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CASE REPORT

A Rare Case of Glandular Odontogenic Cyst Successfully Treated with Surgical Resection

Nimra Iqbal[1], Sidra Fahim[1], Saeedullah Shah[1,2]

1. Department of Oral Medicine, Altamash Institute of Dental Medicine, Karachi, Pakistan.
2. Department of Oral Surgery, Altamash Institute of Dental Medicine, Karachi, Pakistan.

ABSTRACT

A glandular odontogenic cyst (GOC) is a rare developmental cyst of the jaw with the potential for aggressive growth and recurrence. The lining epithelium has a characteristic feature of the presence of mucus cells, ciliated cells, and intraepithelial glandular structures. This case report highlights the diagnosis and management of GOC in a 43-year-old female who presented with painless left facial swelling persisting for four months. Clinical examination revealed a firm swelling in the posterior left mandible involving the mandibular angle and the entire ramus. Radiographic evaluation demonstrated a well-circumscribed unilocular radiolucency with radiopaque margins in the affected region. A diagnosis of the glandular odontogenic cyst was made following a histopathological examination. The cyst was surgically removed, and the patient showed a favorable outcome with no recurrence over a follow-up period. Glandular odontogenic cysts, though rare, should be considered in the differential diagnosis of jaw cysts. Surgical resection was performed which remains the treatment of choice, with careful follow-up necessary due to the risk of recurrence.

Keywords: Odontogenic cyst, Cystic lesion, Mandibular cyst, Glandular cyst

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Corresponding Author
Dr. Sidra Fahim
Department of Oral Medicine, Altamash Institute of Dental Medicine, Karachi, Pakistan
sidra.raf@gmail.com

INTRODUCTION

Glandular odontogenic cysts (GOCs) are rare developmental odontogenic cysts that pose significant diagnostic and therapeutic challenges due to their aggressive behavior and high recurrence rates. First described by Gardner et al. in 1988,[1] these lesions are most frequently encountered in the anterior mandibular region but have also been documented in maxilla,[2,3] emphasizing the need for comprehensive evaluation of jaw cysts regardless of location. Although GOCs are often asymptomatic, they can present clinical features such as discomfort, edema, or jaw enlargement, which complicate early diagnosis and management. [4]

Epidemiologically, GOCs tend to occur slightly more frequently in men than in women and are most commonly observed in patients during their fifth or sixth decade of life.[5] In addition to their typical anterior mandibular localization, approximately 30.9% of cases exhibit tooth displacement or association with an unerupted tooth, 26% show cortical bone perforation, 24.3% present with clinical symptoms, and 13.9% demonstrate root resorption.[6] These features underscore the variability in presentation and the potential for significant osseous involvement.

Histologically, GOCs are characterized by a lining composed of thin layers of squamous and cylindrical cells, with areas of mucinous metaplasia. The presence of mucous cells, ciliated cells, and intraepithelial gland-like structures within an epithelium of variable thickness creates a diagnostic challenge, particularly in distinguishing GOCs from low-grade central mucoepidermoid carcinoma.[3] Immunohistochemical staining, notably with cytokeratin (CK), is often employed to facilitate accurate differentiation between these entities.[7,8]

Given the high rate of recurrence, reported between 25% and 55% after treatment surgical excision is generally preferred over conservative enucleation (4). In more aggressive cases, marginal resection may be indicated to further reduce the risk of recurrence.[9] Although malignant transformation of GOCs is rare, its potential occurrence highlights the importance of thorough diagnostic evaluation and meticulous treatment planning.[10]

This case report describes a patient diagnosed with a GOC who exhibited characteristic clinical and histological features and was successfully managed through surgical resection. By presenting this case, we aim to contribute to the existing literature on GOCs, emphasizing the need for accurate diagnosis and tailored surgical management to achieve favorable long-term outcomes.

Given the high rate of recurrence, the treatment of choice for GOCs is surgical excision rather than enucleation.[4] In more aggressive cases, marginal resection may be considered to minimize the risk of recurrence further.[9] While rare, malignant transformation of GOCs has been noted, underscoring the importance of thorough diagnostic evaluation and appropriate treatment planning.[10] This case report describes a patient diagnosed with a GOC who exhibited characteristic clinical and histological features and was successfully managed through surgical resection. By presenting this case, we aim to contribute to the existing literature on GOCs, emphasizing the need for accurate diagnosis and tailored surgical management to achieve favorable long-term outcomes.

CASE PRESENTATION

A 43-year-old female patient presented to the Department of Oral Medicine/ Oral Diagnosis of Altamash Institute of Dental Medicine, Karachi, Pakistan with a slow-growing painless swelling in the posterior region of the mandible. There was no significant associated family or medical history. On intraoral examination, the patient presented with a mobile second molar and localized swelling that had been causing pain for the past month. No discharge was observed, and the swelling caused the patient to experience difficulty chewing. Extraoral examination revealed a noticeable swelling on the left side of the face, located at the angle of the mandible, extending to the pre-auricular region (Figure 1).

Differential diagnosis

In the present case, the differential diagnosis included odontogenic keratocyst (OKC), ameloblastoma, and dentigerous cyst. OKC was considered due to its potential for aggressive growth and characteristic radiographic presentation, often appearing as a well-defined

radiolucency with possible cortical expansion. Ameloblastoma was also a possibility, given the extent of bone destruction and displacement of adjacent structures, though its typical multilocular appearance was not evident. A dentigerous cyst was included in the differential due to its common association with an impacted tooth; however, the degree of bone resorption and expansion suggested a more aggressive pathology. Ultimately, an incisional biopsy and histopathological examination confirmed the diagnosis of glandular odontogenic cyst (GOC), distinguishing it from these entities based on its unique epithelial features, including cyst-like spaces and glandular structures.

Imaging

Radiographic evaluation showed a large, well-circumscribed unilocular radiolucency, extending from the mandibular and involving the entire ramus. The lesion exhibited radiopaque margins, with significant bone resorption and destruction, particularly in the affected area. The third molar was impacted and displaced by the lesion toward the apical region of the lower left second molar. On the CT scan, the lesion appeared as a fluid-filled cystic structure with aggressive bone destruction and well-defined borders. There was no involvement of the inferior alveolar nerve canal, and no calcifications were present within the lesion.

Diagnosis

After conducting a comprehensive clinical examination and establishing a differential diagnosis, an incisional biopsy was undertaken to confirm the diagnosis of GOC. The diagnosis was confirmed based on histopathological examination. The characteristic features of the GOC were identified, including crypts of small cyst-like spaces within the thickness of the epithelium and a distinctly glandular structure lining.

Treatment

After obtaining informed consent, the patient underwent surgical excision of the lesion through an extraoral submandibular approach. An incision was placed, and a flap was carefully raised to preserve the marginal mandibular nerve. Upon exposure, the cystic lesion was excised completely. A peripheral osteotomy was performed, removing approximately 5 mm of bone around the lesion to eliminate any microscopic extensions and reduce the risk of recurrence (Figure 1).

Hemostasis was achieved, and the surgical site was meticulously closed with 3.0 vicryl sutures. A pressure dressing was applied, and a drain was attached to support post-operative recovery and healing the different phases from diagnosis till treatment of the patient are shown in (Figure 2-7).

Postoperative Outcome

The patient was followed up for 6 months, during which no signs of recurrence were observed. The patient has remained symptom-free and is under regular monitoring till date.

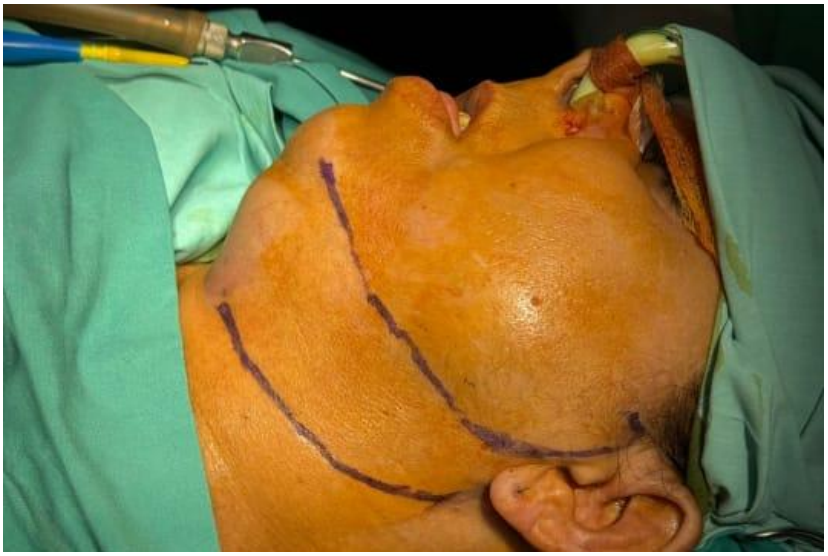

Figure 1: Extraoral extent of swelling with preoperative demarcation of surgical incision

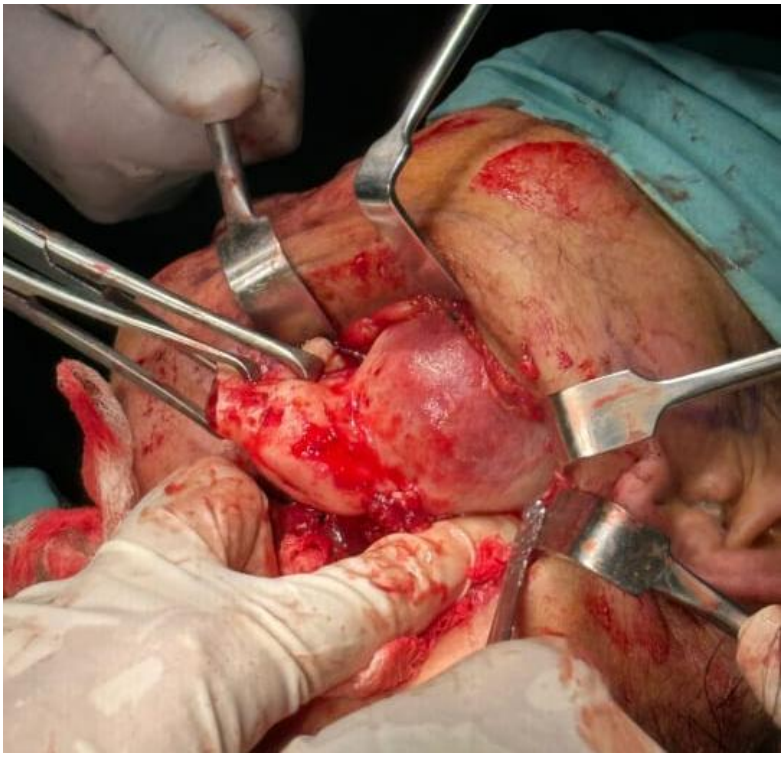

Figure 2: Surgical access achieved; peripheral osteotomy in intraoperative phase

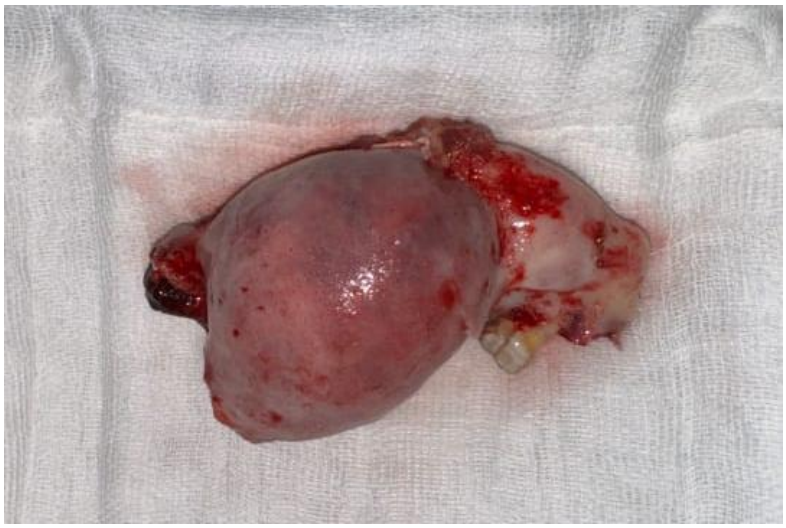

Figure 3: Excised cystic lesion with associated posterior teeth

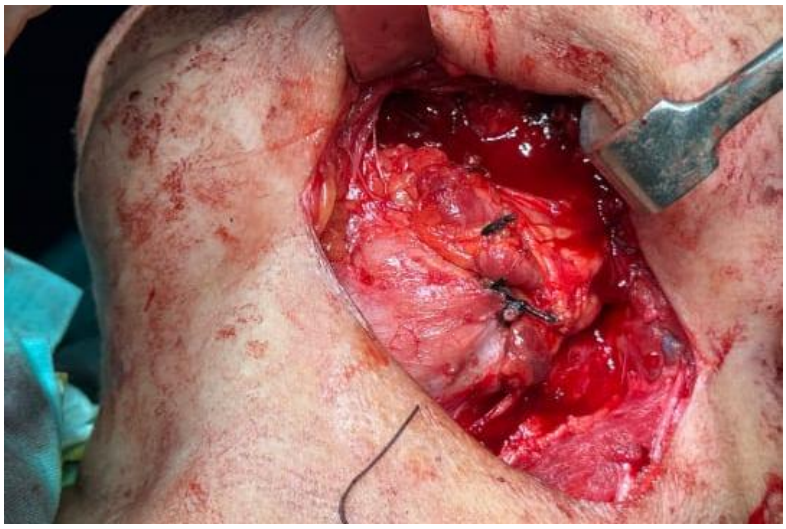

Figure 4: Postoperatively intraoral sutures placed at surgical site

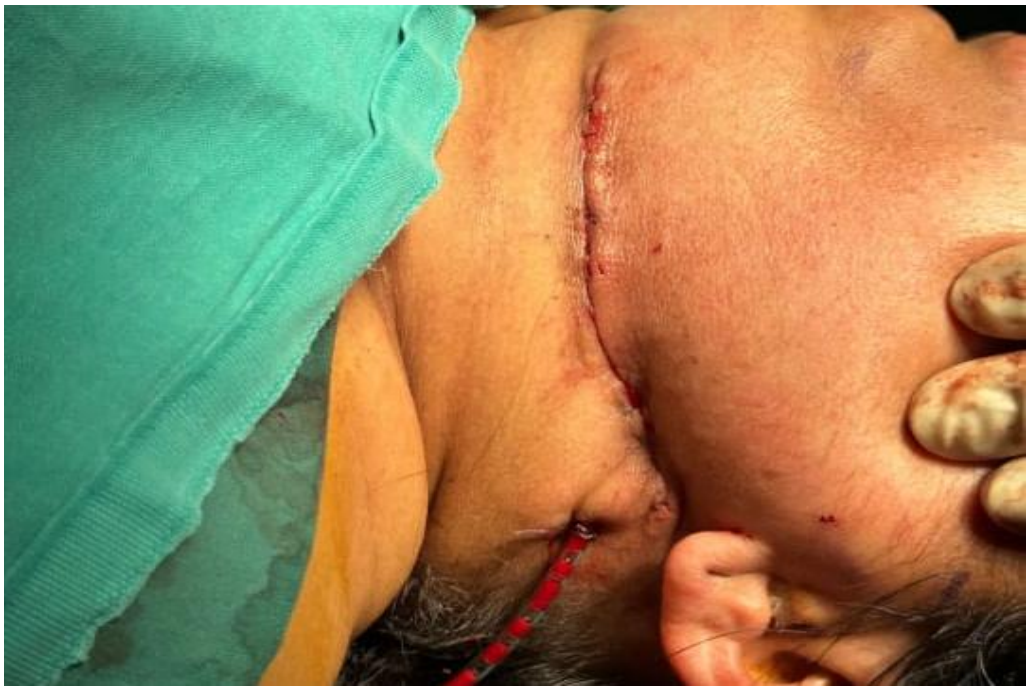

Figure 5: Appearance of the surgical wound after suturing extra orally and drain placement

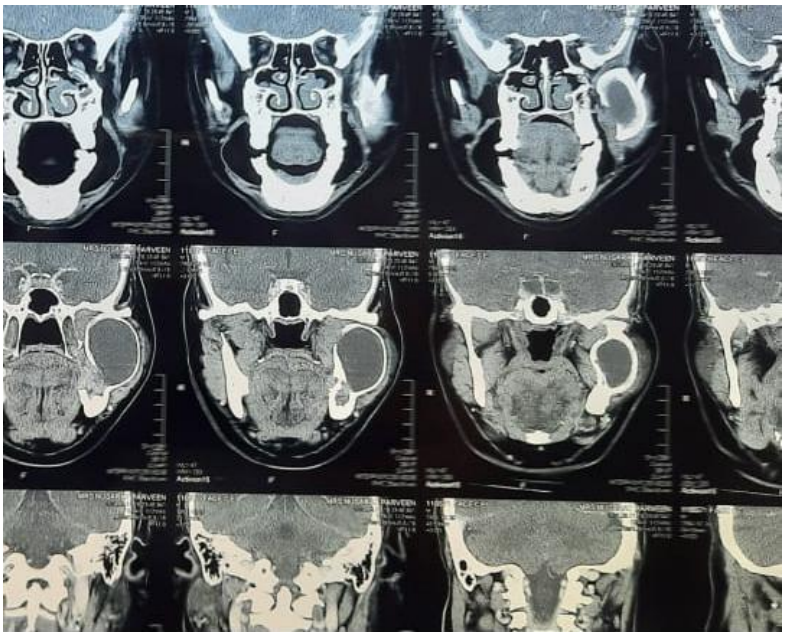

Figure 6: Coronal view of CT displaying cortical bone swelling of left mandible

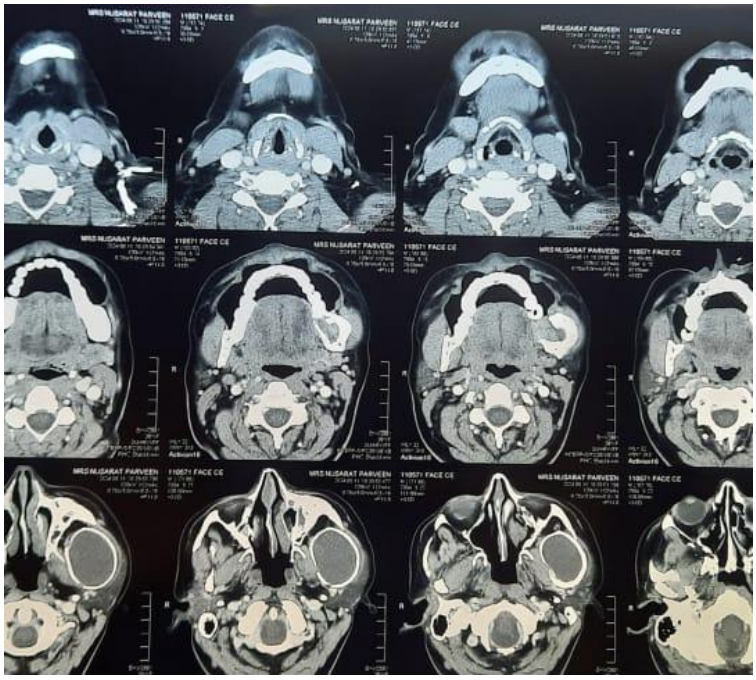

Figure 7: Axial view of CT showing extension of cyst

DISCUSSION

About 0.2% of all odontogenic cysts are glandular odontogenic cysts (GOCs), which are rare odontogenic cysts that lack a distinctive pathognomonic radiological appearance.[3] Even though, GOCs usually occur in the anterior jaw, our case highlighted the variation in presentation by involving a lesion in the posterior mandible.[8,11] The youngest known case of this cyst type included a 12-year-old boy, and it rarely affects people in their first ten years of life.[6] Fascinatingly, the first documented instance of bilateral GOC involving the anterior maxilla in a 29-year-old man underscores the variety of ways this cystic lesion manifests itself.[12]

Glandular odontogenic cysts (GOCs) are diagnostically challenging due to their resemblance to other odontogenic lesions, including lateral periodontal cysts (LPC), botryoid odontogenic cysts (BOCs), radicular and residual cysts with mucous metaplasia, and low-grade mucoepidermoid carcinoma, as noted by Shah et al.[13] The diagnosis was confirmed in this case through histopathological examination, which showed glandular structures, and imaging helped guide surgical planning.

Odontogenic cysts can transform into neoplasms, with factors like chronic inflammation and incomplete removal contributing to this risk. A systematic review reported cases, including glandular odontogenic cysts, progressing to ameloblastoma or carcinoma, highlighting the need for meticulous excision and regular follow-ups.[10] Since conservative enucleation has been linked to high recurrence rates up to 50% surgical resection is recommended for GOC.[4,14] In our case, surgical resection with 5 mm of bone removal was chosen due to the cyst's aggressive nature and high recurrence potential. Additional procedures, such as peripheral ostectomy or 5-fluorouracil (5-FU) application, are used to further reduce recurrence risk, typically involving the removal of about 5 mm of the surrounding bone.[14] Given that recurrence rates are often highest between 3 and 5 years post-treatment, a follow-up period of at least five years is recommended.[14]

Differentiating GOC from low-grade central mucoepidermoid carcinoma is crucial, as both lesions exhibit similar morphological characteristics.[7] Immunohistochemical analysis,

particularly cytokeratin (CK) staining, is a valuable tool in distinguishing between these entities.

CONCLUSION

Glandular odontogenic cysts, though rare, pose a diagnostic challenge due to their overlap with other odontogenic lesions. This case emphasizes the importance of histopathological evaluation in diagnosing GOC and the role of surgical resection in management. Continued follow-up is necessary to monitor for potential recurrence.

Author Contributions

NI, SS and SF: Conceptualization, Performed surgical procedure.

SF and SS: Investigation, Methodology. NI & SF and NI: Methodology, Writing review & editing.

SF and SS: Validation, Supervision, Writing review & editing.

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Institutional ethical board approval

Not applicable.

Informed Consent

The written consent was obtained from all participants in this study.

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None

Availability of data and materials

The data supporting this study's findings are available from the corresponding author upon reasonable request.

Consent for publication

Consent was obtained from all participants

Disclaimer of using AI tools

Not utilized. All ideas, arguments, and conclusions presented in the review, however, are entirely the authors' original work. The authors take full responsibility for the accuracy and integrity of the content.

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