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Case Report

Glandular odontogenic cyst: Case report and literature review

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ABSTRACT

Glandular Odontogenic Cyst (GOC) is a rare and aggressive developmental odontogenic cyst, presenting significant diagnostic and therapeutic challenges. Since its initial characterization, GOC has remained a diagnostic dilemma due to its histopathological similarities with other odontogenic cysts and tumors, such as dentigerous cysts, lateral periodontal cysts, and central mucoepidermoid carcinoma. Accurate diagnosis often requires a meticulous histopathological examination, as well as consideration of clinical and radiographic features to differentiate GOC from other similar lesions.

This case report aims to present a rare occurrence of Glandular Odontogenic Cyst in the anterior maxilla, underscoring the importance of comprehensive diagnostic evaluation, histopathological examination and effective management strategies.

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1. Introduction

The glandular odontogenic cyst (GOC) is a rare type of developmental odontogenic cyst characterized by its local aggressiveness. It comprises 0.4% of all odontogenic cysts.¹ Padayachee et al. first reported the cyst in 1987, describing two cases of atypical odontogenic cysts exhibiting characteristics of botryoid odontogenic cyst (lateral periodontal cyst) and central mucoepidermoid carcinoma (CMEC), alongside a glandular component. They coined the term "sialo-odontogenic cyst" to describe this entity.² However, the histological characteristics of Glandular Odontogenic Cysts strongly suggest an origin from odontogenic tissues rather than salivary glands. Consequently, in 1992, the World Health Organization designated GOCs as a distinct histopathological entity and categorized them as developmental odontogenic epithelial

cysts.³

Certain microscopic characteristics seen in Glandular Odontogenic Cysts (GOC) may resemble features found in other lesions like dentigerous cysts, lateral periodontal cysts, and even Central Mucoepidermoid Carcinoma (CMEC). This underscores the need for careful histopathological assessment to ensure accurate diagnosis and appropriate management.¹

The purpose of this article is to report a case of Glandular Odontogenic Cyst (GOC) occurring in the anterior maxilla due to its relative rarity, highlighting its clinical, radiographic, and histopathological characteristics, along with the successful surgical approach employed for its management.

2. Case Presentation

A 36-year-old male patient reported with a chief complaint of swelling in the anterior region of the palate for 3 months.

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The swelling gradually increased to its current size, and the patient did not experience any functional problems. It was asymptomatic, non-tender, and not associated with any pus discharge.

Examination revealed facial asymmetry with mild swelling in the middle third of the face, obliterating the left nasolabial fold. There was a deviation of the nose towards the right side. Intraoral examination showed a non-tender, non-pulsatile swelling in the anterior hard palate region, extending from teeth 12 to 23, approximately measuring 12 x 10 mm. The overlying mucosa showed no colour change. A prosthesis was present on teeth 11, 21, and 22. Tooth 11 was vital (confirmed with an electric pulp tester), tooth 21 was missing, and tooth 22 had a history of root canal treatment.

Cone-beam computed tomography (CBCT) showed a well-defined unilocular hypodense lesion in the anterior region of the hard palate, crossing the midline and measuring approximately 14.1 mm (antero-posteriorly) x 12.6 mm (mesio-distally) x 20.1 mm (supero-inferiorly). Erosion of the nasal floor was observed in the lesion area, along with thinning and partial loss of palatal cortical bone. External root resorption was noted in the middle and apical third of tooth 22, and internal resorption was observed in the crown of tooth 23. Widened incisive canal noted. Deviated nasal septum noted towards right side (Figure 1).

Treatment plan was explained to the patient and written informed consent was taken from the patient for the procedure and for reporting the case.

Under local anaesthesia and aseptic precautions, a crevicular incision was made from teeth 13 to 23, and a palatal mucoperiosteal flap was reflected. The pathology was exposed. As the pathology did not involve the nasopalatine nerve, the lesion was enucleated in toto without damaging the nerve. The lesion cavity was irrigated thoroughly, haemostasis was achieved, and the palatal flap was closed with 3-0 vicryl sutures (Figure 2).

Histopathology of the excised specimen revealed features consistent with a glandular odontogenic cyst (Figure 3A,B). Histopathological findings included (Figure 4).

Cystic lumen lined by non-keratinized stratified squamous epithelium of variable thickness, partially ciliated, with scattered goblet cells.

Cuboidal basal cells with hyperchromatic nuclei and absence of rete ridges.

Hobnail cells, clear cells, and vacuolated cells with eosinophilic cytoplasm in the epithelial lining.

Mild chronic inflammatory infiltrate in the subepithelial connective tissue, with lymphocytes, plasma cells, and blood vessels.

The diagnosis was confirmed based on Kaplan's criteria for glandular odontogenic cyst, meeting major criteria including squamous epithelial lining, hobnail cells, mucous

cells, and intraepithelial glandular structures. Minor criteria met included the presence of ciliated cells and multiluminal architecture.

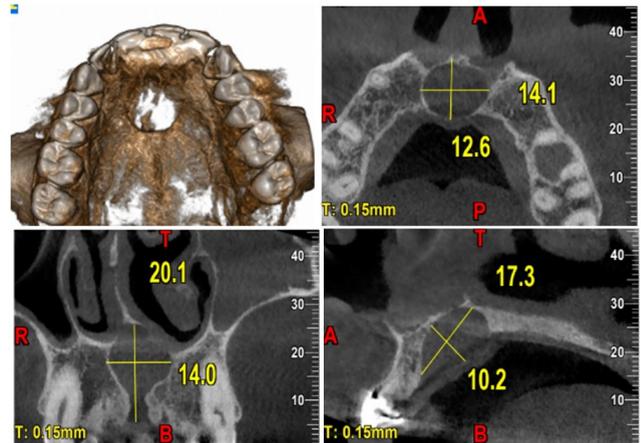


Figure 1: CBCT image

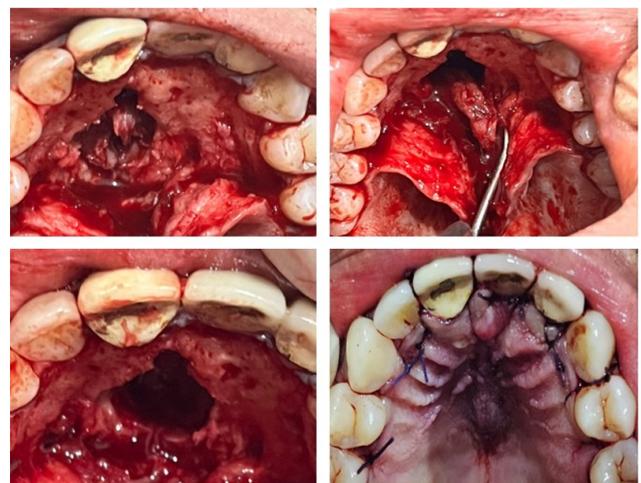


Figure 2: Intraoperative picture

3. Discussion

According to Gurler et al., the prevalence of Glandular Odontogenic Cysts (GOC) ranges from 0.012% to 1.3% of all jaw cysts, with a mean prevalence of 0.17%. Clinically, GOC typically affects the mandibular anterior region and often presents as an asymptomatic, slow-growing swelling.⁴ However, in our case report, the cyst was observed in the anterior maxillary region, highlighting its atypical presentation compared to the common clinical pattern. History of previous Root Canal treatment with upper left central incisor raised the suspicion of Radicular cyst. Aspiration biopsy was performed which showed a clear, watery fluid, not typically seen with radicular cyst. Clinical

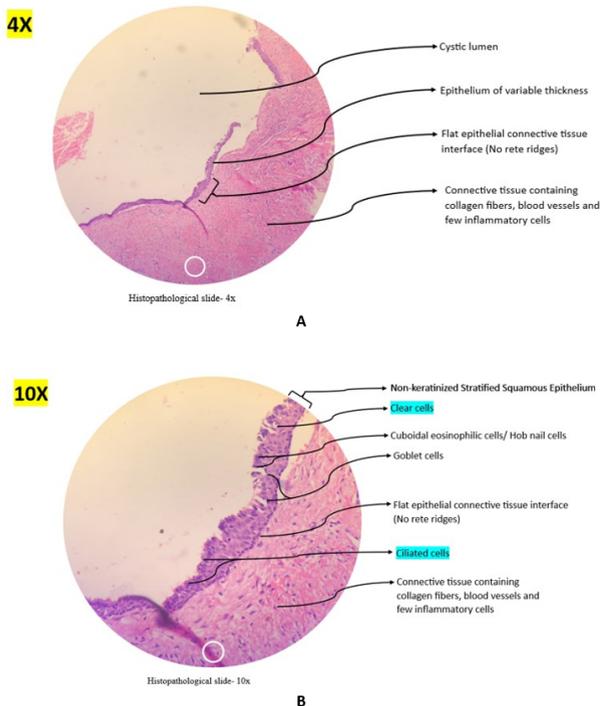


Figure 3: Histopathological images of excised specimen

Histopathological Report

H & E stained section shows single bit of tissue.

The lesional tissue shows presence of cystic lumen lined by non-keratinized stratified squamous epithelium of variable thickness, which is partially ciliated with few goblet cells scattered. The basal epithelial cells are cuboidal with round to flattened hyperchromatic nuclei. There is absence of rete ridges. At places the epithelium is separated from the connective tissue.

The lining epithelium shows presence of cuboidal eosinophilic hobnail cells, clear cell with empty cytoplasm and eccentric nucleus.

The subepithelial connective tissue shows collagen fibre bundles interspersed with fibroblasts. There is mild chronic inflammatory infiltrate composed predominantly of lymphocytes and plasma cells along with endothelial lined blood vessels.

Histopathological Diagnosis: **Glandular Odontogenic Cyst.**

Figure 4: Histopathological report

appearance of the lesion and the location also led to a differential diagnosis of Nasolabial cyst. Arora et al.,² similarly reported a rare case of Glandular odontogenic cyst in anterior maxilla which had a differential diagnosis of periapical abscess, radicular cyst, or a benign tumour of the jaw.

Glandular Odontogenic Cysts (GOC) typically affect individuals aged between their fifth and sixth decades and often manifest as painless swellings in the mandibular symphysis and body. This condition often exhibits aggressive behavior, characterized by cortical expansion and perforation, tooth displacement, and root resorption. Additionally, GOCs have a recurrence rate of approximately 21%. Radiographically, the lesion commonly presents as a

well-defined unilocular radiolucency with sclerotic borders, typically involving the roots and periapical area of erupted teeth.⁵ In this case, external root resorption was seen with 22 which helped in excluding the diagnosis of nasolabial cyst. Gurler et al.,³ in their case series of 5 cases also observed a well-defined unilocular or multilocular radiolucent lesions in all patients. Root resorption was also seen in two of the five patients.

Since the initial reports of the disease, diagnosing and managing Glandular Odontogenic Cysts (GOC) have presented clinical challenges. A significant concern revolves around distinguishing between the histopathological characteristics of GOC and those of low-grade Central Mucoepidermoid Carcinoma (CMEC), highlighting the complexity of achieving precise diagnosis. Fowler et al. suggested that the presence of seven or more microscopic features strongly indicates Glandular Odontogenic Cyst (GOC). These features encompass "hobnail cells in the surface of the cyst lining, intraepithelial microcysts or duct-like spaces, apocrine snouting, clear or vacuolated cells, variable thickness of the cyst lining, papillary projections, mucous goblet cells, epithelial spheres, cilia, and multiple cystic spaces or compartments".⁶ In our case report, the lesion exhibited at least seven microscopic parameters supportive of the diagnosis of GOC.

On Immunohistochemistry, Glandular Odontogenic Cysts (GOC) typically exhibit strong positivity for bcl-2 in the basal and suprabasal cell layers, as well as CK 7, 8, and 19, indicating its odontogenic origin.⁷ Differential diagnosis includes lateral periodontal cyst and central mucoepidermoid carcinoma (CMEC). Lateral periodontal cysts lack ciliated epithelium with duct-like spaces and mucous cells, while CMEC expresses strong positivity for CK18 and Maspin and lacks superficial cuboidal cells, epithelial whorls, ciliated cells, and intraepithelial microcysts.⁸

In the literature, there's no consensus on the preferred treatment for Glandular Odontogenic Cysts (GOC), as it varies based on the surgeon's discretion. Enucleation and curettage are frequently performed procedures. Other successful approaches include marsupialization, jaw resection, and treatments such as applying Carnoy's solution, employing bone grafts, and utilizing cryosurgery.² In our case, the cyst was completely enucleated under local anesthesia, followed by primary closure, leading to an uneventful postoperative recovery.

Fowler et al. documented a 50% recurrence rate for the lesions over an average follow-up period of 8.75 years.⁶ In contrast, Kaplan et al. reported a lower recurrence rate of 29.2% within a follow-up range of 0.5 to 7 years, with a mean follow-up duration of 2.9 years.⁹ Additionally, Chrcanovic et al. reported a recurrence rate of 21.6% over a follow-up period ranging from 0.1 to 20 years, with a mean follow-up duration of 4.5 years.¹⁰

Recurrence is not solely attributed to the biological characteristics of the lesion but is also influenced by the management approach implemented and a long term follow up is recommended owing to its high rate of recurrence.

4. Conclusion

Glandular Odontogenic Cysts (GOC) lack distinctive clinico-radiographic features, often mimicking various other lesions. A meticulous histopathological examination and extended follow-up, are essential to exclude recurrences. Given the overlapping clinical and radiological findings with other odontogenic lesions, thorough histopathological assessment stands as the cornerstone of GOC diagnosis. The high recurrence rate and aggressive behavior of GOC necessitate appropriate management strategies and vigilant long-term monitoring.

All images are original and not taken from any source, written informed consent has been taken from the patient for the procedure and the right to use these photographs.

5. Source of Funding

None.

6. Conflict of Interest

None.

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