

## Case report

## Case series and review of glandular odontogenic cyst with emphasis on treatment modalities

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## ABSTRACT

Glandular odontogenic cyst is a newly determined jaw entity with aggressive behavior and a high rate of recurrence. There is histopathologic resemblance to other lesions of the jaw such as intraosseous mucoepidermoid carcinoma. Although enucleation and curettage are not the treatment of choice for this cystic lesion, they comprise the most common method. On the other hand, filling the defect is a controversial matter, especially in lesions with large size. We introduce 4 cases of GOC, of which 2 are cases of recurrence. We applied bone material substitutes in 3 of these cases with success.

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## 1. Introduction

Glandular odontogenic cyst (GOC) is an uncommon cyst of the jaws. In spite of similarity to salivary gland histologic features, the presence of other odontogenic lesions with GOC simultaneously and immunohistochemistry evaluations have confirmed its odontogenic origin to a great extent (Kramer et al., 1992; Koppang et al., 1998; Hisatomi et al., 2000; Yoon et al., 2006; Vered et al., 2010).

The first case report of this entity with the name “GOC” was published in 1988 (Gardner et al., 1988), although Padayachee and Van Wyk reported this lesion as sialo odontogenic cyst before that (Padayachee and Van Wyk, 1987).

Men are involved more frequently than women. The mandible is affected more than the maxilla. This lesion can cross the midline and is characterized by aggressive behavior; therefore, correct diagnosis is of highest importance. Treatment of GOC is controversial, from enucleation and curettage to en bloc resection with or without bone graft.

In this article, we introduce four cases of GOC with emphasis on treatment modalities and use of biomaterial bone substitutes.

## 2. Case reports

The first case was that of a 62-year-old man, who had been referred to the oral and maxillofacial surgery ward of the dental school at Tehran University of Medical Science with the chief complaint of pain and swelling in the anterior part of the mandible since 6 months previously. As shown in the cone-beam computed tomography (CBCT) scan (Fig. 1), there was a large unilocular radiolucency that passed through the midline and was associated with 2 impacted canines. The coverage of the bone at the labial aspect was complete and intact. After enucleation of the cyst, we made two observations. The first was that the cyst was not filled with liquid. In other words, in aspiration we found a brown creamy material, as if the consistency of the liquid had been increased over time. The second observation, which was more important, was that there was thickening in the cyst lining in some areas, as in a mural ameloblastoma. We also decided to do an aggressive peripheral osteotomy after surgical extraction of two impacted canines. We preferred to fill the defect with biomaterial bone substitute, but as the patient could not afford it, we did not use anything and sutured the flap. The pathologic report was surprising—a glandular odontogenic cyst. The postoperative orthopantomogram (OPG) revealed that there was no recurrence 3 years after surgery (Fig. 2).

The second case was that of a 47-year-old man. In 1994, he had been referred to the oral and maxillofacial surgeon with a chief

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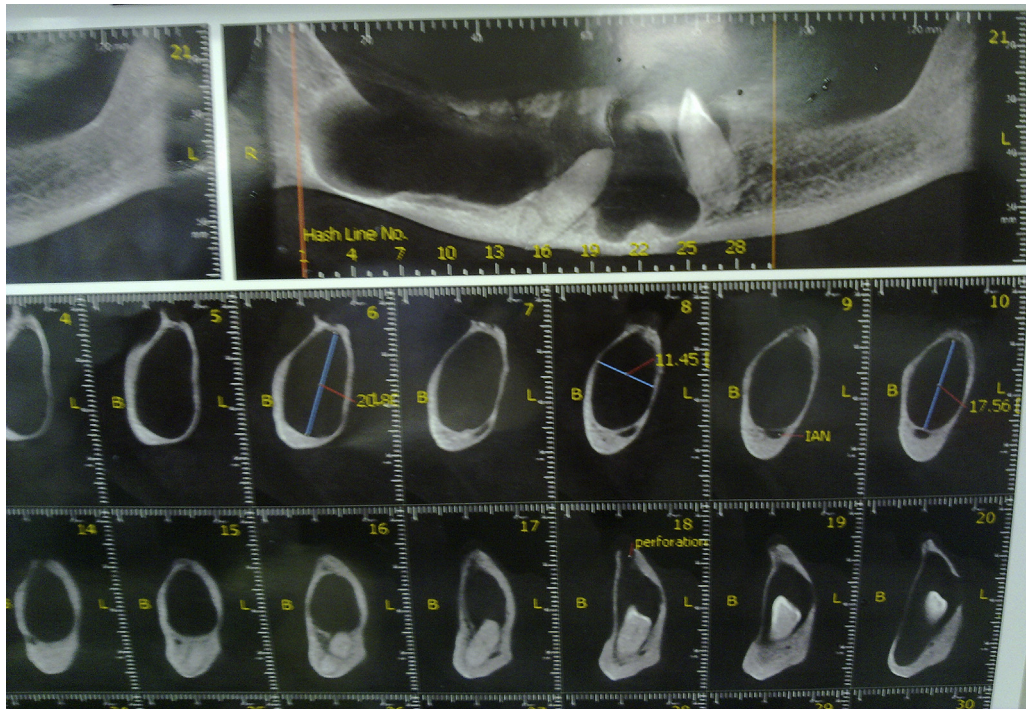


Fig. 1. Cone-beam computed tomogram of a large glandular odontogenic cyst.

complaint of fluctuating inflation at the medial aspect of right side of the mandible near the second and third molars. On preoperative OPG, a multilocular radiolucency was seen. Enucleation and curettage were performed, and the pathologic report was mural ameloblastoma. This patient had no problem for 4 years until 1998. At that time, he had the same chief complaint and had been referred to the oral and maxillofacial surgeon with a new OPG. In this OPG there was a unilocular radiolucency in the same area. Under local anesthesia (LA) the lesion was enucleated again, and the pathologic evaluation revealed a cyst with stratified squamous lining epithelium and mucous cells in some areas. He had no complication for 15 years until 2013. This year he had the same chief complaint and has been referred to us with a new OPG, which revealed a large multilocular radiolucency at right side of the mandible, just like a recurrence of ameloblastoma. After reflecting the mucoperiosteal flap and removing bone, we found nothing but a hollow space covered by a very thin epithelial lining just like a traumatic bone cyst. After performing curettage of the whole

epithelial lining, we did an aggressive peripheral osteotomy in order to induce bleeding in the space to improve bone regeneration. In addition we extracted the third mandibular molar and used an ivy loop for intermaxillary fixation in order to prevent post-operative fracture. The pathologic report was that of glandular odontogenic cyst. On re-evaluation of the first and second pathologic specimens, we found the same histologic pattern, and it seemed that the previous reports were not correct (Figs. 3 and 4).

The third case was a 28-year-old man. He had pain and swelling in the anterior part and left side of the mandible for 1 year previous to the visit. Furthermore, he had a history of trauma to this area 8 years ago. First, we tested all the teeth in the mentioned area for vitality, and referred the patient to an endodontist for root canal therapy of nonvital teeth. As shown on OPG, there was a large

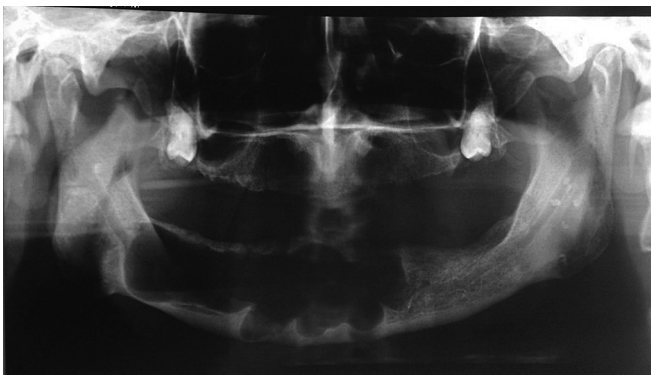


Fig. 2. Postoperative panorex 3 years after surgery.

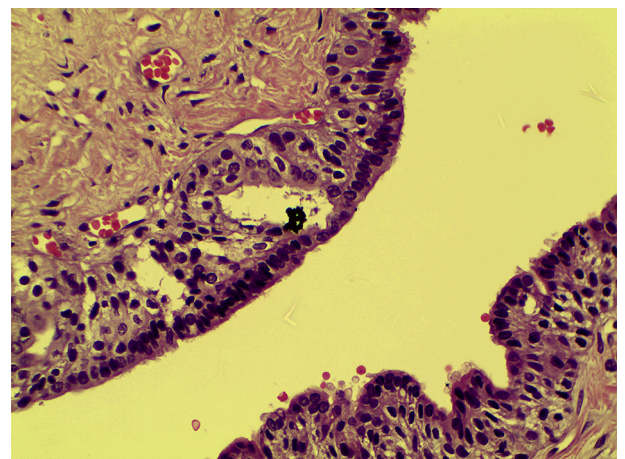


Fig. 3. Photomicrograph showing cuboidal to columnar epithelium and cilia on surface and microcyst formation (hematoxylin and eosin stain, original magnification  $\times 80$ ).



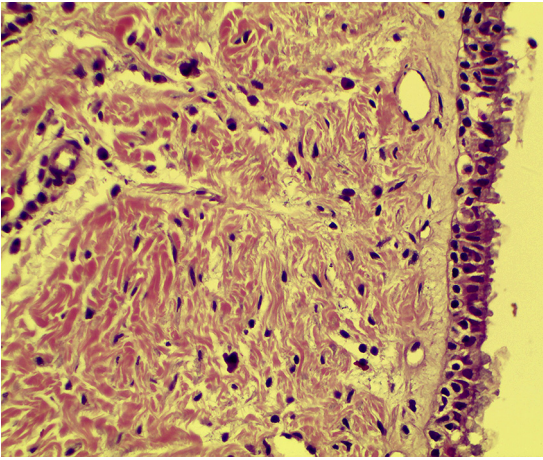


Fig. 4. Photomicrograph showing a hobnail view because of uneven superficial epithelial cells (hematoxylin and eosin stain, original magnification  $\times 80$ ).



Fig. 6. Particulate allogeneic bone insertion into the defect after enucleation and peripheral osteotomy.

radiolucency passing through the midline (Fig. 5). After reflecting the mucoperiosteal envelope flap and removing bone, we had access to the lesion. After enucleating the cyst, we made the same two observations that we saw in the first case reported here: a cyst lumen filled with brown creamy material and a thickening of the lining. Therefore we suspected another glandular odontogenic cyst, and decided to perform an aggressive peripheral osteotomy after doing the retrograde apicoectomy for all roots the apices of which were in the defect. After filling the defect with the allograft bone substitute and covering the allograft with the membrane, the flap was sutured (Figs. 6 and 7). The postoperative panorex revealed no sign of recurrent after 3 years (Fig. 8).

The last patient was a case of recurrence. He was 58 years old. The first surgery was carried out 12 years before, and the pathologic report was GOC. His newly presenting chief complaint was swelling and paresthesia of the lower lip. On aspiration, a red fluid could be seen. A unilocular radiolucency was seen in the anterior part of the mandible (Fig. 9). Root canal therapies of the involved teeth had been done before the first surgery. After reflection of the mucoperiosteal flap, cortical perforation of the buccal bone was obvious. The cyst was removed, and after an aggressive peripheral osteotomy, the defect was filled with allogeneic bone particulates and covered with membrane (Figs. 10 and 11). The mobile teeth were removed. Three months after the surgery, there is no evidence of recurrence, but long-term follow-up is essential (Fig. 12).



Fig. 7. Resorbable membrane on allogeneic bone graft.



Fig. 8. Postoperative panorex 3 years after surgery.



Fig. 5. Preoperative panoramic image.

### 3. Discussion

From the initial reports of GOC in 1987 and 1988, only a few cases of this jaw lesion have previously been reported (Gardner et al., 1988; Padayachee and Van Wyk, 1987). There are similarities between GOC and other lesions, such as low-grade mucoepidermoid carcinoma, lateral periodontal cyst, and botryoid cyst from histologic aspects (Vered et al., 2010; Takeda, 1994; Waldron and Koh, 1990). Because of these similarities and the aggressive





Fig. 9. Panoramic image shows a large radiolucency in the anterior part of mandible.



Fig. 12. Postoperative panorex after 3 months.



Fig. 10. Filling the defect with allograft.



Fig. 11. Membrane covering the bone substitute.

behavior of GOC, definitive diagnosis is of significant importance. There is a predilection to male gender involvement (Kaplan et al., 2008; Shen et al., 2006). Most of the cases are those of adults, with the highest frequency in the third decade of life, but GOC can develop during a wide range of ages (Shen et al., 2006). The most common clinical features are pain and swelling, although other features such as tooth displacement and mobility can occur. The lower jaw is affected more than the maxilla, and the lesion can cross the midline frequency. From radiographic features, most of

the cases are unilocular radiolucencies, but in large cysts the multilocular radiolucency feature is dominant (Kaplan et al., 2008). The border of this cyst is well defined and, in some cases, scalloped. Root resorption, cortical bone thinning and perforation, and tooth displacement may occur. Various kinds of fluids may aspirate from this cyst. Krishnamurthy et al. mentioned a clear, low-viscosity fluid on aspiration (Krishnamurthy et al., 2009); others have reported a brownish-red fluid on aspiration (Araújo de Moraes et al., 2012). In one of our cases, aspiration was negative, whereas in two cases the lesion contained creamy, high-viscosity fluid. Surgical treatment of this lesion differs and depends on surgeon preference. Enucleation and curettage, which is the most common technique, marsupialization, marginal or partial resection of the jaw, and adjunctive therapies such as application of Carnoy solution, filling the cavity with autogenous or allogeneous bone substitute, and cryosurgery are different methods to treat this cystic jaw lesion. The rate of recurrence varies in the literature. Recurrence is more common in lesions of larger size, with cortical bone perforation, and of multilocular radiographic appearance (Thor et al., 2006; Boffano et al., 2010). It is not easy to identify the rate of success for any of the treatment modalities because of the low incidence of this lesion in the population and poor follow-up in previous studies. Araújo de Moraes et al. believed that follow-up visits should continue at least for 3 years after surgery, but most reported cases have been followed for 2 years or less (Araújo de Moraes et al., 2012).

#### 4. Conclusion

In the cases of mandibular pathologic lesion in which the lesion crosses the midline, GOC is one of the most important differential diagnoses. Other signs that are beneficial to diagnosis of GOC are multi-locular radiographic appearance and a high-viscosity fluid on aspiration. Aggressive peripheral osteotomy is strongly recommended by the authors of this case report. Decreasing the dead space with allogeneic bone material will help to reduce postoperative complications.

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