

Glandular odontogenic cyst associated with ameloblastoma occupying maxillary sinus: A rare case report

Riaz Abdulla, Vagish Kumar Laxman Shanbhag¹, Surendranathan Akhila, Maji Jose

Department of Oral Pathology and Microbiology, Yenepoya Dental College and Hospital, Yenepoya University, ¹Department of Oral Medicine and Radiology, Yenepoya Dental College and Hospital, Yenepoya Research Centre, Yenepoya University, Mangalore, Karnataka, India

ABSTRACT

The glandular odontogenic cyst (GOC) is a rare entity with around 111 cases available in the literature. The occurrence of GOC with ameloblastoma as a collision tumor is rarest, and such a case poses problems in the form of diagnostic dilemma and management. The present case report describes and discusses a rare case of GOC with ameloblastomatous component occurring in right maxilla of a 54-year-old male patient with a chief complaint of painful swelling.

Key words: Collision tumor, desmoplastic ameloblastoma, glandular odontogenic cyst, hobnail cells, maxillary sinus

INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare developmental epithelial cyst of the jaws.^[1] It has tendency to occur in anterior mandible with male predilection.^[2,3] Middle-aged patients are frequently affected with mean age of occurrence at 49.5 years. GOC presents aggressive behavior, has tendency to recur and can be mistaken for central mucoepidermoid carcinoma.^[3] Therefore, early diagnosis and recognition of GOC are important. Ameloblastoma is a common tumor of odontogenic epithelium, especially of enamel organ type tissue that has not undergone differentiation to the point of hard tissue formation.^[4,5] It is slow-growing, locally invasive, and exhibits variety of histopathological patterns.^[4,5] The present case report describes a rare case of GOC associated with ameloblastomatous component occurring in right maxilla.

Address for correspondence: Dr. Vagish Kumar Laxman Shanbhag, Department of Oral Medicine and Radiology, Yenepoya Dental College and Hospital, Yenepoya Research Centre, Yenepoya University, University Road, Mangalore - 575 018, Karnataka, India.
E-mail: vagishkumar_12@rediffmail.com

CASE REPORT

A 54-year-old male patient reported with a chief complaint of painful swelling in the right maxillary area since 6 months. Pain was mainly felt in the premolar and molar region. Swelling gradually increased in size. Local examination showed a swelling in the right maxillary quadrant extending from the 11–14, and its surface appeared smooth on palatal side. On palpation, the mass was of 3 cm × 2 cm size, firm in consistency, mildly tender, and fluctuation was absent. Hemogram and other biochemical tests were within normal limits. Cone beam computed tomography (CT) of the lesion was advised. The axial, coronal, and sagittal section in the medium field of view showed a homogenous expansile radiopaque mass within the right maxillary antrum obliterating the sinus [Figure 1]. Erosion of anterior wall with perforation at 13 and 16 regions was observed. Posterior and lateral wall of the sinus was intact. Medial wall of the antrum was thinned out. Orbital floor was intact and thinning of palatal bone with respect to 16 was observed. Based on clinical and radiological examination, a provisional

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

Cite this article as: Abdulla R, Shanbhag VL, Akhila S, Jose M. Glandular odontogenic cyst associated with ameloblastoma occupying maxillary sinus: A rare case report. Clin Cancer Investig J 2016;5:265-8.

Access this article online

Quick Response Code:



Website:

www.ccij-online.org

DOI:

10.4103/2278-0513.182064

diagnosis of ameloblastoma was arrived. Incisional Biopsy was taken from location 11, 12, 13, and 14 regions and sent for histopathological examination. Histopathology with hematoxylin and eosin staining (H and E) showed numerous islands of odontogenic epithelium in a dense collagenous connective tissue stroma. Some odontogenic islands were compressed by the densely collagenous connective tissue stroma and few islands resembled open follicles. The follicles were occasionally lined by peripheral cuboidal cells and showed hypercellular central area with spindle-shaped cells. The connective tissue showed few calcifications and extravasated red blood cells [Figure 2]. Histopathologic diagnosis was desmoplastic ameloblastoma. Correlating with clinical, radiological, and histopathological features, a working diagnosis of desmoplastic ameloblastoma (DA) was arrived. Patient was then referred to competent oral surgeon for surgical treatment. Surgical treatment was carried out, and the tumor was excised with curettage of marginal bone. The excised surgical mass (3 cm × 2 cm) was sent for histopathological examination. Histopathology with H and E showed cystic cavity lined by nonkeratinized stratified squamous epithelium with upper layer composed of ciliated cuboidal cells (hobnail cells) with plaque-like epithelial thickening in some areas. Intraepithelial crypt containing mucous pools were present in the lining of the cystic epithelium [Figures 3 and 4]. The mucous goblet cells were present both singly and in small clusters within the lining epithelium. The underlying connective tissue showed moderate infiltration of chronic inflammatory cells and blood vessels. The section also showed well-encapsulated mass with cystic component and ameloblastomatous-like islands in a dense fibrocollagenous background [Figure 5]. Histopathologic impression was GOC associated with ameloblastoma. Correlating with clinical, radiological, and histopathologic findings, a final diagnosis of GOC associated with DA was made. Thoughtful analysis also suggests the possibility of coexistence of cyst and tumor, i.e., collision tumor.

DISCUSSION

In an analysis of 46 cases of GOC, 80% of cases occurred in mandible, 60% involved anterior jaws, and 71% cases occurred in the fifth to seventh decade.^[6] In our case, posterior maxilla was involved which is uncommon. GOC occurring in posterior part of maxillary sinus and manifesting in CT as cortical perforation on lateral aspect of maxilla in second molar region has been previously reported in the literature.^[7] Hobnail cells were detected in our case. Although hobnail cells are not specific for GOC, they are essential for diagnosis.^[6] Presence of mucous cells with microcysts, epithelial spheres, clear cells, and variable thickness of the cyst lining, occasionally ciliated eosinophilic columnar/cuboidal cells, and multiple compartments

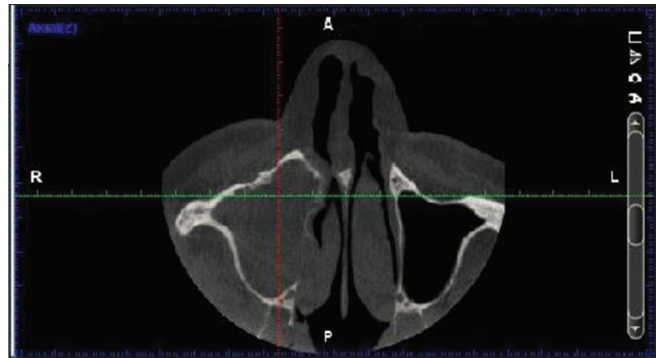


Figure 1: Cone beam computed tomography - axial view

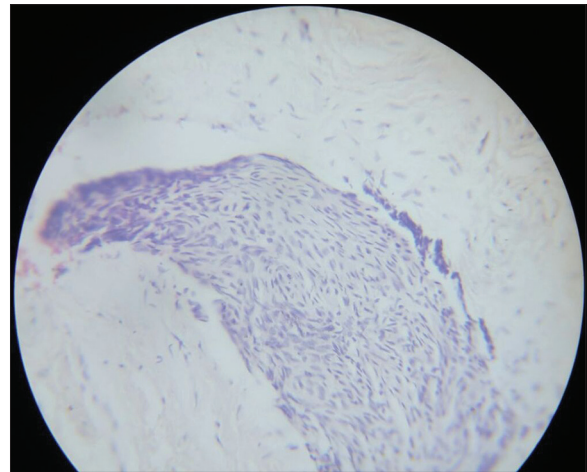


Figure 2: Desmoplastic ameloblastoma-histopathologic picture with H and E, ×40

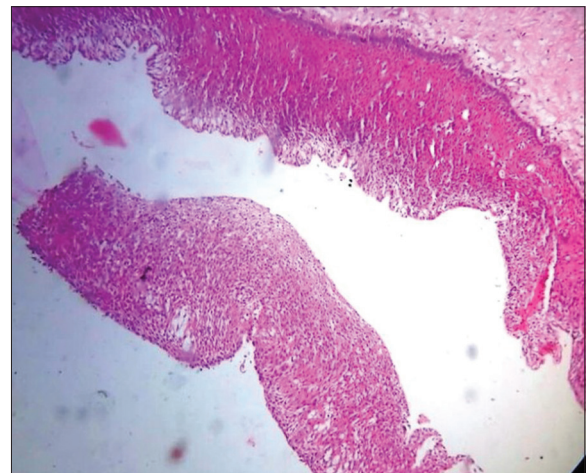


Figure 3: Ciliated cuboidal epithelium showing hobnail appearance (H and E, ×10)

help in differentiating GOC from GOC-like lesions.^[6,8] Principal clinical finding in GOC is nonspecific painless edema.^[7] In our case, the swelling was painful probably due to compression of nerves or secondary infection. GOC presents with unilocular or multilocular radiolucency with well-defined borders, and the diagnosis of cyst is not feasible through clinical and radiological examination alone.^[7,9] Differential diagnosis of GOC includes keratocysts, botryoid

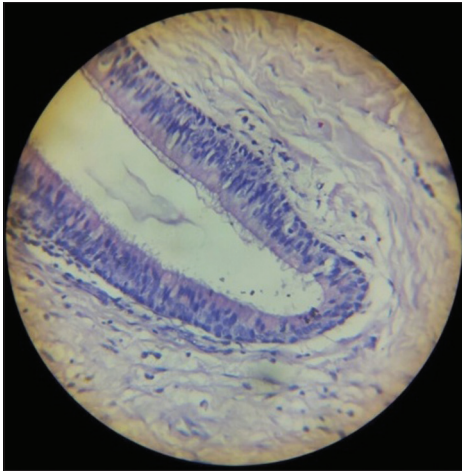


Figure 4: Hobnail cells ($\times 40$)

cysts, ameloblastoma, and central mucoepidermoid carcinoma.^[8]

DA commonly occurs in the region from anterior teeth to premolars in both maxilla and mandible and is usually 2 cm or less.^[10] DA accounts for 4–12% of all ameloblastomas.^[10] It affects both sexes equally between the age range of 20–80 years.^[11] It is also more aggressive than other types.^[5] Frequency of occurrence of DA is relatively higher in the Asian population, and initial chief complaint is painless swelling of the jaw bone.^[12] Histopathological characteristics of desmoplastic ameloblastoma include extensive stromal collagenization.^[5] Previously, a case of maxillary plexiform ameloblastoma manifesting as bulging of sinus floor, erosion of dental sockets and mucosal thickening in the left maxillary sinus in CT has been reported in the literature.^[13] In our case, the tumor entirely occupied the right maxillary sinus which is very rare. Furthermore, the presence of mucous cells within the lining epithelium was seen in this case which is also a very rare finding in tumors-related to ameloblastoma.^[14] The presence of mucous cells was confirmed by periodic acid-Schiff stain.

The case is unique in the aspect of coexistence of GOC and ameloblastoma as a collision tumor. The presence of mucous cells with ciliated epithelium correlated with GOC. Findings of island of ameloblastomatous components in dense collagenous background correlated with ameloblastoma. It is possible that ameloblastic cells in the epithelium morphed to mucous ciliated cells due to inflammatory stimuli leading to the origin of GOC associated ameloblastoma (collision tumor).^[15] CT plays an important role in accurately diagnosing ameloblastoma, especially when occurring in maxilla. CT helps to evaluate the extent, internal structure of the lesion and also assess the state of adjacent structures in three dimensions. Coronal section in CT image helps to assess the extent of the lesion into maxillary sinus and nasal cavity.^[16] Since CT is superior

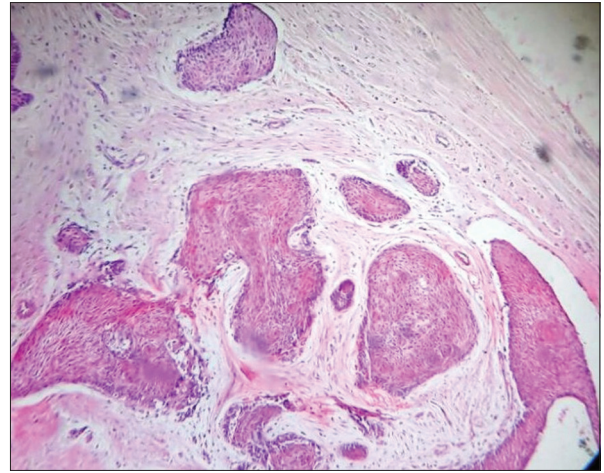


Figure 5: Odontogenic tumor islands in loose fibrous connective tissue stroma (H and E, $\times 10$)

to conventional radiographs with added advantages, occlusal radiograph is not required.

Surgical treatment varies from conservative approaches such as enucleation, curettage, and peripheral ostectomy to marginal and segmental resection.^[9] Conservative approach is correlated with recurrence rates of GOC.^[7] Recurrence in DA is due to ill-defined borders.^[5] Complete removal of the tumor with surrounding normal tissues is the treatment of choice. Wide removal of lateral wall of nasal cavity is recommended to facilitate frequent inspection and detection of recurrence of the tumor in maxillary sinus.^[10]

CONCLUSION

Occurrence of GOC associated ameloblastoma is rare. Both desmoplastic ameloblastoma and GOC are aggressive, and recurrence rate is high. Hence, care should be taken to recognize this rare entity. Early diagnosis and intervention will lead to good prognosis. The present case represents ameloblastoma transforming into GOC or another possibility of GOC coexisting with ameloblastoma (collision tumor). This case report demonstrates that the occurrence of coexistence of aggressive lesions is a possibility and care should be taken to consider this entity during diagnosis and treatment planning.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Oliveira JX, Santos KC, Nunes FD, Hiraki KR, Sales MA, Cavalcanti MG, et al. Odontogenic glandular cyst: A case report. *J Oral Sci* 2009;51:467-70.

2. Raju SP, Reddy SP, Ananthnag J. Glandular odontogenic cyst of the anterior mandible. *N Am J Med Sci* 2015;7:65-9.
3. Tambawala SS, Karjodkar FR, Yadav A, Sansare K, Sontakke S. Glandular odontogenic cyst: A case report. *Imaging Sci Dent* 2014;44:75-9.
4. Jansari TR, Samanta ST, Trivedi PP, Shah MJ. Granular cell ameloblastoma of mandible. *Indian J Pathol Microbiol* 2014;57:305-7.
5. Masthan KM, Anitha N, Krupaa J, Manikkam S. Ameloblastoma. *J Pharm Bioallied Sci* 2015;7 Suppl 1:S167-70.
6. 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.
7. Rao JB, Jeevan Kumar KA, Kumar BP. Glandular odontogenic cyst involving the posterior part of maxillary sinus, a rare entity. *J Maxillofac Oral Surg* 2010;9:72-5.
8. Shah M, Kale H, Ranginwala A, Patel G. Glandular odontogenic cyst: A rare entity. *J Oral Maxillofac Pathol* 2014;18:89-92.
9. Salehinejad J, Saghafi S, Zare-Mahmoodabadi R, Ghazi N, Kermani H. Glandular odontogenic cyst of the posterior maxilla. *Arch Iran Med* 2011;14:416-8.
10. Kato H, Nomura J, Matsumura Y, Tagawa T. A case of desmoplastic ameloblastoma occupying maxillary sinus. *Contemp Clin Dent* 2011;2:234-6.
11. Nair PP, Bhat GR, Neelakantan S, Chatterjee R. Desmoplastic ameloblastoma of mandible. *BMJ Case Rep* 2013;2013. pii: Bcr2013200082.
12. Belgaumi UI, Sundaresh KJ, Varma S, Mallikarjuna R. Desmoplastic ameloblastoma: A rare odontogenic neoplasm with unusual radiographic and histomorphological presentation. *BMJ Case Rep* 2013;2013. pii: Bcr2013009079.
13. Dwivedi N, Raj V, Chandra S, Agarwal A. Maxillary ameloblastoma extending into the maxillary sinus. *Eur J Gen Dent* 2013;2:182-6.
14. Punnya AV, Rekha K. "Ameloblastoma with mucous cells": Review of literature and presentation of 2 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;106:e20-6.
15. Hisatomi M, Asaumi J, Konouchi H, Yanagi Y, Kishi K. A case of glandular odontogenic cyst associated with ameloblastoma: Correlation of diagnostic imaging with histopathological features. *Dentomaxillofac Radiol* 2000;29:249-53.
16. Misra A, Misra D, Rai S, Panjwani S, Ranjan V, Prabhat M, *et al.* Computed tomography in diagnosis of admantinoma. *N Am J Med Sci* 2015;7:229-32.