

Staged Minimally Invasive Surgical Approach in the Management of Glandular Odontogenic Cyst

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ABSTRACT

Aim: The aim of the study was to report a unique case of glandular odontogenic cyst (GOC) in an unusual location of anterior maxilla.

Materials and methods: Considering the age, sex, and location of the lesion, a minimally invasive surgical procedure was opted considering esthetics, such as decompression followed by enucleation and fresh frozen bone (FFB) grafting. To arrive at the diagnosis, cytokeratin (CK)-19 was employed to differentiate from suspected intraosseous mucoepidermoid carcinoma.

Results: With the evidence of pre- and post treatment histopathological changes, the lesion appeared less aggressive justifying the treatment executed.

Conclusion: Glandular odontogenic cyst is a locally aggressive jaw cyst, which has a high rate of cortical perforation and recurrence that poses both diagnostic and therapeutic challenges.

Clinical significance: Treatment of decompression, followed by enucleation may be considered as an effective option in comparison to aggressive surgical intervention.

Keywords: Decompression, Fresh frozen bone graft, Glandular odontogenic cyst, Immunohistochemistry.

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INTRODUCTION

A unique case of GOC in an unusual location of anterior maxilla managed with staged minimally invasive surgical approach is reported here.

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BACKGROUND

Glandular odontogenic cyst is an uncommon developmental lesion, and till date, about 181 cases have been reported. The frequency of occurrence ranges from 0.12 to 0.13% of all jaw cysts.¹ According to Kaplan et al,^{2,3} GOCs occur between 14 and 75 years of age and most commonly in anterior mandible, presenting as an asymptomatic swelling.

Although the radiographic features are not pathognomonic, GOC presents as unilocular or multilocular radiolucent lesion with well-defined or scalloped margins.⁴

Glandular odontogenic cyst shares similar microscopic features with other cysts and tumors, such as radicular or dentigerous cysts with mucous metaplasia, botryoid odontogenic cyst (BOC), lateral periodontal cyst (LPC), and low-grade mucoepidermoid carcinoma (LMEC), hence definitive diagnosis may be difficult to achieve.³

Differentiating GOC from the aforementioned lesions is essential for better therapeutic management. Since GOC behaves as an aggressive lesion, it requires a more radical surgical approach along with a longer follow-up period than dentigerous, radicular, or BOC. Low-grade mucoepidermoid carcinoma, being a malignancy, requires a different management protocol like complete resection of the jaw.⁵

As GOC is a rare lesion and its occurrence in the maxilla is unusual with a histopathologic diagnostic dilemma, a unique case of GOC in the maxilla confirmed with immunohistochemistry and managed with the minimally invasive mode of treatment has been reported.

CASE REPORT

A 20-year-old female reported with the complaint of a slow-growing swelling over the upper anterior gum region since a year (Fig. 1). On intraoral examination, the swelling was noted in 12 to 23 region obliterating the labial vestibule with the same color as that of labial mucosa. On palpation, swelling was hard in consistency and was fixed to the underlying structures. Orthopantomogram revealed single well-defined radiolucency over the upper anterior teeth from 12 to 23 region with displacement of the roots of 11, 21, and 22 (Fig. 2). A



Fig. 1: Intraoral view depicting swelling in the upper labial vestibule

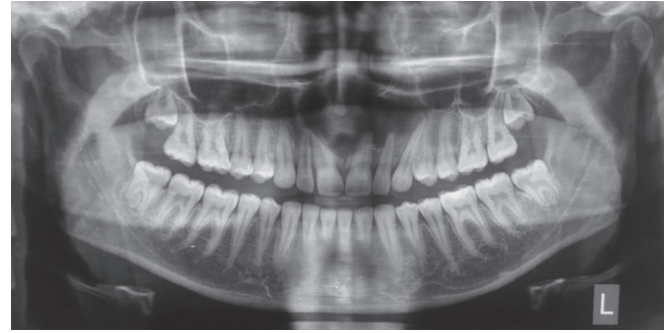


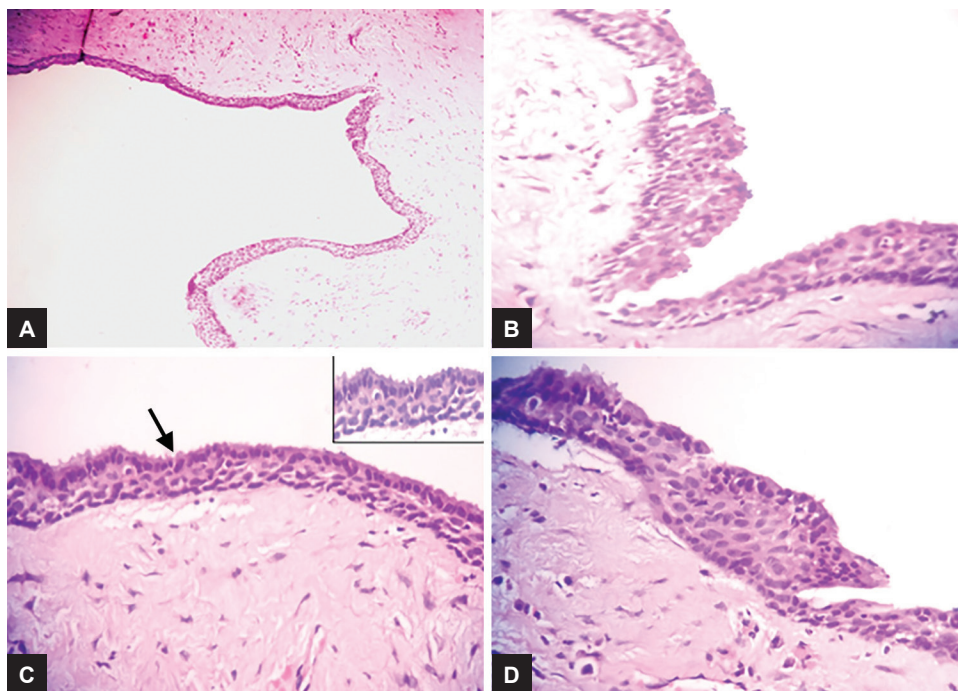
Fig. 2: Orthopantomogram showing well-defined unilocular radiolucency extending from 12 to 23 region

Histopathological and Immunohistochemical Findings

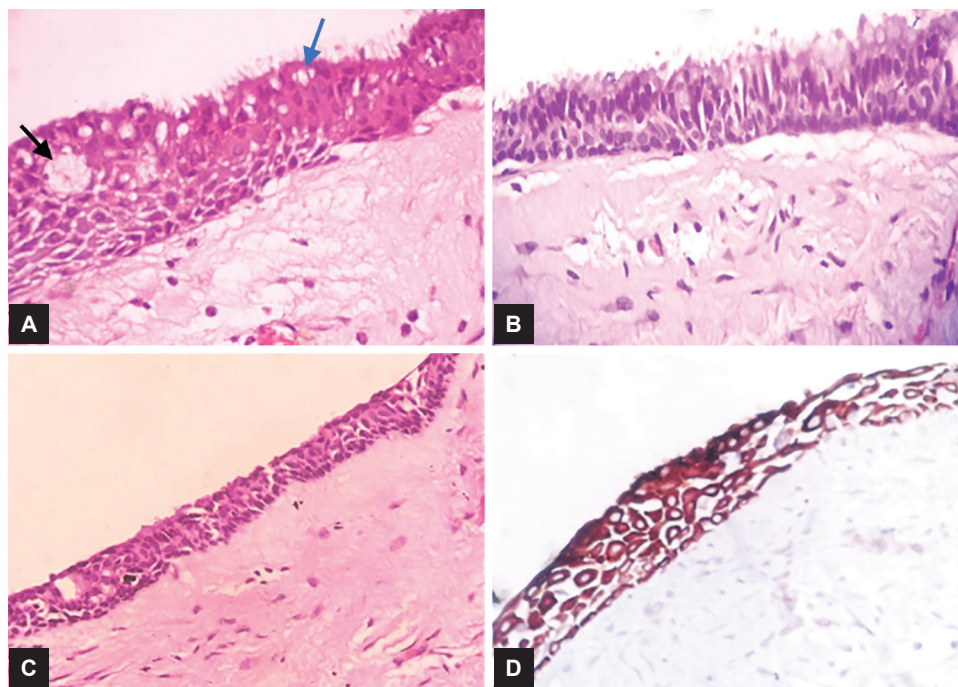
provisional diagnosis of odontogenic keratocyst (OKC) was arrived from the clinical and radiographic findings. Routine hematological investigations were performed and were within normal range. Since the lesion presented in the form of a cyst in the anterior maxilla, it was decided to manage it by a minimally invasive surgical procedure, such as decompression of the cystic cavity followed by enucleation with bone grafting.

Under local anesthesia, straw-colored fluid was aspirated, and incisional biopsy was done and sent for histopathologic examination.

Hematoxylin and eosin (H&E)-stained sections showed a cystic lumen with lining epithelium and a fibrous capsule. Epithelial lining showed variation in thickness and type, ranging from stratified squamous nonkeratinized to reduced enamel epithelium that displayed papillary-like proliferation in focal areas (Figs 3A and B). The superficial cuboidal eosinophilic cells (hob-nail) showed filiform cytoplasmic extensions into the lumen and intraepithelial spherule formation in focal areas (Figs 3C and D). The lining also showed pseudo-stratified columnar epithelium with ciliated cells, mucous cells, clear cells, crypts, and microcysts (Figs 4A to C). The capsule comprised of fibrovascular connective tissue, few



Figs 3A to D: Photomicrographs (H&E stain) showing lining epithelium ranging from stratified squamous nonkeratinized to reduced enamel epithelium with varying thickness (A)(X40) and focal areas of papillary proliferation (B) (X100); superficial cuboidal cells with eosinophilic cytoplasm showing filiform cytoplasmic extensions into the lumen (C) (X100) (inset X400); intraepithelial spherule formation (D)(X100)



Figs 4A to D: Photomicrographs showing lining epithelium with ciliated, mucous (black arrow), clear cells, crypts, and microcysts (blue arrow) (A) (H&E, X100); pseudo-stratified columnar epithelium (B) (H&E, X100); crypts and microcyst formation (C) (H&E, X100); CK-19 membranous and cytoplasmic positive staining (D) (X100)

odontogenic epithelial rests, and minimal inflammatory infiltrate with cholesterol clefts.

The diagnosis of LPC and BOC was excluded based on radiographic examination. Low-grade mucoepidermoid carcinoma was considered as one of the differential diagnosis due to positivity of mucous and clear cells with mucicarmine and Periodic Acid Schiff stains. Immunohistochemical staining was done using CK-19, and it showed positive membranous and cytoplasmic staining of odontogenic epithelium (Fig. 4D). Thus, the diagnosis of GOC was confirmed with the exclusion of LMEC.

Treatment

An elliptical vestibular incision was placed in the anterior maxilla between right central incisor and left lateral incisor, and a bony window was created to access the cystic lesion. Sterile saline was used to lavage the cystic contents. Partial excision of the cystic lining was done. The cavity was packed with betadine-soaked ribbon gauze that was changed every alternate day for a week.

An acrylic stent was fabricated after a week, and patient was advised to wear it all the time. Periodic trimming of the stent was done till the size of the cavity reduced to half the initial size that was confirmed by the panoramic radiograph (Fig. 5A).

The patient was followed up after 6 months for enucleation, and fresh frozen bone (FFB) graft was placed into

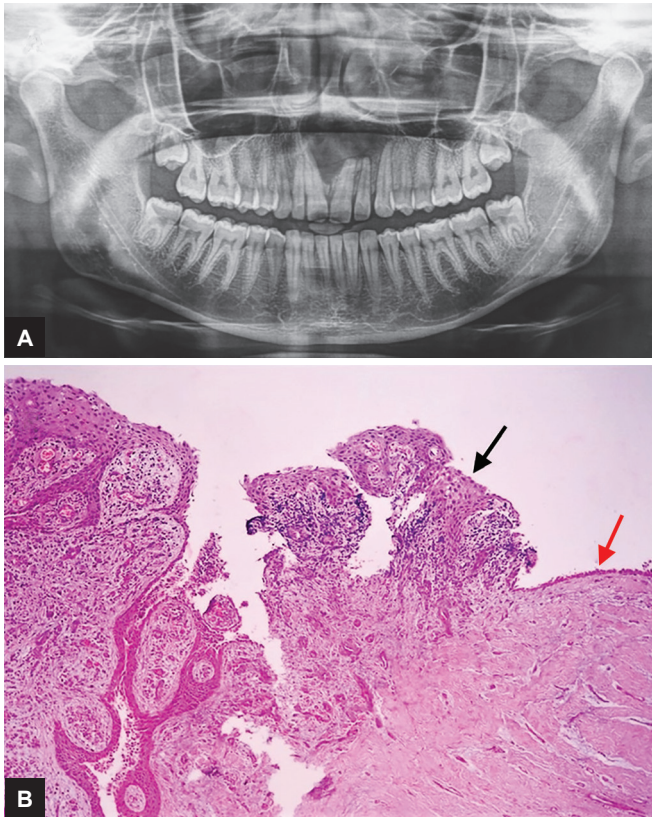
the resultant bony defect. The entire enucleated tissue lining was sent for histopathological examination. Post-enucleation, histopathology was consistent with the previous incisional/decompression procedure histopathology of GOC. However, there was transition seen from GOC cystic lining to stratified squamous epithelium in focal areas (Fig. 5B). Postoperative healing was uneventful. Radiographic examination revealed substantial healing in the bony cavity.

DISCUSSION

Glandular odontogenic cyst is a rare type of jaw cyst that presents with varied radiographic and histological appearances mimicking certain aggressive lesions, such as LMEC and BOC. Thereby numerous designations have been attributed to GOC, such as mucoepidermoid odontogenic cyst, sialo-odontogenic cyst, and polymorphous odontogenic cyst.^{3,6,7} The term GOC was first used by Gardner et al and was adopted by the World Health Organization in the year 1992.³

The histogenesis remains unclear for GOCs, but it is believed that it originates from rests of dental lamina.⁸ The microscopic features of GOC are variable and ambiguous. Therefore, Kaplan et al³ categorized the microscopic features of GOC and divided them into major and minor criteria to facilitate definitive diagnosis.

The histopathological differential diagnosis of GOC include LPC, BOC, and LMEC, and differentiating these



Figs 5A and B: Orthopantomogram showing well-defined unilocular radiolucency extending from 11 to 22 region (A); Photomicrograph showing change of cystic epithelium (red arrow) to stratified squamous epithelium (black arrow)(B) (H&E, X40)

lesions is necessary to avoid over- or undertreatment of the lesion.⁹

Lateral periodontal cyst is an odontogenic cyst of developmental origin which also exhibits epithelial plaques and cells rich in glycogen, analogous to those seen in GOC. Botryoid odontogenic cyst is a polycystic variant of LPC that is locally aggressive and shows microscopic features resembling GOC, such as focal epithelial thickenings and clear glycogen-rich clear cells.¹⁰ However, in the present case, the radiographic findings were helpful in excluding the diagnosis of LPC as the radiolucency was not localized adjacent to the root of a vital tooth and was not polycystic in nature. The presence of ciliated epithelium, hobnail cells, and duct-like spaces with mucous cells favored the diagnosis of GOC rather than LPC and BOC.

Due to overlapping features in histopathology and differences in management, it is essential to differentiate LMEC from GOC. As numerous mucous and clear cells were observed in the lining, the exclusion of LMEC was necessary. Positivity to CK-18 and -19 immunostaining in GOC was the possible aid in distinguishing GOC from LMEC. Therefore, in the present case, CK-19 immunostaining was performed that was homogeneously and strongly positive in the cytoplasm of all layers of the epithelium, which was consistent with the studies of

Mascitti et al⁴ and Shen et al.¹¹ Thus, the diagnosis of GOC was confirmed in the present case.

Pires et al¹² assessed the CK profile of central MEC, GOC, and other odontogenic cysts and tumors. They found expression of CKs 18 and 19 to be highly useful in the differential diagnosis of these lesions. Since CK-19 is a marker of odontogenic differentiation, the low frequency of CK-19 expression in central MECs suggests that they may lose their CK-19 expression during neoplastic transformation or they are of non-odontogenic origin.¹²

The choice of treatment for GOC is still debatable and varies from conservative to aggressive modes of management. Conservative approaches include marsupialization, enucleation, curettage with adjuvant Carnoy solution, curettage with and without peripheral osteotomy, or cryotherapy. Newly recommended aggressive methods include marginal resection and partial jaw resection.^{11,13} However, for the present case, considering the age, sex, and the location of lesion, a minimally invasive surgical procedure was opted, such as decompression followed by enucleation and FFB grafting to enhance the healing. The prognosis after decompression was better clinically which was supported with radiological evidence by significant reduction of bony defect that is half of its initial presentation within a duration of 6 months. At present, the patient is on regular follow-up for the second year, and there is no asymmetry of the face, no visible or palpable bone distortion over the previously diseased area of the maxilla.

The findings of this case report highlight the benefits and the importance of the minimally invasive surgical treatment in the management of GOC which is in concordance with the research of Jayasinghe et al.¹⁴ To reduce the morbidity and recurrence rate, similar study with conservative management for OKC and cystic ameloblastoma was executed by Prasad et al.¹⁵ They evaluated the effect of decompression followed by enucleation with chemical cauterization in 13 cases, and found recurrence in 1 case after 5 years of follow-up.

The high recurrence rate of GOCs is related to incomplete cyst removal because of fragile epithelium that has a tendency to detach from the connective tissue, spread through cancellous spaces of bone and multicystic configuration.¹³ Hence, the treatment of decompression followed by enucleation was opted in the present case instead of enucleation alone, which resulted in decreasing the aggressive nature of the lining epithelium that was confirmed histologically as well as clinically.

CONCLUSION

Glandular odontogenic cyst is a locally aggressive jaw cyst, which has a high rate of cortical perforation and recurrence that poses both diagnostic and therapeutic challenges.

CLINICAL SIGNIFICANCE

Minimally invasive approach showed paramount results without leaving morbidity in the surrounding tissues. Hence, the treatment of decompression, followed by enucleation, can be considered as an option before aggressive surgical intervention. A close follow-up is mandatory to precociously diagnose eventual recurrences.

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