling which the patient claimed had progressively increased in size. The swelling was located in the upper right anterior region of the maxilla. The tumor duration before hospital presentation was approximately 3 months. Radiographic examination revealed a well-defined multilocular radiolucent lesion. Incisional biopsy and histopathological examination were done. A diagnosis of multilocular neoplastic odontogenic cyst consistent with glandular odontogenic cyst was made by the oral and maxillofacial pathologist. Surgical excision was subsequently performed by the oral and maxillofacial surgeon, and the patient was followed up. The patient presented after 10 months with a second painless, fluctuant but progressively increasing anterior maxillary swelling of 4-week duration.

Intraoral examination revealed a bucco-palatal fluctuant swelling that extended from the area of the upper right first premolar to the upper right second molar. The mucosa overlying the swelling was however of normal color and appearance. Radiographically, the lesion was well defined, multilocular and contained few specks of radiopacities. Surgical excision under local anaesthesia was done and submitted for histopathologic evaluation.

Histopathology examination revealed a cyst lined by nonkeratinising epithelium of varying thickness made up of areas with focal proliferations and with papillary fronds that projected into the cystic lumen [Figure 1a and b]. The epithelial lining surface contained some cuboidal cells with somewhat large nucleus [Figure 2]. Microcysts, as well as duct-like structures, were also observed within the epithelial lining. The duct-like structure contained mucoid material in areas [Figure 3]. There were also epithelial lining surface areas that contained calcific materials [Figure 1a and b]. These calcific materials were observed to project into the cystic lumen [Figure 1a and b]. The epithelium-connective tissue interface of the cyst was flat, and the connective tissue wall consisted of endothelial lined vascular channels [Figure 4].

DISCUSSION

GOC is a rare and aggressive developmental odontogenic cyst with a relatively high rate of recurrence; it accounts for about 0.012%–1.3%[10,11] of all jaw cysts. We describe the clinicopathological features of the only case of GOC diagnosed in LUTH over a 47 years period. This accounts for 0.52% in the entire series of orofacial cysts over the same period. This further confirms its rarity. Perusal of the English literature shows a dearth of reports of cases of GOC among Nigerians. GOC has been reported to occur over a wide age range (14–75 years) with a mean age of 45.7 years, a slight male predilection and a male to female ratio of 1.3:1.[7,12] Males have been reported to be more affected in the second to the fourth decades while females predominate in the fifth decade.[5,7,13] The present case occurred in a 33-year-old male which is a pattern of occurrence consistent with previous reports.

Clinically, GOC presents as a slow growing asymptomatic intraosseous swelling with a site predilection for the anterior mandible. Review from large series of GOC by Mohammed Faisal *et al*.[14] and MacDonald-Jankowski[5] show a definite mandibular site predilection with anterior mandible as the most common specific area affected. Cases of bilateral lesions have in addition been reported in the literature.[14] Although the present case occurred in the anterior maxilla, larger sample studies are needed to determine the site predilection among Nigerians. The common radiographic presentation of GOC is a well-defined unilocular radiolucency that sometimes

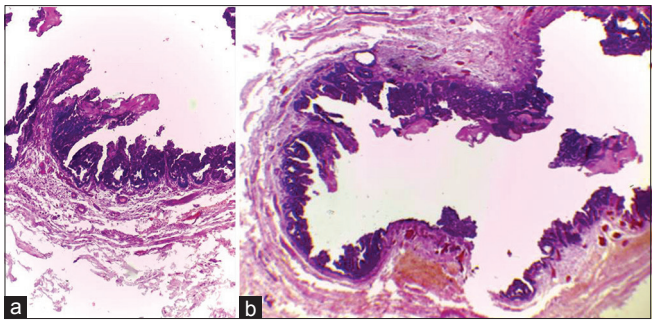

Figure 1: (a) A cyst lined by nonkeratinising epithelium of varying thickness made up of areas with focal proliferations and with papillary fronds that projected into the cystic lumen. Also seen are intraepithelial pseudocystic structure and areas of calcification projecting into the cystic lumen (H and E, ×10). (b) The presence of papillary fronds and calcified structures projecting into the cystic lumen (H and E, ×10)

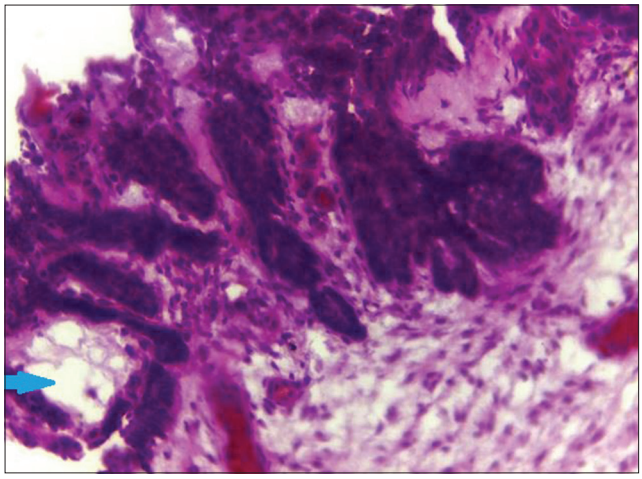

Figure 3: The presence of intraepithelial glandular-like structures containing mucous pools (H and E, ×40)

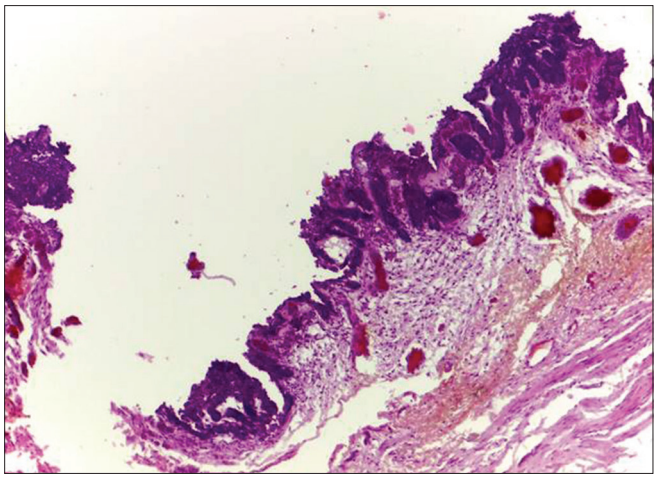

Figure 2: Epithelial surface layer with columnar to cuboidal cells (H and E, ×40)

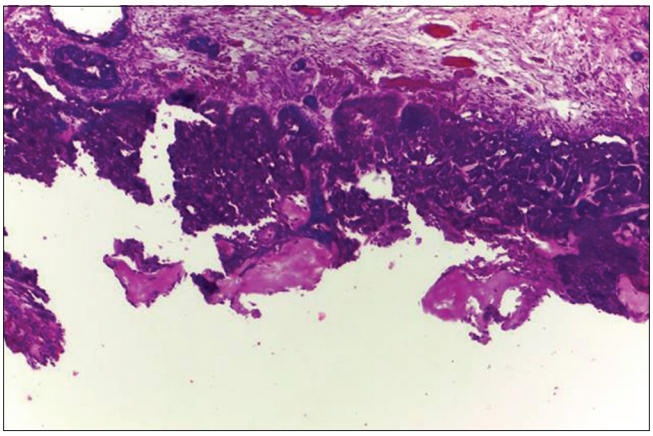

Figure 4: Papillary projections into cystic lumen and flat epithelium-connective tissue interface (H and E, ×10)

occur with buccolingual expansion although lesions with multilocular radiolucency has been reported to occur.[1,5,15,16] Other less frequent presentations are radiolucency's with sclerotic border and scalloping, tooth displacement, root resorption and cortical perforation.[3,6,7] As the clinical and radiographic features of GOC vary considerably, these are not distinctive for a definitive diagnosis.

Histopathological examination is the gold standard for definitive diagnosis of GOC. Kaplan *et al*.[7] stated that to make a diagnosis of GOC, certain criteria must be fulfilled. These criteria have been categorised into major and minor criteria. The major criteria consist of the presence of the following features: (a) non-keratinised squamous epithelial lining with a flat epithelial–connective tissue interface. The epithelial lining should exhibit surface cuboidal 'hob-nail' cells, (b) focal luminal proliferations and (c) intraepithelial structures in the form of glandular microcystic or pseudoglandular structures, mucous cells and intraepithelial mucous pools with or without crypts lined by mucous producing cells. The minor criteria proposed by Kaplan *et al*.[7] consist of the histological presence of the following features: (a) papillary proliferation, (b) Ciliated cells, (c) multicystic/multiluminal architecture of the cyst and (d) clear/vacuolated cells in basal or spinous layer of cystic epithelium. Kaplan *et al*.[7] however, further stated that while the minor criteria may be absent, the focal presence of each of the major criteria is compulsory for definitive diagnosis of GOC. Although the histologic characteristics of the present case are consistent and similar to previous reports,[1,6,8,9] the present case in addition presented with multiple areas of mineralisation/calcification, which is similar to a report by Shah *et al*.[1] where they reported a case of GOC with hyalinised areas suggestive of dentinoid.

Differential diagnosis of GOC consists of lateral periodontal cyst (LPC), BOC and low-grade central mucoepidermoid carcinoma (CMEC).[4,8,17] Although LPC and BOC are histologically similar to GOC, the presence of

intraepithelial structures in form of glandular microcystic spaces and mucous cells which occur in GOC and are absent in LPC/BOC clearly differentiates the lesions.[1,8] Immunohistochemical studies have in addition been used to differentiate GOC from LPC. Pires *et al*.[18] compared GOC and LPC and demonstrated positivity of cytokeratin 18 and 19 with GOC and negative reaction with LPC.

Histopathologic features of the multicystic variant of GOC may mimic a low-grade CMEC making diagnosis challenging and affecting management. The histological presence of, intraepithelial glandular microcystic spaces, superficial epithelial hobnail cells, luminal papillary projections and ciliated cells are distinctive features of GOC and not CMEC.[17] Immunohistological studies have been done to distinguish GOC and low-grade CMEC, and this showed decreased p-53 positivity and increased Ki-67 index for GOC when compared to low-grade CMEC.[19] The aggressive biologic nature and recurrence of GOC have been studied by various scientists.[19,20]

Increased expression of Bcl-2 and Ki-67 index in GOC suggest increased activity and proliferation of cystic lining which may be accountable for the biologic aggressiveness. Enucleation of GOC has been reported to result in high recurrent rate.[7,8,17,21] GOC like keratocystic odontogenic tumor presents with similar unique histologic features in the cystic wall which may make management challenging. Presence of structures such as microcysts, thin epithelial lining in areas, and flat epithelium-connective tissue interface which occur in both lesions have been suggested to be responsible for incomplete removal, especially during enucleation leading to subsequent recurrence.

Recurrence rates as high as of 50.0% and 55.0% have been reported after enucleation of GOC.[8,17,19] It is, however, worth nothing that perhaps some cases considered to have recurred post-enucleation may not be actual recurrence of lesions but relapse due to initial inadequate treatment. The present case was surgically excised and recurred after a period of 10 months. The patient has been followed up for 8 months. Surgeons advocate a more aggressive approach in form of segmental or marginal resection to reduce recurrences. Patients, in addition, must be closely followed up for a long period.

GOC is a rare developmental odontogenic cyst that has been reported to have a high recurrent rate. Detailed histopathologic assessment is required to arrive at a definitive diagnosis due to its similarities to other lesions.[4,8,17] Segmental or marginal resection with long-term follow-up is advised to prevent recurrence.

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Conflicts of interest

There are no conflicts of interest.

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