Glandular Odontogenic Cysts: A Case Report of a Classical Presentation

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ABSTRACT

Background: Glandular odontogenic cysts (GOCs) are rare intrabony unilocular or multilocular pathologically lined cavities of odontogenic origin. It was updated with new diagnostic criteria in the 2017 WHO classification of odontogenic tumours. It has a high tendency for recurrence and can be confused histologically for intraosseous muco-epidermoid carcinoma.
Objective: To report a 10-year history of recurrent mandibular swelling in a patient who has had three surgeries.
Case Presentation: This report describes a case of GOC in the body and symphyseal region of the mandible of a 56-year-old male which is typical in history and clinical examination.
Conclusion: GOC is a rare and aggressive lesion with a relatively high recurrence rate and the need for a careful clinical and radiological evaluation along with a precise histologic examination cannot be overemphasized

Keywords: Odontogenic Cysts, Multiple Reoccurrence, Diagnostic Criteria.

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INTRODUCTION

The glandular odontogenic cyst is a relatively uncommon and newly recognized type of developmental odontogenic cyst that manifests aggressive behavior. Despite its odontogenic origin (rest cells of dental lamina),[1] it also shows glandular or salivary features which can be traced to the fact that the odontogenic epithelium is pluripotential.[2]

It was first described in 1987 by Padayachee and Von Wyk with several synonyms like "sialo-odontogenic cyst", "muco-epidermoid odontogenic cyst" and "mucous-producing odontogenic cyst".[1,2,3] However, in 1988, the name "glandular odontogenic cyst" was proposed by Gardner et al and this was followed by its recognition by the World Health Organization (WHO) in 1992 making it a clinical entity.[3,4]

GOCs tend to occur in the middle-aged though it has a wide age range of 10-90 years with a mean age of 49.5 year.[4] It rarely occurs before the age of 20 and almost 80-85% of cases were said to occur in the mandible showing a strong predilection for the anterior region of the jaws.[4,5] Mandibular lesions tend to cross the midline and can range in size from small to very large irregular expansile masses that can result in pain and parasthesia but it generally has a slow growth rate and is usually asymptomatic.[2,6,17]

The common radiological finding is that of a unilocular or multilocular radiolucency with well-defined margins and a sclerotic rim.[5] Recurrent lesions are usually multilocular. There could be associated resorption of adjacent tooth/roots.

Histologically, the cyst is lined by squamous epithelium, the epithelial and connective tissue interface is usually flat with a cystic wall that is devoid of inflammatory cell infiltrates.[7, 15] There could be 2cilia occasionally and pools of mucin-aminophilic materials. Goblet or mucin producing cells are also visible.[3,7]

Clinical differential diagnosis includes the lateral periodontal cyst which is often considered as its variant because it occurs in the same location and odontogenic keratocyst which it resembles in terms of behaviour. Radiographically, the lesion resembles a unicystic ameloblastoma, odontogenic myxoma and central giant cell granuloma. Histological differentials include dentigerous cyst, radicular cyst, surgical ciliated cyst, botyroid odontogenic cyst and more importantly the intraosseous muco-epidermoid carcinoma.[1,4,6,7]

Treatment options include enucleation, curettage and sometimes marginal or segmental resection due to its high reoccurrence rate. Patients usually require long term follow ups.[8,9]

CASE REPORT

A 56-year-old man who presented at the outpatient clinic of the Oral and Maxillofacial Surgery Department of our facility (Federal Teaching Hospital, Gombe) with a 10-year history of recurrent mandibular swelling for which he has had surgery 3 times. Swelling was asymptomatic localized to the anterior aspect of the mandible with associated buccal expansion and "egg-shell" thinning of the bony buccal cortex as well as obliteration of the sulcus from tooth 35 to tooth 46.

Posterior-Anterior radiographic views of the jaws including right and left oblique laterals were requested and the report showed an expansile "soap bubble" lesion involving the mandible. There is associated erosion of the adjacent cortex with blunting of the adjoining roots of teeth, no calcifications are seen within it; however, an overlying soft tissue swelling is seen. Overall appearance was said to be consistent with ameloblastoma with dentigerous cyst as a possible differential.

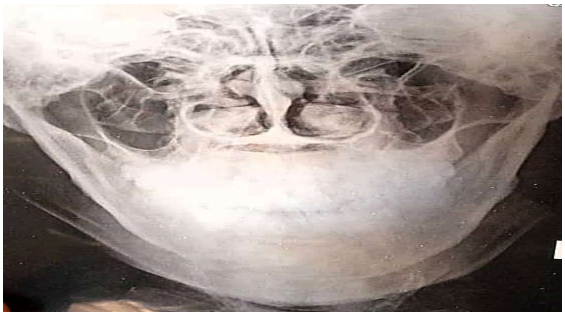

Figure 1: Showing PA jaws with a well-defined multilocular radiolucency with resorption of adjacent teeth.

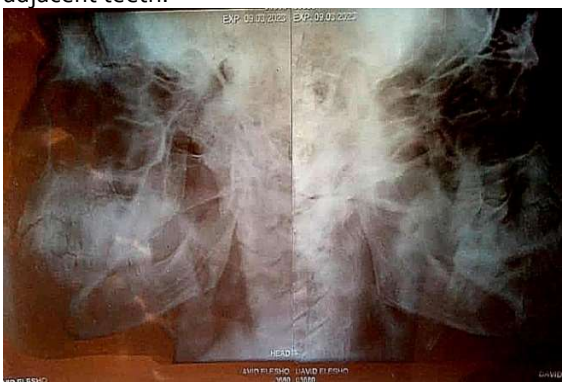

Figure 2: Showing *left and right oblique laterals with a well-defined multilocular radiolucency with resorption of adjacent teeth.*

Aspiration of the mass yielded a brown-coloured fluid that was odourless and produced no shimmering effect. Based on these clinical and radiologic findings, a provisional diagnosis of unicystic ameloblastoma was made and due to the multiple recurrences as suggested in the patient's history, the plan was to do a segmental resection with anterior iliac crest reconstruction.

All pre-operative investigations were performed and the patient was subsequently operated under general anaesthesia via nasotracheal intubation. The pathologic specimen obtained was soft in consistency, creamish-brown in colour with irregular borders alongside associated teeth (12) and a margin of normal bone on both ends.

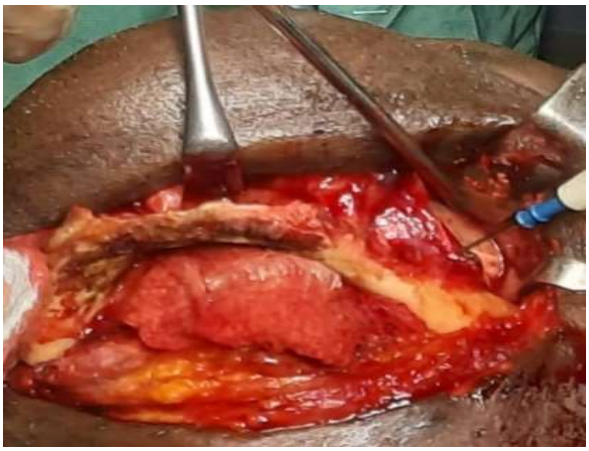

Figure 3: cystic lesion exposed with a margin of normal bone.

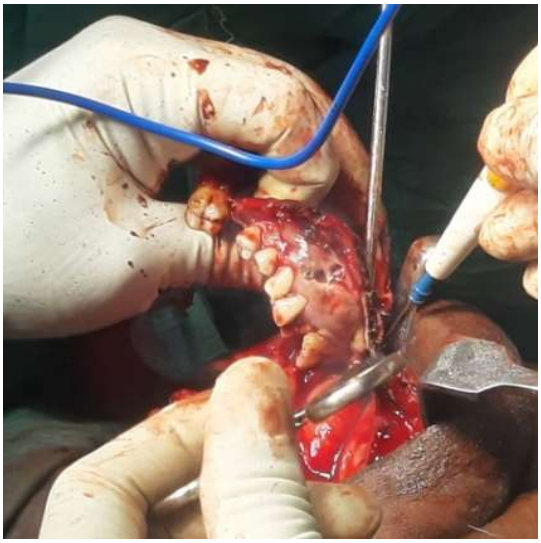

Figure 4: resected mandible with cystic lesion.

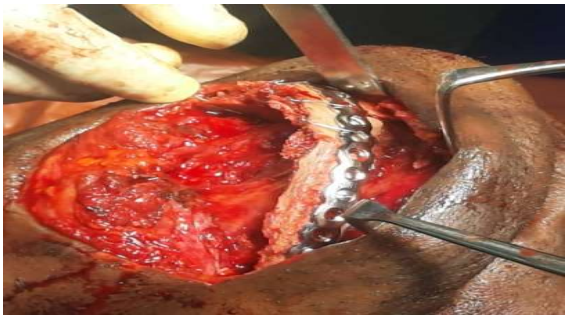

Figure 5: bone graft firmly immobilized with reconstruction plates and screws.

The histological examination of the specimen revealed a multicystic fibrocollagenous cyst walls lined by non-keratinized stratified squamous and partly basaloid epithelium within which are several microcysts, duct-like paces and few mucous cells. Few squamous spherules and "hobnail" cells are also present, with overall features consistent with glandular odontogenic cyst.

The patient has been followed up for 9 months and thus far, there is no evidence to suggest recurrence.

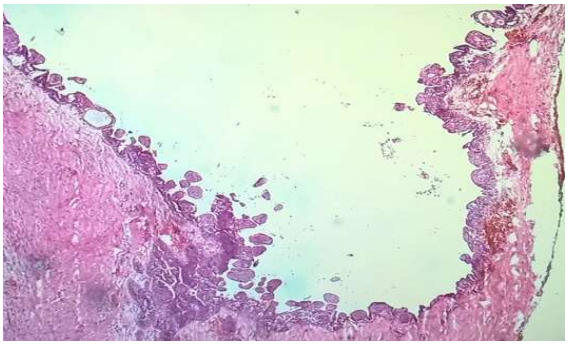

Figure 6: fibro collagenous cystic wall lined by non-keratinized epithelium.

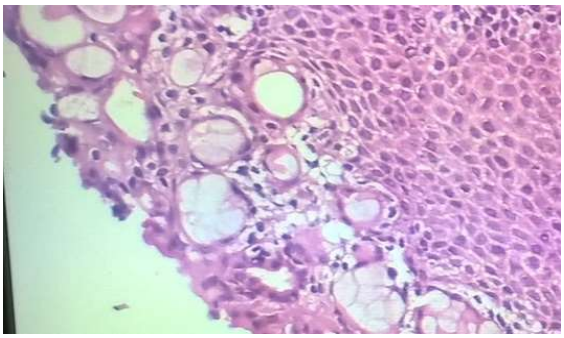

Figure 7: Microcysts, squamous spherules and hobnail cells.

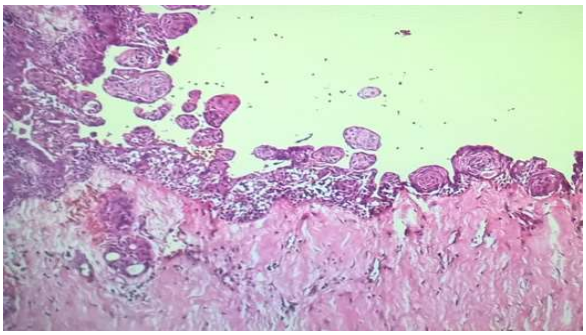

Figure 8:-papillary proliferation of the lining epithelium.

DISCUSSION

Glandular odontogenic cysts are a relatively new clinical entity which was previously referred to as "sialo-odontogenic cyst due to the presence histologically of mucous cells, mucin pools and eosinophilic cuboidal-shaped cells that resemble salivary gland ducts.

The name glandular odontogenic cyst was finally agreed upon in 1988 by Gardner et al when the presence of mucin elements lined by odontogenic epithelium was confirmed.[2,8] The lesion was subsequently adopted by the WHO in 1992 as a separate clinical entity where it was classified as a developmental odontogenic cyst.[8,10]

The GOC remain one of the rarest odontogenic cysts with incidence as low as 0.12%, about 39 cases have been reported in English literature, 23 in Brazil, 4 in Germany (and an additional 7 in 10 years) and 1 case reported in Lagos, Nigeria. In fact, as at 2015, only a total of 181 cases have been reported.[6,11]

This is the first case to be reported in this center and it involved the anterior mandible in a patient aged 56. This finding concurs with most published studies where the majority of GOCs (80-87%) were reported to occur in the anterior mandible of middle-aged with a peak incidence in the 6th decade. Many studies also quoted a slight male predominance.[5,8,20]

The diagnosis of GOCs was previously based on clinical, radiological and histopathologic criteria described by Gardner et al in 1988 but recently Brannon and Kaplan published a set of major and minor criteria that was adopted by the WHO in 2017.[6,11,12]

Major Criteria

1) Squamous epithelial lining with a flat interface with the connective tissue walls.
2) Epithelium exhibiting variable thickness along the cystic lining.
3) Cuboidal eosinophilic cells (hobnail cells)
4) Mucous (goblet) cells with intraepithelial mucus pools
5) Intraepithelial glandular microcystic or duct-like structures.

Minor Criteria

1) Papillary proliferation of lining epithelium
2) Ciliated cells
3) Multicystic /multiluminal architecture
4) Clear cells in basal or spinous layers

The presence of at least 7 of these criteria according to Brannon and Kaplan is sufficient to make a diagnosis of GOC.[12,19]

The multilocularity in GOC when compared to other odontogenic cysts and its tendency to cause expansion of the cortical bone underline its aggressive potential.[14] These aspects were observed in the case reported herein as the patient before presentation had repeated surgeries albeit under local anaesthesia. Our case was considered to be GOC because it fulfilled all the criteria listed by Brannon and Kaplan. GOC recognition based on clinical and radiological evaluation alone is practically impossible, [5,11] a fact that authors of all previous publications have stressed harmoniously. Only histopathological examination allows for a certain diagnosis of the condition.[15,18]

The most important differential diagnosis worthy of consideration is intraosseous muco-epidermoid carcinoma due to its similar histopathologic features. However, GOCs are constantly negative for MAML2 gene arrangements that are present in intraosseous muco-epidermoid carcinoma; also absent were cellular atypia and solid epithelial proliferations.[13,17]

The only feature that has not been reported in intraosseous muco-epidermoid carcinoma that may justify the existence of GOC as a separate clinical entity is the occasional presence of epithelial plaques similar to those seen in lateral periodontal cysts. However, unlike lateral periodontal and botyroid odontogenic cysts that are innocuous, GOC is regarded as considerably aggressive.[5,16]

Several molecular markers have been investigated as new tools to diagnose GOC and differentiate it from other lesions. These include CK19, Ki67, MASPIN (mammary serine protease inhibitors) and EMA (epithelial membrane antigen).[12, 13]

The treatment options for GOC, range from conservative approaches like enucleation, marsupialization, and curettage with adjuvant Carnoy's solution to marginal and segmental resection.[9, 11]

Segmental resection is preferred by a few authors due to the cyst's tendency to recur after conservative treatment.

CONCLUSION

GOC is a rare and aggressive lesion with a relatively high recurrence rate. Hence, a careful clinical and radiological evaluation along with a precise histologic examination must be performed.

Immuno-histochemical investigations when available are also recommended for diagnosing GOC because they provide distinctive information about the lesion that can aid in distinguishing it from its differentials.

Source of Support

Nil

Conflict of Interest

None declared

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