Glandular odontogenic cyst of posterior maxilla: A rare entity

LiFeng Li, Pradeep Singh, Ji Ping, Xian Li

ABSTRACT

Introduction: Glandular odontogenic cyst (GOC) is uncommon jaw cyst of odontogenic origin with unpredictable and potentially aggressive behavior. It is a rare developmental cyst with relatively low frequency of just 0.012–0.03%. Very limited cases of GOC have been reported in literature since it was first described by Gardner et al. in 1988. GOC is found to occur in fifth decade of life and the most common site of occurrence is mandible, especially the mandibular anterior region with slight predilection for males. However, its occurrence in the maxillary posterior region of a 23-year-old female with associated symptoms of pain is quite rare.

Case Report: This case report presents one such rare case of GOC in right maxillary region of a 23-year-old female who was primarily diagnosed as a radicular cyst. Later, due to recurrence of the lesion, patient had to undergo enucleation and partial resection of posterior maxilla, and after a comprehensive histopathological analysis, it was finally diagnosed as GOC.

Conclusion: In conclusion, Glandular odontogenic cyst being such a rare entity, this paper may enhance the existing knowledge about GOC and may guide readers and clinicians to pay special attention to similar cases when encountered in clinical work.

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CASE REPORT

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Keywords: Cytokeratin, Glandular odontogenic cyst (GOC), Ki-67, p53, Radicular cyst

How to cite this article

Li L, Singh P, Ping J, Li X. Glandular odontogenic cyst of posterior maxilla: A rare entity. Int J Case Rep Imag 2016;7(4):254–260.

Article ID: Z01201604CR10632LL

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doi:10.5348/ijcri-201644-CR-10632

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Received: 09 December 2015

Accepted: 05 January 2016

Published: 01 April 2016

INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare developmental cyst of odontogenic origin. Padayachee and Van Wyk initially reported it as a sialo odontogenic cyst in 1987 [1] but its odontogenic origin was first described by Gardner et al. in 1988 who also proposed the name 'GOC' [2]. The term 'polymorphous odontogenic cyst' for this cyst was proposed by High et al. in (1996), because of its aggressive growth pattern [3]. Sadeghib in 1991 used the term mucoepidermoid cyst or mucous producing cyst due to the microscopic findings of mucus producing cells and squamous cells [4]. Moreover, WHO

histological typing of odontogenic tumors also includes glandular odontogenic cyst under the terms, GOC or Sialo odontogenic cyst. Magnusson et al. observed that only 0.012% of the cysts seen on the oral cavity have fulfilled the criteria of GOC microscopically [5].

The GOC is a rare developmental cyst with relatively low frequency of 0.012–0.03% and prevalence rate 0.17%. ‘High recurrence rate’ and an ‘aggressive growth potential’ [6] are considered to be the most important clinical characteristics of GOC. The following three possibilities can be attributed to the development of GOC:

(a) Odontogenic primordial origin, wherein epithelial lining undergoes prosoplasia into glandular epithelium.

(b) As a low-grade mucoepidermoid carcinoma that appears as a single cystic space as opposed to usual multicystic spaces.

(c) As a true cyst of glandular origin, in which the entrapped salivary gland primordia or undifferentiated primitive epithelial rests differentiate into glandular epithelium [7].

Clinically, the most common site of occurrence is mandibular anterior region [8], where it presents as a slow growing intraosseous lesion. The GOC occurs primarily in middle-aged patients (mean age of 49.5 years) [9, 10] with slight male predilection [11]. Radiographically, the lesion can be described as a unilocular or, more commonly, multilocular radiolucency with well-defined sclerotic margins [12]. Histologically, GOC is characterized by a non-keratinized stratified squamous epithelial lining with papillary projections, focal plaque like thickenings within the lining, microcysts or intraepithelial crypts containing mucin, ‘mucous lakes’ and hyaline bodies. It also includes cuboidal basal cells, sometimes vacuolated and absence of inflammation in the subepithelial connective tissue [12, 13]. The relative rarity of this lesion is the reason behind presentation of this case. In this paper, we present one such unique case of a GOC and its clinic-histopathological features.

CASE REPORT

A 23-year-old female patient presented to the department of oral and maxillofacial surgery, with the chief complaint of pain in the right upper back tooth region of jaw since four months. Pain was mild and recurrent in nature. There were no associated symptoms apart from mild tenderness of the maxillary right buccal region. Meticulous dental history with patient revealed that he had a previous history of swelling and intermittent pain in the same region five years ago, for which she was diagnosed with radicular cyst in 16, and had to undergo surgery for enucleation of the cyst. One year post operatively, patient underwent extraction of upper right first molar at some other dental facility, because of continuous pain. Four months ago she underwent root canal therapy for 17, at some other dental facility. Past medical, and family history were noncontributory and there was no previous history of trauma.

Clinical examination

Extra-orally there was no apparent facial asymmetry. On intra-oral examination any significant mass and swelling was non-apparent, except mild tenderness of the maxillary right posterior buccal region, extending from 15 to 18 tooth region. The associated teeth were tested vital. Cystic fluid was reddish in color on needle aspiration. There were no obvious signs of infection, and limitation of mouth opening. Preoperatively panoramic radiograph was taken and postoperatively histologic examination was done.

Radiological examination

Panoramic radiograph revealed a large oval well-circumscribed multilocular radiolucent lesion measuring around 3.00x1.80 cm, located under the maxillary sinus region and extending from the root apices of 15 to 18 tooth region (Figures 1 and 2). 3D cone beam computed tomography (CBCT) reconstruction image showed perforations of the cortical bone extending from the periapical region of 15 to 18 (Figure 3).

Initial Histological examination

Microscopic examination of H&E stained section of the specimen revealed non-keratinized stratified squamous epithelial lining of varying thickness with

Figure 1: Initial preoperative panoramic radiograph (five years ago) showing 3.00x1.80 cm oval well-circumscribed multilocular radiolucent lesion (arrow) near the root apices of 15 to 18.

Figure 2 : Recent preoperative panoramic radiograph showing cystic lesion near the root apices of 15 to 18, extraction socket of 16, and R.C.T treated 17 can be well appreciated.

epithelial hyperplasia (irregular proliferation and elongation). However, some mucus gland-like cells were also observed within some selective areas which were not so typical (Figure 4A–B). The nature, anatomical location, radiographic findings, and histopathological findings of the lesion were compatible with the diagnosis of radicular cyst. Ameloblastoma and odontogenic keratocyst were considered for the differential diagnosis of the same.

Current Operative Procedure

Considering the age and subjective requirements, and with patient's full consent, it was finally decided to treat the case with enucleation and block resection of the maxilla, simultaneously extracting 15, 17 and 18. The surgery was performed under general anesthesia and during the surgery some thick cystic lining was found which could be easily enucleated from the bony cavity (Figures 5 and 6). In order to further elucidate the nature of the lesion and to provide final diagnosis, we carried out an incisional biopsy and a part of cystic lining was excised through the perforations of cortical bone and the specimen was subjected to histopathological and immunohistochemical examination.

Recent Histopathological examination

Microscopic examination of the specimen showed, glandular structures lined by mucous cells within the non-keratinized squamous epithelium. Spinous cell layer appeared as vacuoles (Figure 7A–B). No single mucinous cell was observed in the epithelium. No significant signs of mucoepidermoid carcinoma were seen in the sections examined.

Immunohistochemical examination

To assess the proliferate nature of the cyst in the present case, an immunohistochemistry (IHC) staining was done using Ki-67, p53 and cytokeratin (CK) was

found to be positive for p53 (Figure 8A), Ki-67 (Figure 8B) and CK (Figure 9). However, the expression of CK was moderately positive in the basal layer and slightly positive in the parabasal and surface layers and duct forming cells.

Histopathological and immunohistochemical examination findings were indicative of 'glandular odontogenic cyst' thus a final diagnosis of GOC was

Figure 4: (A, B) Photomicrograph of the initial specimen showing non-keratinized stratified squamous epithelial lining of varying thickness with epithelial hyperplasia. Some mucus gland-like cells can also be seen in selective areas (Magnification Figure 4A: H&E stain, x100, Figure 4B: H&E stain, x200).

Figure 5 : Intraoperative image showing cystic enucleation.

Figure 6 : Enucleated specimen from the bone cavity.

Figure 3: Recent preoperative 3D CBCT reconstruction image showing perforations of the cortical bone extending from the periapical region of 15 to 18.

Figure 7 (A, B) : Photomicrograph of the specimen (recent) showing non-keratinized squamous epithelium and glandular structures, lined by mucous cells. Spinous layer cell appeared as vacuoles. (Magnification: Figure 7A: H&E stain, x100, Figure 7B: H&E stain, x200).

Figure 8 (A, B): Immunohistochemical findings for p53 and Ki-67 in the lining epithelia of Glandular odontogenic cyst of the maxilla. Photomicrograph of immunohistochemically stained (A) p53 and (B) Ki-67 showing scattered positive cells in GOC lining (IHC stain, x 200).

Figure 9: Immunohistochemical findings for cytokeratin in the lining epithelia of glandular odontogenic cyst of the maxilla. Photomicrograph showing positive immunoreactions to CK protein (IHC stain, x200). The CK was positive in the whole layer of the lining epithelia.

Figure 10: Postoperative panoramic image after partial maxillary resection.

made. Postoperative recovery was uneventful (Figures 10 and 11). Patient is currently subjected under a long-term follow-up. One year follow-up showed improved bone healing with no signs of relapse.

DISCUSSION

The GOC is a rare entity with relatively low frequency of 0.012–0.03% [5] and prevalence rate 0.17%. Magnusson et al. in their study evaluated 5900 cases of jaw bone cysts and found only seven cases of GOC, i.e., about 0.12% [5]. In another similar study by Van Heerden et al., only 1.3% cases were reported [14]. Literature review showed that GOC may mimic a wide clinicopathologic spectrum ranging from LPC to a destructive malignant neoplasm such as central mucoepidermoid carcinoma (CMEC) [15].

As there are very few cases of GOC reported in English literature, its incidence, clinical manifestations, radiological findings, histological features, treatment and prognosis has no unified opinion. Clinically, GOC is often manifested as a slow growing painless mass, few accompanied by pain, paresthesia and numbness. The lesion may cause pain due to compression of a neurovascular bundle or secondary infection [16]. In the present case the patient is a middle-aged female with recurrent pain in right maxillary posterior region. However existing literature reports, slight male predilection and, mandibular anterior being the most common site of occurrence. In this case, the recurrent pain the patient complained of is considered to be due to compression of the posterior superior alveolar nerve by the lesion.

Lack of consistency in the clinical manifestations, and the intraosseous development of these lesions, and similarities with various other intrabony pathologies demonstrate the importance of radiographical and histopathological evaluation. Radiographically, GOC

Figure 11: 3D cone beam computed tomography (CBCT) reconstruction image after partial maxillary resection.

may appear as intraosseously localized, multilocular or unilocular radiolucent lesion with well-defined borders. In some instances it may also present scalloped, and peripherally osteosclerotic borders, together with root resorption and displacement of the teeth. In the presented case, radiographic examination shows unilocular multilocular radiolucencies with well-defined margins. It is often diagnosed as an odontogenic cyst or a tumor because this disease has no special characteristics.

Histologically the presented case consisted of certain characteristic features of GOC like non-keratinized squamous epithelium varying in thickness and cuboidal or ciliated epithelium with mucus-producing cells on the surface. Owing to the fact that lateral periodontal cyst (LPC) and CMEC exhibit substantial overlap between histological features, their histopathological differentiation becomes difficult and must be performed with considerable care. In particular the differentiation of low-grade CMEC from GOC is more important and difficult. However, the identification of intraepithelial microcysts or duct-like structures, epithelial whorls, ciliated cells, and superficial cuboidal cells in GOC differentiates it from low-grade CMEC [17]. Likewise, the absence of duct like spaces with mucous cells and ciliated epithelium in the histological sections of LPC, favors the diagnosis of GOC [18].

While some authors believe the distinction between GOC and central mucoepidermoid carcinoma depends largely on the degree of epithelial proliferation [19] others have recommended the use of immunohistochemical markers to distinguish these two diseases [20]. Immunostaining with Ki-67, p53, CK-19 and their positivity in GOC may help in differentiating GOC from Mucoepidermoid carcinoma (MEC). Certain studies have reported an increased Ki-67 index and decreased P53 positivity suggesting that GOC lining displays increased proliferation, but not malignant transformation potential. Kaplan et al. found that GOC showed lower p53 immunoreactivity but significantly higher Ki-67 proliferative index than MEC [21]. Besides, cell kinetics in the lining epithelium might be associated with the tendency for recurrence and aggressive nature of GOC. Furthermore, Tosios et al. demonstrated increased Bcl2 (an anti-apoptotic protein) in their study and suggested dysregulation of cell death in lining epithelium to be associated with the biological behavior of GOC [22].

Several treatment options including curettage, enucleation, en block resection and partial osteotomy are available for the treatment of GOC but the treatment of choice is still controversial. Another factor is, treatment by enucleation or curettage alone is associated with a high recurrence rate. Multicystic lesions treated by curettage or enucleation demonstrated increased recurrence rate of 55% with an average of 4.9 years [23]. In this case, en block resection was considered to be the treatment of choice in order to cure the disease and avoid further surgery. The cyst has an aggressive behavior and a high recurrence rate, so follow-up during three to five years

should be carried out.

Reviewing the histologic examination of radicular cyst, we can find some mucus gland-like cells within some selective areas, although these cells are not so typical. Analyzing the clinical features, radiological and histological examination of the two lesions (GOC and Radicular cyst), it is questionable to diagnose whether this is actually a GOC evolving from a radicular cyst or just a new lesion arising from the same area. Review of literature has shown that in one of the case reports GOC presented as dentigerous cyst. At this point, both alternatives are possible and more case reports and studies should be encouraged to support these possibilities.

CONCLUSION

In conclusion, glandular odontogenic cyst (GDC) being such a rare entity, this paper may enhance the existing knowledge about GOC and may guide readers and clinicians to pay special attention to similar cases when encountered in clinical work. Mucus gland-like cells found in the histological examination of radicular cysts should draw particular attention to the lesion, whether leading to recurrence or evolving to GOC. Close follow-up must be put in schedule.

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Author Contributions

LiFeng Li – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Pradeep Singh – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Ji Ping – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Xian Li – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

The authors Li LiFeng and Singh Pradeep contributed equally to this case report, and the authors Li LiFeng and Singh Pradeep should be regarded as first joint authors.

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REFERENCES

1. 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 Nov;16(10):499–504.
2. Gardner DG, Kessler HP, Morency R, Schaffner DL. The glandular odontogenic cyst: an apparent entity. *J Oral Pathol* 1988 Sep;17(8):359–66.
3. High AS, Main DM, Khoo SP, Pedlar J, Hume WJ. The polymorphous odontogenic cyst. *J Oral Pathol Med* 1996 Jan;25(1):25–31.
4. Lin CC, Chen CH, Lai S, et al. Glandular odontogenic cyst: a case report. *Kaohsiung J Med Sci* 2000 Jan;16(1):53–8.
5. Magnusson B, Göransson L, Odesjö B, Gröndahl K, Hirsch JM. Glandular odontogenic cyst. Report of seven cases. *Dentomaxillofac Radiol* 1997 Jan;26(1):26–31.
6. Shen J, Fan M, Chen X, Wang S, Wang L, Li Y. Glandular odontogenic cyst in China: report of 12 cases and immunohistochemical study. *J Oral Pathol Med* 2006 Mar;35(3):175–82.
7. Mark RE, Stern D. Oral and maxillofacial pathology. A rationale for diagnosis and treatment. 2ed. New Delhi: Quintessence Publishing co, Inc; 2012.
8. Shafer WG, Hine MK, Levy BM. Shafer's textbook of Oral Pathology. 5ed. India: Elsevier; 2005.
9. Manor R, Anavi Y, Kaplan I, Calderon S. Radiological features of glandular odontogenic cyst. *Dentomaxillofac Radiol* 2003 Mar;32(2):73–9.
10. Purohit S, Shah V, Bhakhar V, Harsh A. Glandular odontogenic cyst in maxilla: A case report and literature review. *J Oral Maxillofac Pathol* 2014 May;18(2):320–3.
11. van Heerden WF, Raubenheimer EJ, Turner ML. Glandular odontogenic cyst. *Head Neck* 1992 Jul-Aug;14(4):316–20.
12. Shah M, Kale H, Ranginwala A, Patel G. Glandular odontogenic cyst: A rare entity. *J Oral Maxillofac Pathol* 2014 Jan;18(1):89–92.
13. Manzini M, Deon C, Corte LD, Bertotto JC, Abreu LB. Glandular odontogenic cyst: an uncommon entity. *Braz J Otorhinolaryngol* 2009 Mar-Apr;75(2):320.
14. Osny FJ, Azevedo LR, Sant'Ana E, Lara VS. Glandular odontogenic cyst: case report and review of the literature. *Quintessence Int* 2004 May;35(5):385–9.
15. Kasaboglu O, Basal Z, Usubütün A. Glandular odontogenic cyst presenting as a dentigerous cyst: a case report. *J Oral Maxillofac Surg* 2006 Apr;64(4):731–3.
16. Prabhu S, Rekha K, Kumar G. Glandular odontogenic cyst mimicking central mucoepidermoid carcinoma. *J Oral Maxillofac Pathol* 2010 Jan;14(1):12–5.
17. Shear M. Cysts of the Oral Region. 3ed. Mumbai: Varghese Publishing House; 1992.
18. Waldron CA, Koh ML. Central mucoepidermoid carcinoma of the jaws: report of four cases with analysis of the literature and discussion of the relationship to mucoepidermoid, sialodontogenic, and glandular odontogenic cysts. *J Oral Maxillofac Surg* 1990 Aug;48(8):871–7.
19. Chung GC, Han WJ, Kim EK. A huge glandular odontogenic cyst occurring at posterior mandible. *Korean J Oral Maxillofac Radiol* 2004;34(4):209–13.
20. Kaplan I, Anavi Y, Manor R, Sulkes J, Calderon S. The use of molecular markers as an aid in the diagnosis of glandular odontogenic cyst. *Oral Oncol* 2005 Oct;41(9):895–902.
21. Tosios KI, Kakarantza-Angelopoulou E, Kapranos N. Immunohistochemical study of bcl-2 protein, Ki-67 antigen and p53 protein in epithelium of glandular odontogenic cysts and dentigerous cysts. *J Oral Pathol Med* 2000 Mar;29(3):139–44.
22. Hussain K, Edmondson HD, Browne RM. Glandular odontogenic cysts. Diagnosis and treatment. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995 May;79(5):593–602.
23. Thor A, Warfvinge G, Fernandes R. The course of a long-standing glandular odontogenic cyst: marginal resection and reconstruction with particulated bone graft, platelet-rich plasma, and additional vertical alveolar distraction. *J Oral Maxillofac Surg* 2006 Jul;64(7):1121–8.

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