ORIGINAL RESEARCH PAPER
Volume - 12 | Issue - 04 | April - 2023 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

GLANDULAR ODONTOGENIC CYST, A RARE ENIGMA – A CASE REPORT & LITERATURE REVIEW

Maxillofacial Surgery

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ABSTRACT

The glandular odontogenic cyst (GOC) is a rare cyst that was first described in 1988 as a distinct entity. This lesion can occur in either jaw and is a rare developmental odontogenic cyst that resembles several other odontogenic lesions. However, GOCs have non-specific clinical and radiographic findings, and diagnosing them can be very difficult given their rarity. Furthermore, GOCs may be difficult to diagnose because they have overlapping histologic features with other intraosseous lesions, such as the lateral periodontal cyst and its botryoid variant, and the dentigerous cyst with metaplastic changes. In this study, we report a new case of GOC in a 42-year-old female patient and review previous cases reported in the literature. We discuss the different kinds of features which will help a clinician distinguish GOC from other lesions.

KEYWORDS

Glandular Odontogenic Cyst, Odontogenic Cyst, Mandible

INTRODUCTION

The glandular odontogenic cyst (GOC) was first described by Padayachee and Van Wyk in 1987.[1] In the beginning, the authors coined the term sialo-odontogenic cyst for the lesions. A year later, Gardner et al. (1988) reported eight additional cases of the lesion and proposed the term "glandular odontogenic cyst," which is still used today.[2] The GOC is a developmental cyst with epithelial features that simulate salivary gland or glandular differentiation, according to the most recent classification of head and neck tumors by the WHO (Speight et al., 2017).[3] Although considered rare, it is crucial to correctly diagnose this lesion, as it presents with non-specific clinical and radiographic findings, is more aggressive, requires more radical surgery, and has higher chances of recurrence compared to other odontogenic cysts.

GOCs often affect patients in their fifth to seventh decades of life, with a slight male predilection. They typically appear as asymptomatic swellings, frequently located in the anterior mandible and often crossing the midline.[4] Radiologically, they appear as well-demarcated unilocular, or less often multilocular, radiolucent lesions. Bony expansion is common, whereas tooth displacement, root resorption, and cortical perforation are infrequent findings.[4,5,6]

Diagnosing GOCs can be a challenge because they share histologic features with other intraosseous lesions, including the lateral periodontal cyst and its botryoid variant, and the dentigerous cyst with metaplastic changes. To distinguish GOCs from other entities with similar appearance, Fowler et al. proposed a set of 10 histologic criteria.[7]

Glandular Odontogenic Cysts are typically treated with conservative surgical enucleation, but this approach often results in a high recurrence rate. Recurrence rates of 21% to 50% have been reported, with a mean recurrence period of 62 months.[4,7,8] However, recurrence rates may be underestimated since many cases in the literature only report a 24-month follow-up.[9] Kaplan et al. recommend enucleation for small unilocular GOCs and peripheral ostectomy/marginal resection for larger multilocular lesions.[8] Due to the high recurrence rate, confirmed GOC cases require a 3- to 7-year follow-up period.[8]

Partial information regarding the clinical and radiologic features of GOCs is available in the literature. Therefore, epidemiological studies of rare lesions like GOCs are important for improving diagnostic accuracy and refining treatment plans, ultimately leading to better clinical outcomes.

The aim of this study is to present a case report of a middle-aged woman and to conduct a comprehensive comparative analysis of the clinical, radiologic, and histopathological features of GOCs based on available literature.

CASE REPORT

A 42-year-old female patient presented to the Department of Oral and Maxillofacial Surgery at our institute with complaints of swelling on the right side of her jaw and pain in the posterior region of the jaw, specifically in the area of the lower right first and second molars, for the past three months. The symptoms had significantly worsened in the previous month. The patient had no significant medical history and reported no history of trauma to the region or any deleterious habits. The patient gave a history of 48 extraction.

During extra-oral examination, a firm swelling was palpated in the right mandibular angle region. The overlying skin was intact and showed no signs of discoloration. No extra-oral discharging sinus or bleeding was observed. (Figure 1) The intraoral examination revealed swelling in the posterior mandible region with respect to teeth 46 and 47. This was accompanied by bucco-lingual cortical expansion. The lesion was tender upon palpation and exhibited crepitus in some regions. Signs of fluctuation were also present. Overlying mucosa was intact and there were no signs of infection or sensory loss. (Figure 2)

The panoramic radiograph revealed a well-defined, unilocular, radiolucent lesion extending from the distal aspect of 47 to the sigmoid notch laterally. The condyle and coronoid processes were unaffected. (Figure 3) The lesion extended over the mandibular canal and there was erosion of the lingual cortex with resultant thinning. Provisional diagnoses of odontogenic keratocyst, residual cyst, unicystic ameloblastoma and central giant cell lesion were given. The result of aspiration was a serous, brownish-red fluid.

Under general anaesthesia, the lesion was enucleated in one big piece and few small fragments followed by application of Carnoy's solution and peripheral ostectomy. (Figure 4). The teeth 46 and 47 were extracted. An intraoral vestibular incision was given and the buccal

cortical bone was removed. Careful curettage was done to avoid damage to the inferior alveolar nerve. The overlying mucosa was also excised.

Figure 1: Extra-oral Facet Of The Patient.

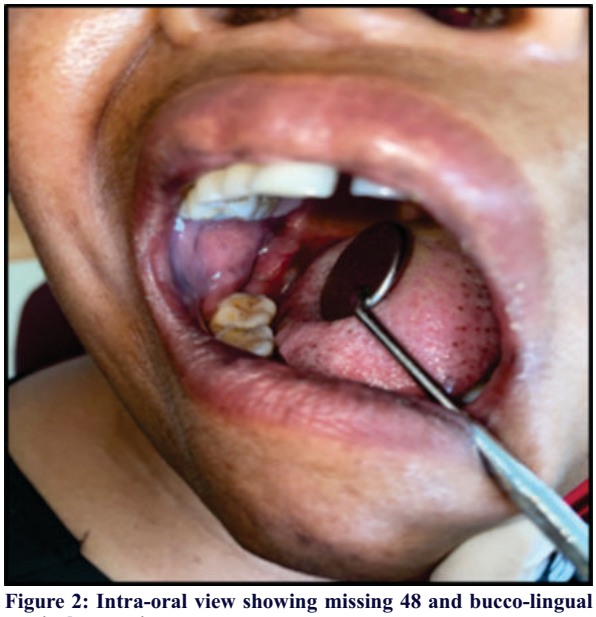

Figure 2: Intra-oral view showing missing 48 and bucco-lingual cortical expansion.

The cavity was kept open and Bismuth Iodoform Paraffin Paste (BIPP) gauze was placed which was changed every 1 week for the next 2 months.

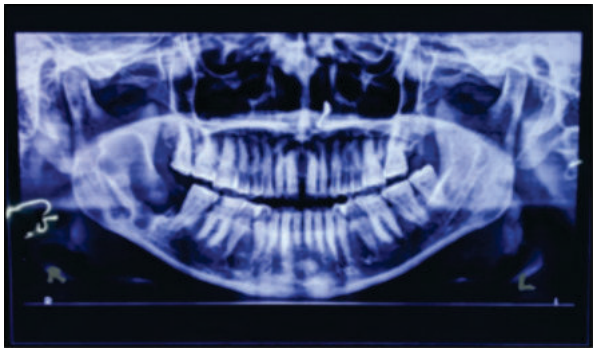

Figure 3: Panoramic Radiograph

On macroscopic examination, there was one fragment of tissue 2 cm x 1.5 cm in dimension, brownish in colour and multiple bits of cystic fragment. The surgical specimen was sent for histological examination which confirmed the diagnosis of GOC. Nine months following the surgical procedure, a postoperative panoramic radiograph was taken, which revealed evidence of bone healing and complete resolution.

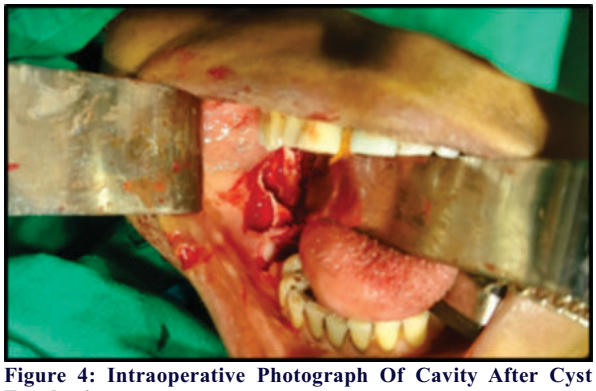

Figure 4: Intraoperative Photograph Of Cavity After Cyst Enucleation.

Kaplan et al. divided the microscopic characteristic of GOC into major and minor criteria, with the purpose of facilitating the diagnosis. (Figure 5, Figure 6, Figure 7)

1. Squamous epithelial lining, with a flat interface with the connective tissue wall.
2. Epithelium exhibiting variations in thickness along the cystic lining with focal luminal proliferation.
3. Cuboidal eosinophilic cells or "hob-nail" cells.
4. Mucous cells with intraepithelial mucous pools, with or without crypts lined by mucous-producing cells.
5. Intraepithelial glandular, microcystic or duct-like structures.
6. Papillary proliferation of the lining epithelium.
7. Ciliated cells.
8. Multicystic or multiluminal architecture.
9. Clear or vacuolated cells in the basal or spinous layers.

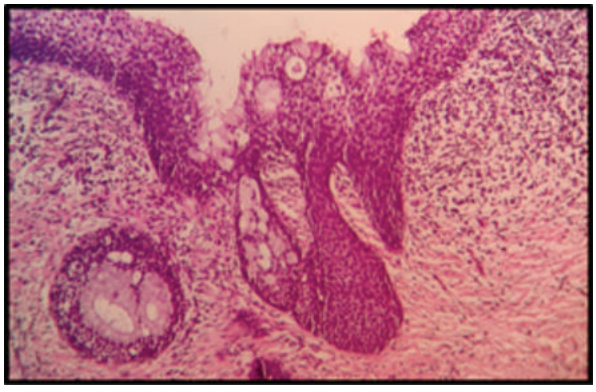

Figure 5: A. Epithelium Exhibiting Variations In Thickness Along The Cystic Lining With Focal Luminal Proliferation.

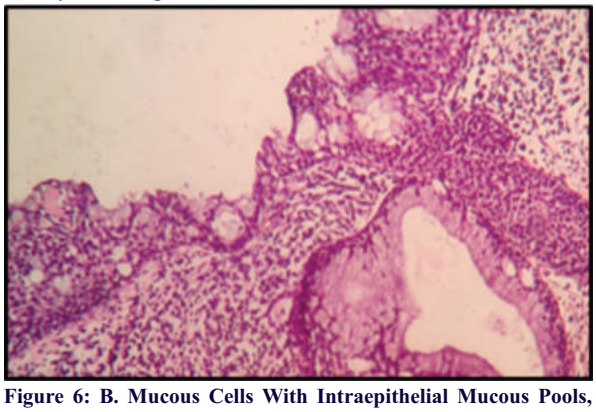

Figure 6: B. Mucous Cells With Intraepithelial Mucous Pools, With Or Without Crypts Lined By Mucous-producing Cells. C. Clear Or Vacuolated Cells In The Basal Or Spinous Layers. D. Intraepithelial Glandular, Microcystic Or Duct-like Structures.

DISCUSSION

GOC makes up just 0.2% of all odontogenic cysts, making it a very rare condition.[10] Less than 250 instances have been documented in the literature [4,11,12,13]; the biggest single research sample was 46 individuals.[7] This study presents a case of GOC in a 42-year-old female and reviews the relevant literature to clarify the key elements of a reliable diagnosis.

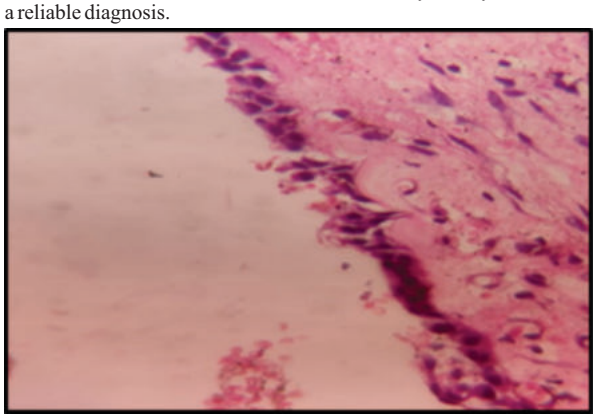

Figure 7: E. Cuboidal Eosinophilic Cells Or "hob-nail" Cells.

Demographics & Clinical Features

GOCs commonly affect patients during the fifth to seventh decades of life (mean age of 48 years), with most cases presenting after 30 years of age.[14] The gender distribution of GOCs is roughly evenly split, with a slight male predilection.[4,7]

GOCs may manifest as asymptomatic swellings or may be accidentally found on radiographic examination.[4,15] There have been a few isolated cases described in the literature that manifest with pain and paresthesia. Just one instance from each nation had tooth movement or paresthesia as the initial presenting feature, highlighting how uncommon these findings are. [14]

In the current study the age of the patient is 42 and is female who presented with pain as the main symptom along with swelling.

Location

GOCs exhibit a preference for the mandible especially the anterior mandibular region, and they frequently cross the midline.[4,5,7,8] 36% of GOCs were found in the anterior/premolar area of the mandible, according to a study of the literature.[4]

The GOC reported here is located in the posterior right mandibular region stretching from the mandibular molar area to the ramus in the current investigation. Few earlier studies have reported this location which is confirmed in this location. [4,5,6]

Radiologic Features

The anterior mandible often has well-defined uni- or multilocular radiolucency that span the midline as characteristic radiologic markers of GOCs. Although it seems that GOC borders are clearly defined, there have been a few isolated instances of ill-defined borders documented. GOCs manifest radiologically as unilocular or, less frequently, multilocular radiolucencies, with some studies demonstrating comparable rates of uni- and multilocularity. A unilocular radiolucency that extends laterally into the posterior ramus is described in the current case.

The current investigation found evidence of cortical enlargement linked to GOCs, which is a frequent radiologic finding. A review of the literature revealed that 86% of GOCs caused cortical thinning or perforation, demonstrating the aggressive propensity of the condition.[9]

The odontogenic keratocyst, radicular cyst, ameloblastoma, nasopalatine duct cyst, and dentigerous cyst are among the radiologic differential diagnoses for GOC.[15]

Surgical Management

In 122 cases, the treatment for the lesions was known; 108 of these cases involved conservative surgery (27 curettages, 81 enucleations), and 12 had marginal or segmental resection. 21 (21.6%) of them recurred (4 curettages, 16 enucleations, and 1 marginal resection). Two of the enucleated cases - Cano et al. (2012) and Motooka et al. (2015) - were previously subjected to marsupialization and were thus included in this analysis as examples of "enucleation." Neither case has recurred.[4]

Before curettage or enucleation, five cases were exposed to Carnoy's solution (Bhat et al., 2014; Qin et al., 2005), all of which had no recurrences. These cases were all taken into consideration when determining whether to do a curettage or an enucleation.[4]

After enucleation, three cases were submitted for peripheral osteotomy (Lee et al., 2014; Lin et al., 2000; Reyes et al., 2008); one case had no recurrence, while the other two lacked any information on the status of the patients.[4]

Considering the past literature and the age of the patient, we chose the treatment modality of enucleation followed by peripheral ostectomy and chemical treatment with carnoy's solution.

CONCLUSION

GOCs often affect adults in their fifth to seventh decades of life and present as asymptomatic swellings that usually involve the mandible. From benign cysts with identifiable GOC-like features to more aggressive tumours, they may display a wide variety of clinical and radiologic features. Common observations were cortical growth and loss of cortical integrity, either through thinning or disintegration. Paresthesia and clinical symptoms of pain appear to be key indicators for differentiating GOCs from central MECs.

The patient in the current research presented with a well-defined unilocular radiolucent lesion with pain as their primary complaint. Despite some similarities in the radiological findings, this case does not fall within the typical range of GOCs. Even if it's an uncommon occurrence, one shouldn't rule it out as an option for an oral and maxillofacial surgeon or a dental practitioner to confront. As such, the potential of it should not be discounted and it should be kept as a differential diagnosis.

Consent: Written consent was taken from patient for this publication.

Financial support and sponsorship: Nil.

Conflicts of interest: The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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