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Review Article

Glandular odontogenic cyst – Literature review and report of a paediatric case

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ABSTRACT

Glandular odontogenic cyst (GOC) is an extremely rare lesion occurring in the jawbones. The present paper is a review of 181 cases of GOCs reported in English literature, since it was first reported by Padayache and Van Wyk in 1987. Mandible was involved in 130 cases and maxilla in 51 cases. Anterior mandible was the most common area of involvement. Radiographic appearance was that of a unilocular radiolucency in 98 of 176 reported cases. Rest presented as multilocular radiolucency. Cortical expansion was observed in 136 of the 180 reported cases while cortex breach or perforation was seen in 81 cases. The treatment of choice was that of minor procedures that included enucleation with or without curettage, peripheral ostectomy, cryotherapy, etc. in 157 of the total 177 reported cases. Marginal jaw resection, segmental mandibulectomy etc. was reported in 20 cases. Although minor surgical procedures were the treatment of choice in most studies, two major studies of Kaplan *et al.* and Fowler *et al.* involving 111 and 46 cases, recorded a recurrence rate of 35.9 and 19.6%, respectively. The age range was between 11 and 82 years. The respective mean age of patients in the above mentioned studies was 45.7 for Kaplan's and 51 years for Fowler's whereas in our study, the mean age was 45.9 years. Very rarely does GOC presents itself in a paediatric patient. The paper also reports a case of an 11-year-old child whose histopathology came out to be a case of a GOC.

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1. Introduction

Glandular odontogenic cyst is an uncommon cyst of the jaw bone with only about 181 cases described till date, since it was first described by Padayache and Van Wyk¹ in 1987. They described two multilocular cystic lesions with histological features similar to that of botryoid odontogenic cyst with the

lining epithelium containing areas of cuboidal or columnar epithelium and scattered foci of mucous cells and termed it as “sialo-odontogenic cyst”. A year later Gardener *et al.*² described eight cases with similar histological features and suggested the term Glandular Odontogenic Cyst. In 1992, the second edition of the World Health Organization's (WHO) histological classification of odontogenic tumours recognized it as “a cyst arising in the tooth-bearing areas of the jaws and

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characterized by an epithelial lining with cuboidal or columnar cells both at the surface and lining crypts or cyst-like spaces within the thickness of the epithelium", and retained the term glandular odontogenic cyst (Fig. 1).

Mean Age of appearance at diagnosis is in the middle-aged adults 45–50 years of age. Few cases have been reported in the second decade of life.

Radiographic appearance is nonspecific. Radiographically the lesion may appear as unilocular or multilocular radiolucency, usually with well-defined margins and scalloped border. The scalloping also occurs interdentally and at interradicular areas (Figs. 2–4).

Clinically, the most common site is the mandible, especially in the anterior region. The cyst commonly presents as a slow growing intraosseous lesion in the anterior mandible frequently crossing the midline. Small cysts may be asymptomatic; however, large cysts often produce clinical expansion, which sometimes can be associated with pain and rarely paresthesia may be seen. Clinical and histological diagnoses are significant as GOC have shown a tendency towards recurrence similar to the odontogenic keratocyst, and its resemblance microscopically with the central mucoepidermoid carcinoma (CMEC) so much that few authors doubt the rarity of this entity considering historically reported cases of very low grade CMEC to be actually as GOC's. Kaplan et al.³ in their review of GOC's in English literature reported 111 cases in 20 years since it was first described. The present paper is a review of 181 cases reported till date, which includes published case series and reports since 2008 and literature from the Asian and the Indian subcontinent as well. We also report a case of highly aggressive GOC in a paediatric patient 11 years of age, extensively involving the anterior and posterior mandible. Keeping in mind the clinical history at first presentation of reported cases of GOC's, this seems to be the only reported case of a GOC that probably developed in the first decade of life.

Methodology: A literature review was performed using the search terms "glandular odontogenic cysts", "sialo-odontogenic cyst" on the Pub Med interface. Further literature search was identified from the reference list of the papers accessed via *Pub med*, *Springer online*, *Wiley online* and by searching the

Fig. 1 – Frontal view.

Fig. 2 – Orthopantomogram showing multilocular cyst.

Fig. 3 – Coronal section showing dimension of cyst in premolar regionst.

Fig. 4 – Extent of cyst from left molar to rt second premolar.

World Wide Web. The search included abstracts, books and online publications from year 1987 to 2015.

2. Review of literature

Sl no	Study title	No of cases	Gender and age	Location	Radiographic characteristics	Cortical plate integrity	Treatment	Recurrence	Follow up
1	Kaplan et al. ³	111	1.3:1, 14–75 Mean 45.7	Mand-70% Max-30% Ant-24.1% Post-25.3% Ant + post-50.6%	Unilocular-53.8% Multilocular-46.2%	Expansion (E)-88.5% Perforation (P)-61%	Curettage/enucleation-83.5% Resection 17%	35.9% 0%	Mean 2.7 years
2	Manzini et al. ⁴	1	27/F	Ant. Max.	Uniloc.	E- P-	Enucleation	Not reported	Not reported
3	Anuthama	2	42/F	Ant mand	Multilocular	E+	En bloc resection	NR	2 years
4	Krishnamurthy et al. ⁵	21/M	21/M	A + p mand	Multilocular	P+	En bloc resection	NR	9 months
5	Jefferson X. Oliveira et al. ⁷	1	54/F	Ant mand	Multilocular	E+,P+	Curettage	Recur	5 years
6	Mariana Camilo	1	37/M	Ant + post mand	Multilocular	Not recorded	Curettage + peripheral ostectomy	NR	5 years
7	Negreiros Lyrio et al. ¹²	1	47/F	Post max	Unilocular	E+	Enucleation	NR	5 years
8	Prabhu et al. ¹³	2	68/M 82/F	Post mand Post mand	Multilocular Unilocular	E+ E+	CURRETAGE Enbloc excision.	NR NR	2 years 6 years
9	C. Fowler et al. ¹⁰	46	Mean age 51 years. M:F = 1:1	36-mand Ant mand-19 Max-9 7 ant maxilla	Unilocular-27 Multilocular-14 5 cases-nr	E+20 cases P+3 cases	Enucleation/curretage	19.6%	8.75 years
10	J. Salehinejad et al. ¹⁵	1	28/M	Post max	Unilocular	E+	Enucleation + peripheral ostectomy	NR	2 years
11	Cano et al. ¹⁶	2	54/M 63/F	Post mand Post mand	Multiloc. Multiloc	E+ E+	Curettage Marsupialization	NR NR	3 years 3 years
12	Korkmaz et al. ¹⁷	1	52/M	mand	Multilocular	E	Curettage	NR	6 years
13	Amberkar et al. ¹⁸	1	29/M	Post Maxilla	Uniloc	E+	NR	NR	nr
14	Guruprasad et al. ¹⁹	1	17/F	max	Unilocular	E+ P-	Enucleation with curretage	NR	1 year
15	Araújo de Moraes et al. ²⁰	1	56/M	Post mand	Unilocular	E+ P+	Enucleation+ curretage	NR	8 months
16	Pornpop Rattana-arpha et al. ²¹	1	49/F	Ant mand	Unilocular	E+ P+	Enucleation with curretage	NR	18 months
17	M. Mascitti et al. ²²	1	28/F	Ant mand	Unilocular	E- P-	Enucleation	NR	18 months
18	Fatih ASUTAY et al. ²³	1	38/F	Ant max	Unilocular	E+	Enucleation	NR	17 months
19	Shahnaz S. Tambawala et al. ²⁴	1	17/F	Post mand	Multilocular	E+ P+	Curretage	NR	3 months
20	David Booth et al. ²⁵	1	36/M	Post mand	Multilocular	E+ P+	Enucleation	NR	6 months
21	Kumaraswamy Naik et al. ²⁶	1	14/M	Ant + post mand	Multilocular	E+ P+	NR	NR	nr
22	Byung-Do Lee et al. ²⁷	1	78/F	Post mand	Unilocular	E+ P+	Enucleation with peripheral ostectomy	NR	–
23	Monali Shah et al. ²⁸	1	40/M	Ant + post mand	Multilocular	E+ P+	Enucleation	Not reported	nr
24	Present study	1	11/M	Ant + post mand	Multilocular	E+ P+	Enucleation with curretage	NR	1 year

nr: not reported in literature.

3. Case report

An 11-year-old relatively healthy male child presented to the hospital with a complaint of painless swelling on his chin and lower anterior jaw bone. The swelling had a 3-month history of presentation and had slowly increased in size. Patient denied any significant medical history or family history. Extra oral examination revealed slight facial asymmetry with slight expansion of both buccal and lingual cortices in the anterior mandible. Neither cervical lymphadenopathy nor sign of inflammation was noted. The expansion was more pronounced on facial surface than on lingual surface. The overlying oral mucosa was intact. The right central incisors, lateral incisors, and lower anterior teeth had grade 1 mobility.

Radiographic presentation: Panoramic radiograph revealed 6×4 cm, multilocular radiolucency with corticated, scalloped border. The main lesion extended from roots of premolar on the right side to distal root of molar on left side. Cone beam CT scan revealed well demarcated multilocular lesion with a thinned out lower border of mandible. A large, hypodense, and multilocular lesion with cystic pattern and well defined borders, extending from the right side second premolar to the midline and extending beyond the mental foramen and first molar of the left side. The inferior limits ranging from the interdental areas of teeth, alveolar processes and to the mandibular base (Figs. 2–4). Severe buccal expansion and thin septa within the lesion were noted. In some areas, the buccal cortical bone was perforated. No involvement of soft tissues was depicted clinically and radio graphically.

Histopathological examination After an incisional biopsy revealed nonkeratinized stratified squamous epithelium lining with a flat interface with subsequent stroma. The epithelium shows plaque-like thickenings and whirling pattern of squamous epithelial cells. Other areas of epithelium showed pseudo-glandular pattern with few ciliated and mucus cells. Histopathology was suggestive of glandular odontogenic cyst (Figs. 7 and 8).

Treatment: A month later, complete enucleation and subsequent curettage were performed under local anaesthesia. Very thick cystic lining was removed from the cavity. Primary closure was done (Figs. 5 and 6). Specimen was sent for

Fig. 5 – Surgical enucleation.

Fig. 6 – Thick lining removed from multiple locules of the cyst.

histopathology, which again revealed glandular odontogenic cyst. One-year follow-up the case has revealed no recurrence.

4. Results

4.1. Demographics

The mean age at diagnosis for all cases ($N = 181$) was 45.9 years. The age range was between 11 and 86 years. Most of the cases were diagnosed in the 4th, 5th and 6th decades. 17% i.e. 4/23 cases in reported after 2008 were in the 2nd decade of life. The case in the present study was diagnosed at age of 11 years and going by the clinical history and first presentation the lesion appears to have developed in the first decade. No sex predilection was seen ($n = 71$, cases reported after 2008).

4.2. Location

Of the 181 cases reported 130 were in the mandible and 51 in the maxilla. Anterior jaw bone was twice more commonly involved than the posterior jaw. (117:56).

4.3. Radiographic appearance

Radiographic features were defined in 176 out of the 181 cases. Unilocular radiolucency was seen in 98 cases ($n = 176$) and

Fig. 7 – Parakeratinized squamous epithelial lining exhibiting cuboidal.

Fig. 8 – Lining epithelium.

multilocular appearance was observed in 78 cases ($n = 177$). Five cases in one study had not defined the locularity.

4.4. Cortical plate integrity

Swelling and expansion of the cortical bone were seen as a predominant feature. Of the 180 cases that recorded the cortex expansion, 136 cases had positive expansion of cortical plates. Cortical plate breach or perforation was seen in 81 cases.

Treatment: Out of 177 cases that reported the treatment modality, 157 cases reported conservative management with minor procedures including enucleation with or without curettage, peripheral ostectomy, chemical cauterization with carnoys solution, cryotherapy, etc. Major procedures (marginal resection/partial jaw resection) were reported in 20 cases.

Follow-up: The mean follow up period was 2.2 years. The range was 0.25–8.75 years. The present case had a follow-up period of 1 year.

Recurrence: Recurrence was reported in 50 of the 179 cases that reported recurrence.

5. Discussion

The GOC is a rare lesion, with a frequency rate ranging from 0.12% to 0.13% of all jaw cysts and has a high recurrence potential. Mascitti et al.²² reported 111 cases of GOC with majority of cases in 4th and 5th decades of life which was coincident with that of MacDonald-Jankowski.⁸ However their study reported the age at presentation of GOCs in the East Asian and sub-Saharan African population was nearly a decade younger. According to Fowler et al.¹⁰ the mean age at diagnosis was 51 years with a range of 20–82 years ($N = 45$). 32 (71.1%) of his cases were diagnosed in the 5th, 6th, and 7th decades.

According to Kaplan et al.^{3,6} GOCs occur within a wide age range of 14–75 years, but has never been reported in children <10 years of age. Literature survey reveals very few cases in second decade of life and none in first decade of life. The case reported here is of an 11-year-old child. Only one case of an 11-year-old child has been reported earlier in literature. The growth pattern of the present case and comparative study of literature of the previous studies have made us conclude that in the present case the GOC had developed when the child was less than ten years of age in the first decade of life.

GOC has a clear preference for the mandible, with 71.8% of cases in this region and only 28% in the maxilla which is similar to other studies. The anterior mandible was the most common area of involvement. The involvement of anterior jaws both anterior mandible and anterior maxilla was twice that of the posterior jaws.

Radio graphically, the lesions typically presented as a radiolucent unilocular or multilocular lesion. Unilocularity of the cyst was the more common presentation with 56% cases. This was similar to other reviews of Kaplan et al. and Fowler et al.^{10,11}

Histologically the lesion revealed a lining of nonkeratinized stratified squamous epithelium with a flat interface with subsequent stroma. The epithelium shows plaque-like thickenings and whirling pattern of squamous epithelial cells. Other areas of epithelium showed pseudo-glandular pattern with few ciliated and mucus cells. Histopathology was suggestive of glandular odontogenic cyst as defined by Gardner et al.² In their review Kaplan et al.^{3,6} divided the microscopic characteristic of GOC into major and minor criteria, to facilitate its diagnosis. According to this author, a certain diagnosis of GOC could be made with the presence of all the major signs.

The major criteria include:

1. Squamous epithelial lining, with a flat interface with the connective tissue wall, lacking basal palisading.
2. Epithelium exhibiting variations in thickness along the cystic lining with or without epithelial “spheres” or “whorls” or focal luminal proliferation.
3. Cuboidal eosinophilic cells or “hob-nail” cells.
4. Mucous cells with intraepithelial mucous pools, with or without crypts lined by mucous-producing cells.
5. Intraepithelial glandular, microcystic or duct-like structures.

The minor criteria include:

1. Papillary proliferation of the lining epithelium.
2. Ciliated cells.
3. Multicystic or multiluminal architecture.
4. Clear or vacuolated cells in the basal or spinous layers

Two major studies of Kaplan et al.³ and Fowler et al.¹⁰ have reported a high recurrence rate following conservative surgical management (enucleation, curettage, etc.). According to Kaplan et al.³ the aggressive biologic behaviour and propensity for recurrence might be associated with cell kinetics in the lining epithelium, i.e. infoldings, microcysts and plaques, which are suggestive of active cell proliferation. These areas of epithelial thickening may be comparable to the proliferative changes seen in dental lamina underlying the odontogenic nature of the GOC. Fowler et al.,¹⁰ in their study of 46 cases, reported an average time period from initial treatment to first recurrence of 8 years. Since 2008, the now of cases reporting recurrence have been less with reported recurrence only in two studies of Jefferson et al. and Fowler et al.¹⁰. In cases prior to 2008, Kaplan³ had reported a recurrence rate of 35.9%. The main reason for a decrease in the recurrence in our study may be due to an inadequate follow up. The mean follow-up period of all cases was 2.2 years.

Conclusion: Rarity of the glandular odontogenic cyst makes it difficult to make a diagnosis and study the biologic behaviour. Few case studies with long-term follow-up have been reported. More than half of the published cases have been followed for periods less than 2 years and recurrences have been reported even 8 years postoperatively when treated conservatively. The principles of treatments are still conservative. With cases of GOCs as the one reported here, the glandular odontogenic cyst should be kept in differential diagnosis of very small children too and conservative management with a long-term follow-up up to 10 years should be undertaken.

Conflicts of interest

The authors have none to declare.

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;16(10):499.
2. Gardner DG, Kessler HP, Morency R, Schaffner DL. The glandular odontogenic cyst: an apparent entity. *J Oral Pathol.* 1988;17(8):359.
3. Kaplan I, Anavi Y, Hirshberg A. Glandular odontogenic cyst: a challenge in diagnosis and treatment. *Oral Dis.* 2008;14(7):575.
4. Manzini M, Deon C, Corte LD, Bertotto JC, Abreu LB. Glandular odontogenic cyst: an uncommon entity. *Braz J Otorhinolaryngol.* 2009;75(2):320.
5. Krishnamurthy A, Sherlin HJ, Ramalingam K, et al. Glandular odontogenic cyst: report of two cases and review of literature. *Head Neck Pathol.* 2009;3(2):153.
6. Kaplan I, Gal G, Anavi Y, Manor R, Calderon S. Glandular odontogenic cyst: treatment and recurrence. *J Oral Maxillofac Surg.* 2005;63(4):435.
7. Oliveira JX, Santos KCP, Nunes FD. *J Oral Sci.* 2009;51(3):467–470.
8. Macdonald-Jankowski DS. Glandular odontogenic cyst: systematic review. *Dentomaxillofac Radiol.* 2010;39(3):127.
10. C. Fowler, R. Brannon, OOOOE e30 Abstracts December 2010.
11. 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(4):364.
12. Lyrio MC, De Assis AF, Germano AR, De Moraes M. Treatment of mandibular glandular odontogenic cyst with immediate reconstruction: case report and 5-year follow-up. *Br J Oral Maxillofac Surg.* 2010;48:651–653.
13. Prabhu S, Rekha K u. Glandular odontogenic cyst mimicking central mucoepidermoid carcinoma. *J Oral Maxillofac Pathol.* 2010;14:12–15.
14. Boffano P, Cassarino E, Zavattero E, et al. Surgical treatment of glandular odontogenic cysts. *J Craniofac Surg.* 2010;21:776–780.
15. Salehinejad J, Saghaei S, Zare-Mahmoodabadi R, Ghazi N, Kermani H. Glandular odontogenic cyst of the posterior maxilla. *Arch Iran Med.* 2011;14(6):416.
16. Cano J, Benito DM, Montans J, Rodriguez-Vazquez JF, Campo J, Colmenero C. Glandular odontogenic cyst: two high-risk cases treated with conservative approaches. *J Craniomaxillofac Surg.* 2012;40(5):e131–e136.
17. Korkmaz YT, Hocaoglu TP, Baris E, Erkmen E. Surgical treatment of a rare mandibular glandular odontogenic cyst with 6-year follow up: a case report. *F U Sağ Bil Tı p Derg.* 2011;25:97–101.
18. Amberkar Vikram S, Jahagirdar Abhishek. Ahmed Mujib BR. Glandular odontogenic cyst: report of an unusual bilateral occurrence. 2011;22(2):364.
19. Guruprasad Y, Chauhan DS. Glandular odontogenic cyst of maxilla. *J Clin Imaging Sci.* 2011;1:54.
20. Araújo de Moraes HH, José de Holanda Vasconcellos R, de Santana Santos T, Guedes Queiroz LM, Dantas da Silveira É. Glandular odontogenic cyst: case report and review of diagnostic criteria. *J Craniomaxillofac Surg.* 2012;40(2):e46–e50.
21. Pornpop Rattana-arpha. Ploi Yongvanichit. *Int J Oral Maxillofacial Pathol.* 2013;4(2):34–39.
22. Mascitti M, Santarelli A, Di Fede O, Rubini C, Lo Muzio L. *Annali di Stomatologia.* (Suppl. 2):2013;(Suppl. 2):1–48. 29.
23. Asutay F, Acar AH, Yolcu Ü, Karadağ N, Geçör O. Glandular odontogenic cyst mimicking dentigerous cyst: a case report. Dentijeröz kisti taklit eden glandüler odontojenik kist: olgu sunumu. *J Istanbul Univ Fac Dent.* 2014;48(1):65–70.
24. Tambawala SS, Karjodkar FR, Archana Yadav. Kaustubh Sansare. Subodh Sontakke. *Imaging Sci Dentistry.* 2014;44:75–79.

1. Padayachee A, Van Wyk CW. Two cystic lesions with features of both the botryoid odontogenic cyst and the central

-
25. David Booth. Jerry Bouquot. Clinico-Pathologic Conference: Case 6. *Head Neck Pathol.* 2010;4:351–355.
 26. Kumaraswamy Naik LR, Agarwal R, Amberkar VS, Ahmed Mujib BR. A rare variant of ameloblastoma associated with a glandular odontogenic cyst. *Indian J Pathol Microbiol.* 2008;51:576–577.
 27. Byung-Do Lee. Wan Lee. Glandular odontogenic cyst mimicking ameloblastoma in a 78-year-old female: a case report. *Imaging Sci Dentistry.* 2014;44:249–252.
 28. Shah M, Kale H, Ranginwala A, Patel G. Glandular odontogenic cyst: a rare entity. *J Oral Maxillofac Pathol.* 2014;18:89–99.