

Recurrent glandular odontogenic cyst treatment

Reküren glandüler odontojenik kist tedavisi

Sibel Turalı, DDS,¹ Duygu Yazıcıoğlu, DDS, PhD,² Kutay Can Ergül, DDS,¹
Nazife Tuba Telcioğlu, DDS,³ Hakan Alpay Karasu, DDS., PhD.¹

¹Department of Oral and Maxillofacial Surgery, Ankara University, Faculty of Dentistry, Ankara, Turkey;

²Ankara University, Faculty of Dentistry, Dental Clinics, Ankara, Turkey;

³Samsun Oral and Dental Health Center Clinic, Samsun, Turkey

The glandular odontogenic cyst (GOC) is a rare odontogenic cyst. Although none of the clinical or radiographic features of GOC are unique or pathognomonic, the lesion has a potentially aggressive behavior. Treatment of GOC includes curettage and enucleation or excision and cryotherapy; however marginal resection is usually considered a more reliable approach due to the low incidence of recurrence. In this article, we present a case with GOC which recurred five years after the initial surgical treatment.

Key Words: Carnoy's solution; enucleation; glandular odontogenic cyst; recurrence.

Glandüler odontojenik kist (GOK), nadir bir odontojenik kisttir. Bu kistlerin klinik veya radyografik özellikleri benzersiz veya patognomik olmamasına rağmen, lezyon agresif bir tutum sergileme potansiyeline sahiptir. Glandüler odontojenik kistler küretaj ve enükleasyon veya eksizyon ve kriyoterapi ile tedavi edilmekle birlikte, düşük rekürens insidansı nedeniyle marjinal rezeksiyonun daha güvenilir bir yaklaşım olduğu düşünülmektedir. Bu yazıda, ilk cerrahi tedaviden beş yıl sonra tekrarlayan bir GOK olgusu sunuldu.

Anahtar Sözcükler: Carnoy çözümü; enükleasyon; glandüler odontojenik kist; nüks.

The glandular odontogenic cyst (GOC) is a rare odontogenic cyst, originally classified in 1988.^[1-10] According to the literature review these cysts are classified as "sialo-odontogenic cyst" due to the microscopic features.^[2-4] Glandular odontogenic cyst is the most-used term according to the World Health Organization (WHO), because the possible salivary gland origin of these cysts has not yet been established.^[5,11] The lesions are presumed to be odontogenic in origin, since they appear as radiolucencies on radiographs and are associated with jaw swellings and unique histopathological features.^[5,6]

Glandular odontogenic cyst is a rarely-seen pathology among the reported cases in the literature.^[3] The most common location is the mandible, especially the anterior region.^[3,4,6] It is seldom found in association with an unerupted tooth.^[6] Recurrence has been reported in about half of cases.^[4] Glandular odontogenic cyst occurs primarily in middle age.^[3,4] A slight predilection for males can be seen, with a female-to-male ratio of 19:28. An asymptomatic, slow-growing swelling is frequently seen.^[3]

Glandular odontogenic cyst has no specific or pathognomonic radiographic features. It



Figure 1. Preoperative view of the lesion.



Figure 2. Panoramic radiography of the lesion.

may be seen as a multilocular or a unilocular radiolucency or as a perifollicular radiolucency, simulating a hyperplastic follicle or dentigerous cyst or may occur as a separate and distinct cavity, possibly simulating selective odontogenic tumors and lesions of bone. Similarly, the borders may be well-defined and sclerotic or ill-defined.^[11] The histologic features of GOC include a thin layer of stratified squamous cell epithelium with or without surface cilia, glandular or pseudoglandular structures containing mucicarmine-positive material, and interspersed mucous cells.^[7,12] Glandular odontogenic cyst has been reported as a significant lesion by clinicians and pathologists due to two reasons. The first is the obvious overlap in histomorphological features between GOC, lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC), and central mucoepidermoid carcinoma (CMEC) of the jaws. The second is the potential aggressive behavior of the lesion.^[11] The number of typical features which are necessary for the diagnosis of GOC remains unclear, and there are no specific stains for the differential diagnosis of GOC. Although none of the clinical or radiographic features of GOC are unique or pathognomonic, the lesion has a potentially aggressive behavior.^[12] In certain features, treatment of GOC includes curettage and enucleation or excision and cryotherapy, although some authors report marginal resection as a more reliable treatment due to the tendency of the cyst to recur after enucleation or curettage.^[3,11]

In the present study, a case of GOC that recurred five years after the initial surgical treatment is reported.

CASE REPORT

A 45-year-old male patient who had no systemic diseases was referred to our clinic with a painless

swelling in the anterior mandible that had been present for one year.

On clinical examination, a perforation on the vestibular gingival mucosa of the incisor teeth accompanied the swelling with drainage of cystic fluid and there was no numbness around the lower lip (Figure 1). The past medical history revealed that the patient had undergone an operation in the same region in 2004, but he disregarded the follow-up period.

Panoramic radiograph revealed a large, multilocular, cystic lesion extending from the right second premolar to the left first molar, displacement of the incisors and left premolar and root resorption of the left premolar and molar teeth (Figure 2). Computed tomography revealed a lesion in the anterior mandible, destroying the buccal cortex of the alveolar bone.

Incisional and fine-needle aspiration biopsies were performed under local anesthesia and the initial histopathological diagnosis was "glandular odontogenic cyst." Under general anesthesia, enucleation and curettage was performed (Figure 3). The bone cavity was lined by a thick epithelium that was easily enucleated. Teeth related with the lesion [premolar to premolar] were extracted (Figure 4). The cavity was milled with hard milling cutter, after which Carnoy's solution was applied twice to the cavity;^[12] first for three minutes, and second for one minute and rinsed with 0.9% isotonic sodium chloride solution (Figure 5). The mucosa was then sutured. The postoperative period was uneventful except for unsuccessful primary closure around the mucosal perforation site. Two months after surgery the patient had no complaints, the mucosal perforation site was also healed successfully (Figure 6, 7).



Figure 3. Intraoperative view of the lesion.

Specimens were sent for further histopathological examination. The cyst was 5x2x2 cm in dimension during the macroscopic examination. The multilocular cyst epithelium and the extracted teeth were completely sampled for microscopic examination. Cystic lesions were localized apically to the incisor teeth.

After using a microtome, an Hematoxylin and Eosin (H-E) Staining protocol was applied to the specimens. Hematoxylin was applied for four minutes, after washing with 0.3% acid alcohol, eosine was applied for two minutes. The results of the staining are below:

- Collagen..... pale pink
- Acidophilic cytoplasm..... red
- Basophilic cytoplasm..... purple
- Nuclei..... blue
- Erythrocytes..... cherry red

Microscopic examination of the tissue showed a multilocular cystic lesion with non-keratinized



Figure 5. Intraoperative cavity after application of Carnoy's solution.

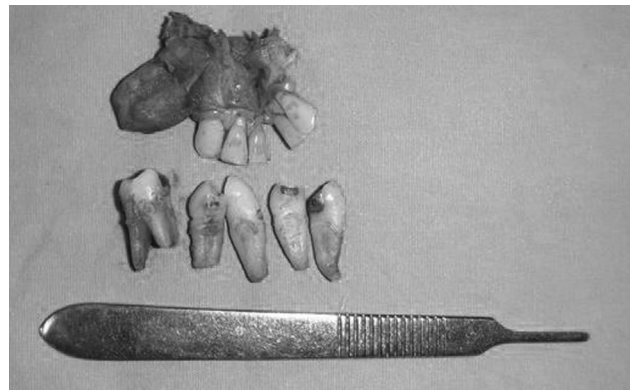


Figure 4. Enucleated lesion.

stratified squamous epithelium which focally revealed plaque-like nests, glandular structures and goblet cells. Keratinization was not evident. The wall of the cyst consisted of inflammatory connective tissue and widespread intraepithelial cysts. Hematin exhibits indicator-like properties, being blue and less soluble in aqueous alkaline conditions, and red and more soluble in alcoholic acidic conditions. Lymphoplasmocytic inflammatory cell infiltration was also seen in the wall of the cells and presented red areas. There were no remarkable findings that could suggest a malignancy. Thus, the lesion was diagnosed as a GOC (Figure 8).

DISCUSSION

According to the literature, GOC may have a wide clinicopathological spectrum ranging from benign pathology to a destructive malignant neoplasm.^[3] The differential diagnosis of a slowly-growing radiolucent lesion of the mandible includes cysts, myxoma, central giant-cell granuloma, fibrous dysplasia, ameloblastoma and central mucoepidermoid carcinoma.^[5] Preoperative examination of GOCs has to be the same as the procedure performed for any large radiolucent



Figure 6. Postoperative intraoral view.



Figure 7. Postoperative panoramic radiography.

lesion of an indeterminate origin and is significant to facilitate appropriate operative treatment.^[5] Fine-needle aspiration biopsy can be misleading due to the fact that the epithelium of the cyst is diagnostically valuable and the aspiration biopsy material may not contain any cystic wall cells. An incisional biopsy is essential to differentiate between GOC, ameloblastoma and other lesions.^[5] In the present case, both biopsies were performed under local anesthesia before enucleation which was performed under general anesthesia. The enucleated material was also examined histopathologically.

The clinical aspects of the present case are consistent with those reported for most of the GOCs. The lesion has a slight predilection for men and occurs mostly in middle-aged patients. The age range has been from 14 to 90 years with a mean age of 49.5 years.^[2,4,10] According to the literature GOCs are found mostly in the anterior mandible.^[6,12] A multilocular radiolucency is usually seen, although unilocular lesions have also been described.^[4] The GOC is a rarely-seen lesion but should be considered for the differential diagnosis of unilocular and multilocular radiolucencies of the jaws.^[9]

Periapical, panoramic radiographs and CT can be used to visualize the lesions. Radiologically GOC can be seen as a unilocular or multilocular radiolucency, usually in the anterior and anterolateral portions of the jaws.^[1,6,12,13] In this case report, the teeth related multilocular radiolucency of the anterior mandible which could be seen clearly in the panoramic radiograph was significant and compatible with the former diagnosis and newly performed incisional biopsy, therefore no other radiodiagnostic technique was requested.

The recurrence rate of GOC ranges between 21% and 55%.^[12] The high rate of recurrence may

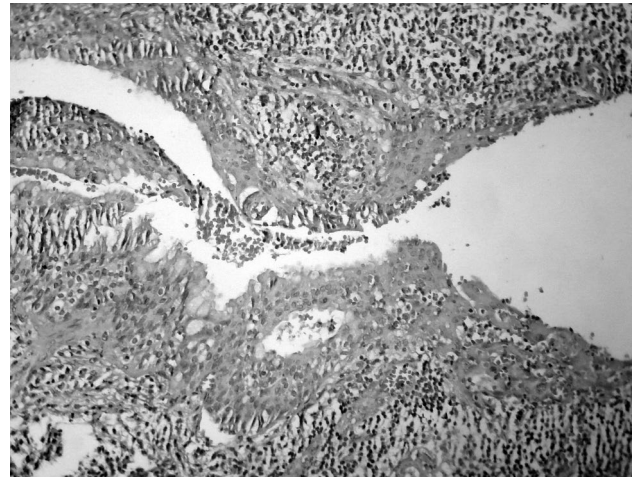


Figure 8. Histological cross-sectional view of the lesion (H-E x 200).

be an indication of the aggressive nature of GOC. The reason of recurrence may be the teeth which are related with the lesion. Another reason of aggressiveness is the compromised cortical integrity. According to the literature there are few cases that reported the use of CT for the radiographic examination of GOC.^[12] The patients with recurrence reveal a higher percentage of cortical compromise findings.^[12] Cortical perforation should be recognized and considered during the treatment planning and appropriate modalities such as occlusal plane radiographs and CT should be performed to evaluate this feature.^[12] The cortical perforation in this case could also be seen in the clinical examination, therefore CT and occlusal plane radiographs were not necessary to evaluate the perforation.

The treatment for GOC remains controversial due to the rarity of reported cases. The lesions vary considerably in size and aggressiveness and treatment ranges from simple curettage to marginal resection.^[9] The treatment of the GOC includes curettage, enucleation, conservative surgery, and partial or total resection, excision and cryotherapy.^[3,4,11] In the present case the cystic lesion was enucleated and curettage was performed. Teeth related with the lesion were extracted and Carnoy's solution was applied after enucleation due to the recurrence and questionable patient reliability for follow-up examinations.^[14] Cryotherapy was not considered as a treatment option for this case, due to the tendency of the bone to fracture after cryotherapy. Reconstruction with iliac graft is planned after one-year follow-up period.

The literature review revealed two previous reports with inclusion of an impacted tooth and one odontoma in the cyst cavity. Because of this contradiction it can be questionable whether this was actually a GOC arising in a dentigerous cyst or just mimicking one.^[3,13] In our case there was no impacted tooth or any other calcified lesion related to the cystic lesion. In the tooth-related lesions enucleation and retrograde amalgam placement may be performed.^[1] In our case we did not consider retrograde amalgam filling as a treatment option and preferred to extract the related teeth because of the higher risk of recurrence due to the second operation of the related region. A central mucoepidermoid carcinoma is one of the lesions that are significant for the differential histopathological diagnosis for GOC. The difference between these two lesions is the grade of the epithelium proliferation.^[3,13] The lesion may not be homogeneous in the histopathological view. Gardner et al.^[1] had a 19-year-old male patient with the complaint of swelling in the mandible and performed enucleation of the lesion. The histopathological examination showed a part of the lesion was typical pseudoglandular odontogenic cyst except for lack of mucous pools; the other part was typical ameloblastoma. The histopathological examination of our case showed no non-homogeneous pattern which would lead to another lesion and confuse the diagnosis.

In conclusion, histopathological examination of GOC is clear and unambiguous, but it can be confusing on clinical examination. Further studies can be performed to be aware of GOCs, their tendencies to recur and the importance of the longer follow-up periods for clinicians and pathologists.

Acknowledgements

The authors would like to express thanks to Dr. Ömer Günhan (MD, Professor, Department of Pathology, Gülhane Military Medical Academy, Ankara, Turkey) for sharing his valuable experience on pathology, kindly interpreting the pathology specimens and providing histopathological diagnosis on this case.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Gardner DG, Kessler HP, Morency R, Schaffner DL. The glandular odontogenic cyst: an apparent entity. *J Oral Pathol* 1988;17:359-66.
2. Patron M, Colmenero C, Larrauri J. Glandular odontogenic cyst: clinicopathologic analysis of three cases. *Oral Surg Oral Med Oral Pathol* 1991;72:71-4.
3. Kasaboğlu O, Başal Z, Usubütün A. Glandular odontogenic cyst presenting as a dentigerous cyst: a case report. *J Oral Maxillofac Surg* 2006;64:731-3.
4. de Sousa SO, Cabezas NT, de Oliveira PT, de Araújo VC. Glandular odontogenic cyst: report of a case with cytokeratin expression. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;83:478-83.
5. 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.
6. Qin XN, Li JR, Chen XM, Long X. The glandular odontogenic cyst: clinicopathologic features and treatment of 14 cases. *J Oral Maxillofac Surg* 2005;63:694-9.
7. Takeda Y. Glandular odontogenic cyst mimicking a lateral periodontal cyst: a case report. *Int J Oral Maxillofac Surg* 1994;23:96-7.
8. Ficarra G, Chou L, Panzoni E. Glandular odontogenic cyst (sialo-odontogenic cyst). A case report. *Int J Oral Maxillofac Surg* 1990;19:331-3.
9. Chavez JA, Richter KJ. Glandular odontogenic cyst of the mandible. *J Oral Maxillofac Surg* 1999;57:461-4.
10. Ramer M, Montazem A, Lane SL, Lumerman H. Glandular odontogenic cyst: report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;84:54-7.
11. Sittitavornwong S, Koehler JR, Said-Al-Naief N. Glandular odontogenic cyst of the anterior maxilla: case report and review of the literature. *J Oral Maxillofac Surg* 2006;64:740-5.
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. Meral G, Akalın AG, Saysel M, Taşar F. Glandular odontogenic cyst: a case report. *GÜ Dışhek Fak Derg* 2005;22:117-20.
14. Stoelinga PJ. The treatment of odontogenic keratocysts by excision of the overlying, attached mucosa, enucleation, and treatment of the bony defect with carnoy solution. *J Oral Maxillofac Surg* 2005;63:1662-6.