

A vacillating swelling in maxillary anterior region- Diagnostic challenges and management

ABSTRACT

In most of the cystic lesions appearing in the oral cavity, proper diagnosis and treatment remains a challenge. As glandular odontogenic cyst (GOC) is a rare type of developmental odontogenic cyst, it was not included in our differential diagnosis. The report of GOC was quite surprising as it was rare and never came in this discussion. A 25-year-old male patient was reported to our center with a chief complaint of painless swelling in the right anterior region of the maxilla. The swelling was oval shaped and diffuse extending from the distal aspect of 11 to the mesial aspect of 14 and superiorly extending into the sulcus. Working diagnosis was a periapical cyst, so conventional treatment of root canal treatment, cyst enucleation, and apicoectomy was done. This article presents a rare case which was primarily misdiagnosed and treated and has been subsequently revealed histopathologically as a rare clinicopathological entity.

Keywords: Cystic swelling, glandular odontogenic cyst, maxillary swelling, odontogenic cyst, palatal swelling

INTRODUCTION

In most of the cases, cysts of the oral cavity pose a challenge in its diagnosis. This article presents a case of cystic swelling in the maxillary anterior region that was primarily misdiagnosed and treated as a periapical cyst and on histopathologic examination found to be a rare case of glandular odontogenic cyst (GOC). As GOC is a rare type of developmental odontogenic cyst, it was not included in our differential diagnosis. GOC was not considered in the initial differential diagnosis due to the extreme rarity of its appearance in the anterior maxillary region. GOC is a rare, benign cystic lesion that occurs predominantly in the anterior region of the mandible. Here, a rare case of GOC is presented along with its differential diagnosis, clinical, radiological, histopathological features, and management.

CASE REPORT

A 25-year-old male patient was referred to the oral and maxillofacial surgery department with a chief complaint of painless swelling in the right anterior region of the maxilla. Clinical examination revealed a swelling at the mucogingival junction approximately 1.5 cm × 2 × cm 1.8 cm in size, in

relation to the periapical and lateral region of 11, 12, and 13. The swelling was oval shaped and diffuse extending from the distal aspect of 11 to the mesial aspect of 14 and superiorly extending into the sulcus [Figure 1].


The swelling was not fluctuant. History revealed that the patient noticed an asymptomatic swelling 11 months back, which gradually increased in size. Intraoral periapical radiograph revealed a unilocular radiolucency, extending between the roots of right central, lateral incisor, and canine with well-defined sclerotic borders [Figure 2]. There was resorption of roots of the right central incisor.

VINOD NAIR SREEKUMAR, SHIAD SALIM, SANGEETH K CHERIAN¹, RICHIE JOHN²

Departments of Oral and Maxillofacial Surgery, ¹Prosthodontics, PMS College of Dental Science and Research, ²General Practitioner, Thiruvananthapuram, Kerala, India

Address for correspondence: Dr. Vinod Nair Sreekumar, Department of Oral and Maxillofacial Surgery, PMS Dental College, Venkode, Vattapara, Thiruvananthapuram - 695 028, Kerala, India. E-mail: vinodnair145@gmail.com

Received: 14 November 2018, **Revised:** 04 January 2019, **Accepted:** 10 April 2019, **Published:** 16 December 2020

Access this article online	
Website: www.njms.in	Quick Response Code 
DOI: 10.4103/njms.NJMS_84_18	

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Sreekumar VN, Salim S, Cherian SK, John R. A vacillating swelling in maxillary anterior region- Diagnostic challenges and management. Natl J Maxillofac Surg 2020;11:292-7.

The involved teeth 11, 12, and 13 were nonvital, and there was a discoloration in relation to 11. No incidence of trauma was reported, and involved teeth were not mobile. Mucosa over the swelling was slightly bluish in color and no associated palatal swelling was seen. Multiplanar cone-beam computed tomography (CBCT) was obtained using NewTom GIANO Scanner (Field of vision 5 cm × 5 cm) with 1-mm thin slices.

Radiographically a well defined mildly expansile homogenous periapical radiolucency with loss of sclerotic borders of size 16.4mm mediolaterally X 16.2mm superoinferiorly X 15.8mm anteroposteriorly in relation to 11,12,13 and 14 region. It extends from mesial aspect of 11 extending distally till the middle third of 14 [Figure 3].

CBCT gives an impression of infected periapical cyst in relation to 11 and 12 involving the mesial third and middle third of 13 with labial and palatal cortical plate fenestration with respect to 11 and 12 and thinning of the nasal floor in relation to 13. There is associated external root resorption of 11.

Differential diagnosis

Differential diagnosis of a well-circumscribed, asymptomatic, radiolucent lesion of the right maxillary anterior region on the basis of its position, clinical features, and radiological features comprises several pathologies, including globulomaxillary cyst (GMC), keratocystic odontogenic tumor (KCOT), adenomatoid odontogenic tumor, squamous odontogenic tumor, dentinogenic ghost cell tumor, and periapical cyst.^[1]

KCOT is an odontogenic tumor which is aggressive in nature and is having a high recurrence rate. The World Health Organization Classification of the Head and Neck Tumors in 2005 reclassified odontogenic keratocyst (OKC) from a cystic to a neoplastic lesion and renamed as “keratocystic odontogenic tumor” (KCOT).^[1] The KCOT contributes approximately 11% of cysts of the jaws and is most commonly located in the mandibular ramus region in the third molar region. Hence, KCOT was added to our differential diagnosis.

Adenomatoid odontogenic tumor has a remarkable tendency to occur in the anterior portion of the jaws, commonly in the maxilla than in the mandible. It has a striking tendency to occur in the anterior portions of the jaws and is found twice as often in the maxilla as in the mandible. They are relatively small and seldom exceed 3 cm in the greatest diameter. Peripheral form occurs as sessile masses usually of small size on the facial gingiva of the maxilla. Clinically, it is difficult to differentiate adenomatoid odontogenic tumor from



Figure 1: Intraoral photograph showing swelling extending from distal aspect of 11 to mesial aspect of 13



Figure 2: Intraoral periapical interpreting unilocular radiolucency with external root resorption of 11



Figure 3: Cone-beam computed tomography showing well-defined mildly expansile homogeneous periapical radiolucency with loss of sclerotic border

gingival fibrous lesions. They are frequently asymptomatic and are discovered during the course of routine radiographic examination. Less often, the adenomatoid odontogenic

tumor is a well-delineated unilocular radiolucency that is not related to an unerupted tooth but rather is located between the roots of erupted teeth.^[1]

Squamous odontogenic tumor is haphazardly distributed throughout the alveolar process of both maxilla and mandible, found in age groups ranging from 8 to 74 years. It has no sex or site predilection. It appears as a painless or slightly painful gingival swelling along with the mobility of involved teeth. Radiographic finding is not specific or diagnostic and consists of a triangular radiolucent defect lateral to the root or roots of teeth. In some instances, this suggests vertical periodontal bone loss. The radiolucent area may be somewhat ill-defined or may show a well-defined sclerotic margin. It seldom exceeds 1.5 cm in diameter.^[1]

Since this cystic lesion was asymptomatic and the involved teeth were nonvital, presumptive diagnosis of periapical cyst was also made. The tooth from which the periapical cyst originates usually does not respond to thermal and electric pulp testing. There will be a loss of lamina dura along the adjacent root, and a rounded radiolucency encircles the affected tooth apex. Root resorption is common. Significant growth is possible, and lesions occupying an entire quadrant have been noted. Root canal therapy is advised on those nonvital teeth.^[1]

The GMC was thought to be a fissural cyst originating from epithelial inclusions at the line of fusion between the medial nasal process and the maxillary process.^[2] GMC has a developmental origin. GMC was classically seen between the roots of maxillary lateral incisor and cuspid teeth, although occasionally GMC has been reported between the central incisors and lateral incisors.^[3] Classical radiographic appearance of GMC is a well-circumscribed unilocular, inverted pear- or tear-shaped radiolucency between the teeth. As the lesion expands, tipping of the tooth roots may occur. Now, the so-called GMC, a fissural cyst which is caused by entrapped epithelium between the nasal and maxillary processes, is no longer considered for its own entity.^[4] Because a fissural cyst in this region probably does not exist, the term GMC is no longer used. When a radiolucency between maxillary lateral incisor and canine is encountered, the clinician should first consider an odontogenic origin for the lesion.^[1]

Management

Although we have many differential diagnoses, our working diagnosis was a periapical cyst, so conventional treatment of root canal treatment, cyst enucleation, and apicoectomy was planned. With proper anesthesia, a two-sided full-thickness

mucoperiosteal flap was elevated within the area from tooth 14 to 21 [Figure 4a]. The lesion was identified in relation to 11, 12, and 13. Clinically labial cortical plate overlying the lesion was perforated at some levels. Once the overlying bone was removed, cystic lining was evident. Following cystic enucleation, surgical bed demonstrated bone loss at the mesial margin of 13 [Figure 4b]. Later, root apical end resection and retrograde filling were done in relation to 11, 12, and 13 [Figure 5]. The excised specimen was sent for histopathological examination, and suturing of surgical site was done [Figure 6].

Diagnosis

Histopathological findings of the excised soft-tissue section show a cystic lumen lined by nonkeratinized epithelium of varying thickness with a flat epithelial-connective tissue interface. Epithelium exhibits pseudostratified columnar appearance with areas of plaque such as thickening. Superficial cells of the epithelium are either cuboidal or columnar with some showing filiform extensions of cytoplasm. Within the pseudostratified ciliated columnar epithelium, there were goblet cells. Intraepithelial microcysts surrounded by cuboidal cells and containing eosinophilic material are seen [Figure 7]. Underlying connective tissue is moderately collagenous with focal collection of chronic inflammatory cell infiltrate and hemorrhage. Periodic acid–Schiff staining also showed microcysts and goblet cells [Figure 8]. The histopathologic picture gave an impression of GOC. Differentiation of GOC from other odontogenic cysts with mucous prosoplasia is quite challenging for a pathologist, as the presence of mucous-producing cells and duct-like structures is not an infrequent feature of GOC. It is thus of immense importance to differentiate between the two since the treatment and prognostic interference varies. Immunohistochemical staining was done with CK19 and Ki67. Ki67 showed negative/<1% staining [Figure 9]. The epithelium showed strong positivity for CK19 [Figure 9]. The asymptomatic swelling was diagnosed as GOC.

DISCUSSION

As GOC is a rare and recently recognized type of developmental odontogenic cyst, it was not included in our differential diagnosis. The report of GOC was quite surprising as it was rare and never came in this discussion. Although it is accepted as of being odontogenic origin, because of the pluripotentiality of the odontogenic epithelium, it can show glandular or salivary features.^[1] The GOC has two clinically important attributes: it has “a high recurrence rate”^[5] and it displays “an aggressive growth potential.”^[6]

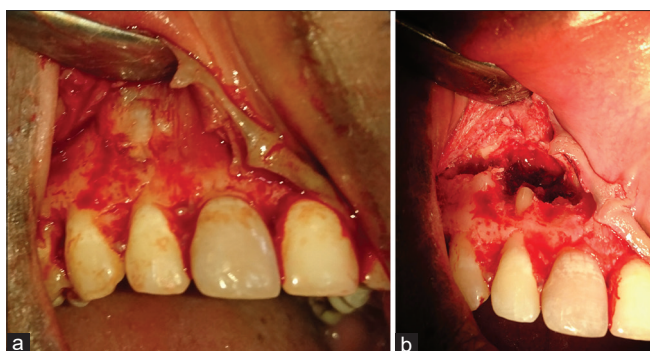


Figure 4: (a) The lesion was identified in relation to 11, 12, and 13. (b) Surgical bed



Figure 5: Retrograde filling



Figure 6: Postoperative sutured site

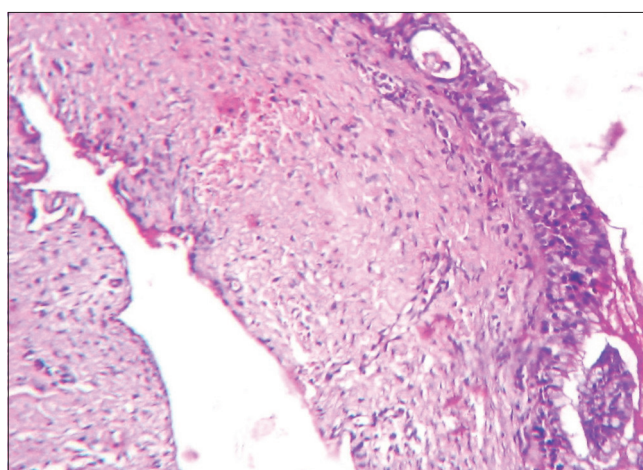


Figure 7: Intraepithelial microcysts (H and E, $\times 10$)

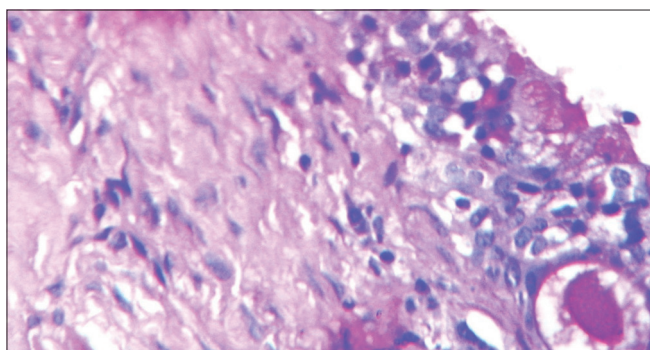


Figure 8: PAS-positive epithelium showing goblet cells and microcysts (PAS, $\times 40$)

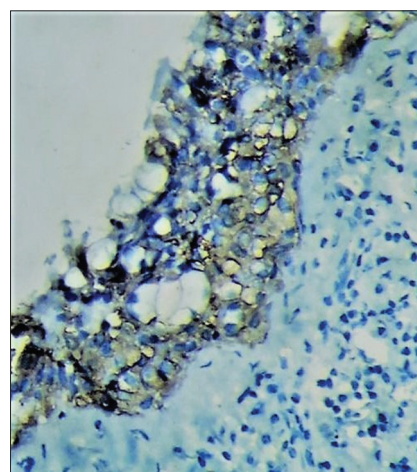


Figure 9: CK, 19×40

GOC was first suggested by Padayachee and Van Wyk in 1987 by reporting two cases that were similar to the botryoid odontogenic cyst but with a gland element and suggested the name “sialo-odontogenic cyst.”^[7] Later, it was described in detail by Gardner *et al.* in 1988.^[8] High *et al.* in 1996 described the polymorphous nature of GOC and coined it as a polymorphous odontogenic cyst.^[9] This cyst was recently listed by the World Health Organization as a developmental odontogenic epithelial cyst. The GOC occurs commonly in middle-aged adults with a mean age of onset of 49 years at

the time of diagnosis; rarely does it occur before the age of 20 years. GOC has a preference for the anterior regions, especially in maxillary lesions. The size of the cyst can vary from small lesions < 1 cm in diameter to large destructive lesions that may involve most of the jaws. Small cysts may

be asymptomatic; however, large cysts often produce clinical expansion, which sometimes can be associated with pain or paresthesia.

Between 1977 and 1995, Magnusson *et al.*^[10] analyzed 5,800 biopsies of jaw cysts and observed that only seven cases fulfilled GOC criteria, which comprised 0.012% of the total. In 2012, Araújo de Moraes *et al.* stated that approximately 114 cases of GOC have been reported in the literature by 2010.^[11]

Radiographically, the lesion may appear as a unilocular, or more commonly, a multilocular radiolucency with a well-defined sclerotic rim or as a perifollicular radiolucency. Radiographic findings of this lesion play a major role in diagnosis as there is a lack of consistency in the clinical manifestations and the intraosseous development of these lesions. Gardner *et al.* proposed the histopathological characteristics of GOC,^[8] and Kaplan *et al.* proposed a list of microscopic criteria for GOC that includes nonkeratinized stratified squamous epithelium, eosinophilic cuboidal or columnar cells, epithelial whorls or spheres within the lining, which are occasionally ciliated, and presence of mucous cells with microcystic areas.^[12] The subepithelial connective tissue is usually free of inflammation. This case satisfied all the characteristic features of GOC. The recurrence rate of GOC ranges between 21% and 55%.^[5] The high rate of recurrence shows the aggressive nature of GOC. The aggressive nature of GOC might be associated with cell kinetics in the lining epithelium.^[12]

There are several articles in which root canal treatment has been done due to misdiagnosis, on a preassumption that it was a periapical cyst. Similarly, we also did root canal treatment (RCT) in the presumption that it is a periapical cyst.^[8]

Retrospectively, we feel that GOC is underdiagnosed because the strict criteria put forwarded by Gardener are not followed. Moreover, GOC is always mismanaged as in our case because we initially thought it was a periapical cyst. Similar reports are there, where GOC was mismanaged as a periapical cyst.^[9]

Different treatment modalities of GOC have been recommended. Ficarra *et al.* proposed the treatment of GOC as complete enucleation and fixation of the surrounding bone with Carnoy's solution.^[13] According to Hussain *et al.*, it was local *en bloc* excision with primary reconstruction because of its aggressive nature and tendency for recurrence.^[14] Bhatt *et al.* proposed that conservative treatment is enough for GOC.^[15] However, in our case, the diagnosis of GOC comes only in excisional biopsy. Hence, we had done only a

conservative enucleation. Since there are conflicting reports in the literature, regarding the treatment modality, and since only a few cases are reported in the world literature, we have opted to observe the patient periodically.

Mascitti *et al.*^[16] reported a 19.8% recurrence rate, but it was contradicted by Fowler *et al.*^[17] in their study who showed a 50% recurrence rate and suggested that a long-term follow-up is required. In this case, he is on routine follow-up, and on 12-month review, there is no evidence of any recurrence.

CONCLUSION

Hence, this is a unique case which appeared as a radiolucent lesion in relation to 11, 12, and 13 which was initially misdiagnosed and mismanaged as a periapical cyst and on histopathologic examination found to be a rare case of GOC.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Neville B, Douglas D, Allen CM, Bouquot J. Oral Maxillofacial Pathology. 3rd ed. Philadelphia: Saunders; 2009.
2. Shear M. Developmental odontogenic cysts. An update. J Oral Pathol Med 1994;23:1-11.
3. Shear M. Cysts of the jaws: Recent advances. J Oral Pathol 1985;14:43-59.
4. Dammer U, Driemel O, Mohren W, Giedl C, Reichert TE. Globulomaxillary cysts – Do they really exist? Clin Oral Investig 2014;18:239-46.
5. Boffano P, Cassarino E, Zavattoni E, Campisi P, Garzino-Demo P. Surgical treatment of glandular odontogenic cysts. J Craniofac Surg 2010;21:776-80.
6. Macdonald-Jankowski DS. Glandular odontogenic cyst: Systematic review. Dentomaxillofac Radiol 2010;39:127-39.
7. Padayachee A, Van Wyk CW. Two cystic lesions with features of both the botryoid odontogenic cyst and the central mucoepidermoid tumour: Sialo-odontogenic cyst? J Oral Pathol 1987;16:499-504.
8. Gardner DG, Kessler HP, Morency R, Schaffner DL. The glandular odontogenic cyst: An apparent entity. J Oral Pathol 1988;17:359-66.
9. High AS, Main DM, Khoo SP, Pedlar J, Hume WJ. The polymorphous odontogenic cyst. J Oral Pathol Med 1996;25:25-31.
10. Magnusson B, Göransson L, Odesjö B, Gröndahl K, Hirsch JM. Glandular odontogenic cyst. Report of seven cases. Dentomaxillofac Radiol 1997;26:26-31.
11. Araújo de Moraes HH, José de Holanda Vasconcellos R, de Santana Santos T, Guedes Queiroz LM, Dantas da Silveira ÉJ. Glandular odontogenic cyst: Case report and review of diagnostic criteria. J Craniomaxillofac Surg 2012;40:e46-50.
12. Kaplan I, Gal G, Anavi Y, Manor R, Calderon S. Glandular odontogenic cyst: Treatment and recurrence. J Oral Maxillofac Surg 2005;63:435-41.
13. Ficarra G, Chou L, Panzoni E. Glandular odontogenic

- cyst (sialo-odontogenic cyst). A case report. *Int J Oral Maxillofac Surg* 1990;19:331-3.
14. Hussain K, Edmondson HD, Browne RM. Glandular odontogenic cysts. Diagnosis and treatment. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995;79:593-602.
 15. Bhatt V, Monaghan A, Brown AM, Rippin JW. Does the glandular odontogenic cyst require aggressive management? *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;92:249-51.
 16. Mascitti M, Santarelli A, Sabatucci A, Procaccini M, Lo Muzio L, Zizzi A, *et al.* Glandular odontogenic cyst: Review of literature and report of a new case with cytokeratin-19 expression. *Open Dent J* 2014;8:1-12.
 17. Fowler CB, Brannon RB, Kessler HP, Castle JT, Kahn MA. Glandular odontogenic cyst: Analysis of 46 cases with special emphasis on microscopic criteria for diagnosis. *Head Neck Pathol* 2011;5:364-75.