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RESEARCH ARTICLE

GLANDULAR ODONTOGENIC CYST OF ANTERIOR MAXILLA IN A YOUNG PATIENT- A RARE CASE REPORT

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ABSTRACT

The glandular odontogenic cyst is a rare developmental jaw bone cyst of odontogenic origin described as a distinct entity by Gardner *et al*. in 1988. The cyst occurs primarily in middle- aged individuals and has slight predilection for mandible. Glandular odontogenic cysts (GOC) accounts for 0.012 to 1.3% of all jaw cysts, show aggressive behavior and presents as an expansion with or without pain or paresthesia with a high potential of recurrence which can be due to incomplete removal of the lining following conservative treatment. This article presents a rare case report of glandular odontogenic cyst in a 14-year old male patient in the anterior region of the maxilla.

INTRODUCTION

Glandular odontogenic cyst (GOC) is an uncommon developmental cyst. [1] GOC has a frequency rate of 0.012 - 1.3% of all the jaw cysts [2]. Although GOCs are relatively rare, their correct diagnosis is of major clinical importance because they have aggressive growth potential, asymptomatic with or without paresthesia, high incidence of cortical perforation and a relatively high rate of recurrence. [3]

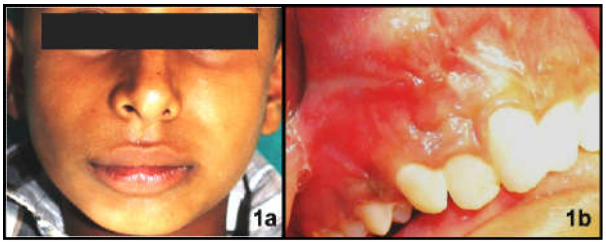
In 1987, Padayachee and Van Wyk described the microscopic details of two cases with features of GOC. They speculated on the possibility of salivary gland origin and proposed the term sialo-odontogenic cyst. [4] This cyst has been established as a distinct entity by Gardner *et al*. in 1988. [5] Though the lesion was initially referred to as a sialo-odontogenic cyst, its name was changed to glandular odontogenic cyst by Gardner *et al* because of the lack of evidence of salivary gland origin, and the term was later adopted by the World Health Organization. [6]

Mostly GOCs occur in middle-aged individuals involving mandible anterior region. [1] Several authors have suggested for aggressive treatment approach of the cyst due to a high recurrence rate. [2] Thus, the aim of this article is to discuss a rare case of GOC involving maxillary anterior region in a young male patient who was treated with enucleation of cyst followed by chemical cauterization using Carnoy's solution and showed no sign of recurrence after 1 year of follow-up.

Case report – A 14-year-old male, reported to our department with a chief complaint of asymptomatic swelling present on the right side of face since 4-5 months. According to the patient, initially swelling was small in size and gradually increases to the present size without any signs of pain and paresthesia. The patient's medical history was non-contributory.

On extra-oral examination, a solitary diffuse swelling was present with obliteration of ala nose involving right side of face (Figure 1a). The swelling is extending superior-inferiorly from inferior orbital foramen till the vermilion border of upper

lip and anterio-posteriorly from philtrum of upper lip till 3cm away from tragus of ear. The swelling was approximately 3cm x 4cm in size, oval in shape, smooth surface with obliteration of right side of ala of nose. It was firm in consistency and non-tender on palpation.



1a. A solitary diffuse swelling involving right side of face obliterating ala of nose.
1b. Intraoral view of the swelling in right anterior maxilla showing intact mucosa with obliteration of labiobuccal sulcus i.r.t 21, 11,12, 13, 14

On Intra-oral examination, a solitary, well defined swelling was present on the right side of maxilla with obliteration of the labio-buccal vestibule. The swelling extends from the mesial aspect of 11 to distal aspect of 14. The coronal portion of 11 is overlapping on 21. The swelling was approximately 2cm x 2cm in size, oval in shape. It was firm in consistency and non-tender. The adjacent oral mucosa was intact and normal in appearance (Figure 1b). All the teeth were sensitive to percussion. On chair side investigation, electric pulp vitality test was performed on which all teeth were vital. On further investigation, a panoramic radiograph was made, which revealed a well-defined, unilocular radiolucency present in the anterior maxilla crossing the midline, involving the roots from distal aspect of maxillary left lateral incisor & extending upto distal aspect of maxillary right second premolar. The radiolucency has sclerotic borders with displacement of right maxillary sinus. The divergence of roots of 11, 12, 13, 21 and 22 was well appreciated. (Figure 2). Correlating history, clinical examination and radiographic findings, a differential diagnosis of Adenomatoid odontogenic tumor, Calcifying epithelial odontogenic tumor and Odontogenic keratocyst was made.

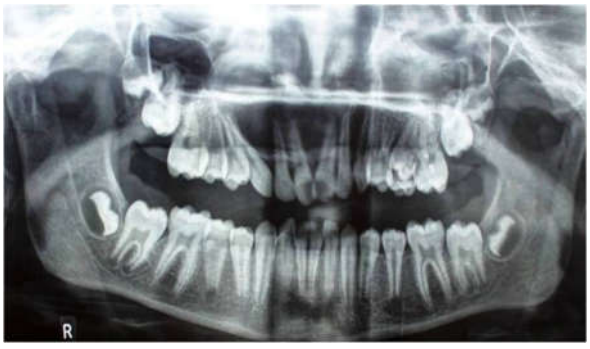
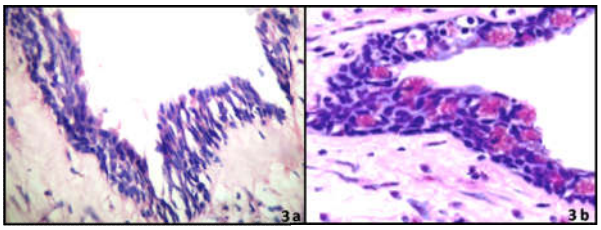


Figure 2 Panoramic radiograph showing a well defined unilocular radiolucency extending from maxillary left lateral incisor to right second premolar

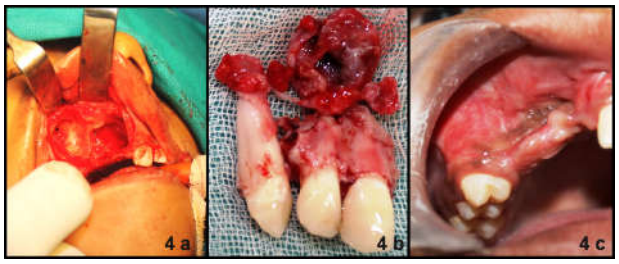
For further investigations, the incisional biopsy specimen was taken and sent for histopathological examination. The histopathological examination of the biopsy sample reveals cystic cavity lined by non keratinized stratified squamous epithelium surrounded by fibrous connective tissue wall. The epithelium connective tissue junction was flat and the stroma was devoid of inflammatory infiltrate. The epithelium exhibited variable thickness (plaque formation) containing vacuolated basal cells and areas of glandular differentiation were seen. (Figure 3a) The epithelial lining consisted of columnar and cuboidal cells having mucous cells with filiform extensions of the cytoplasm at few places. Few cholesterol clefts were also appreciated in stroma. Periodic acid Schiff (PAS) staining was performed and several PAS-positive mucous cells, clear cells and pseudo-glandular structures were seen in the epithelial lining. (Figure 3b) The histopathological diagnosis of Glandular odontogenic cyst was made.



3a. Photomicrograph showing variable thickness of epithelial lining with mucous cells and clear cells.
3b. PAS-positive mucous cells (pink stained) and clear cells.

Correlating history, clinical examination, radiographic findings and histopathological details, diagnosis of Glandular odontogenic cyst was made and final treatment was planned.

Under general anaesthesia, mucoperiosteal incision was given and the flap was raised. The cyst was enucleated and the bony cavity was fixed with Carnoy's solution. 11, 12 and 13 were extracted along with cyst. (Figure 4a,4b) Then flap was replaced and the wound was closed primarily. On seventh day, sutures were removed and healing was uneventful. Further prosthetic rehabilitation for esthetic purpose and function was done. The patient was recalled regularly for follow-up examination. (Figure 4c) The patient was clinically and radiologically examined and till now no sign of recurrence was observed.



4a. Intraoperative view after excision of the lesion
4b. Enucleated cyst along with extracted 11,12 and 13
4c. Postoperative area showing complete healing

DISCUSSION

Glandular odontogenic cyst (GOC) is an uncommon jaw bone cyst of odontogenic origin. Clinically, the most common site of occurrence is the mandible anterior region. GOC occurs over a wide age range primarily in middle- aged individuals and has a predilection for men [1], but in our case, patient was 14 years old and lesion involved anterior maxilla crossing the midline which is a rare site for GOC.

The lesion is generally painless, slow growing and its size can vary from less than 1 cm in diameter to large dimensions. [1] Small cysts may be asymptomatic, while large ones can cause bone expansion accompanied by pain and paresthesia. In our case, patient was asymptomatic with swelling due to slow bone expansion of approximately 3cm x 4cm in size. An association with impacted teeth, resorption and tooth displacement is common. In our case, root resorption was not present but displacement of roots was present along with displaced maxillary sinus.

Radiologically, these cysts may present as unilocular or multilocular radiolucent lesion, usually with well-defined borders. [3] In our case, a well-defined unilocular radiolucency was present with sclerotic border. The microscopic features show a cystic cavity lined with non-keratinized stratified squamous epithelium with localized plaque-like thickenings of the epithelium. Variable numbers of mucous-secreting cells in the surface layer of the epithelium, eosinophilic cuboidal or columnar cells that may be ciliated, intraepithelial gland like structures and the absence of inflammation in the subepithelial stroma may be seen[7,8]. In our case, variable thickness of epithelium and PAS-positive mucous cells, clear cells and some pseudo glandular structures along the epithelial lining were seen.

The histogenesis of GOC is still controversial. Kaplan *et al*. (2008) stated that "due to similarities in microscopic characteristics between GOC and lesions such as botryoid cyst, radicular and dentigerous cysts with mucous metaplasia and low-grade mucoepidermoid carcinoma (MEC), a definitive diagnosis can be difficult to make.[3]

According to Toida *et al*. (1994) it is important to differentiate GOC from central mucoepidermoid carcinoma (MEC), particularly the low-grade and predominantly cystic type. [9] Although, no definite separation can be made between central low-grade MEC and GOC, but immunopositivity of the epithelial mucous cells favors a diagnosis of central MEC.[7] Table 1

Table: 1 Immunohistochemical aspects (Oliveria et al, 2009)

CK 7+	Surface layer epithelium
CK 8+	Surface, suprabasal layer
CK 10+	Suprabasal and plaque
CK 14+	Suprabasal, basal and plaque
CK 19+	All layers

Note: CK- Cytokeratin

Kaplan *et al* have divided certain histological characteristic of GOC into major and minor categories.[10] Table 2

The histological features of the present case fulfilled some of the histopathological criteria for GOC like variable thickness (plaque formation), glandular differentiation, vacoulated basal cells flat epithelium connective tissue junction etc and thus final diagnosis was established.

Table 2- Histological characteristic of GOC (Kaplan et al, 2005)

Major criteria	Minor criteria
1.Squamous epithelial lining, flat interface	1.Papillary projections
2.Variations in thickness of the lining with or without epithelial "spheres" or "whorl", no palisades	2.Ciliated cells
3.Cuboidal eosinophilic cells or "hob-nail' cells	3.Multicystic or multiluminal architecture
4.Mucous "goblet" cells with interepithelial mucous pools with or without crypts lined by mucous-producing cells	4.Clear or vaculated cells in basal or spinous layer
5.Interepithelial glandular microcystic or duct like structures	

Treatment recommendations for GOC in the literature vary from enucleation, marsupialization, curettage with adjuvant Carnoy solution to a more aggressive approach including marginal resection or partial jaw resection. Several authors have suggested for aggressive approach to be a more reliable treatment due to a tendency of the cyst to recur. [2] In our case, enucleation followed by chemical cauterization using Carnoy's solution was done with extraction of 11,12 and 13. Prosthetic rehabilitation for function and esthetics was done. The present case showed no sign of recurrence after a year of follow-up.

CONCLUSION

In conclusion, considering the aggressive biological behavior and high recurrence rate of GOC, careful evaluation and regular radiological follow-up are necessary. The present case showed no sign of recurrence after a year of follow-up. Though GOC is rare in maxilla, still the possibility of such cyst cannot be ruled out while making differential diagnosis. It is important to consider both radiological and pathological findings for the final diagnosis.

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