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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.

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 ostectomy 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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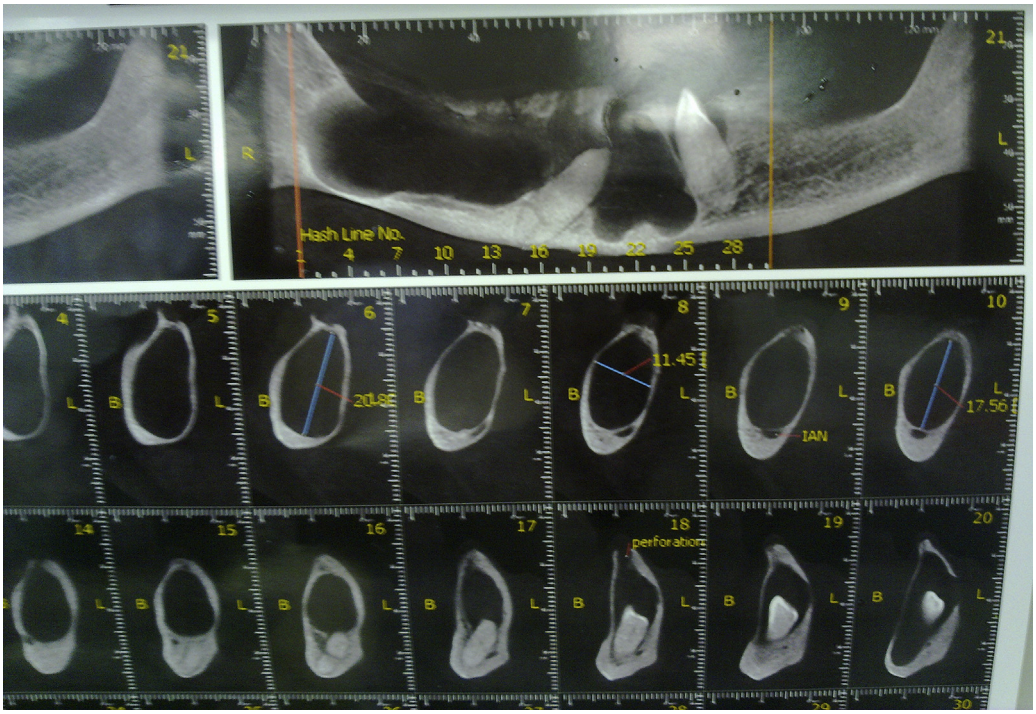

Fig. 1. Cone-beam computed tomogram of a large glandular odontogenic cyst.

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 ostectomy 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 postoperative 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

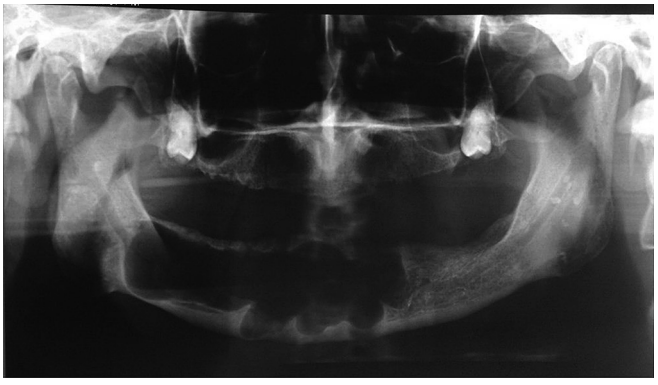

Fig. 2. Postoperative panorex 3 years after surgery.

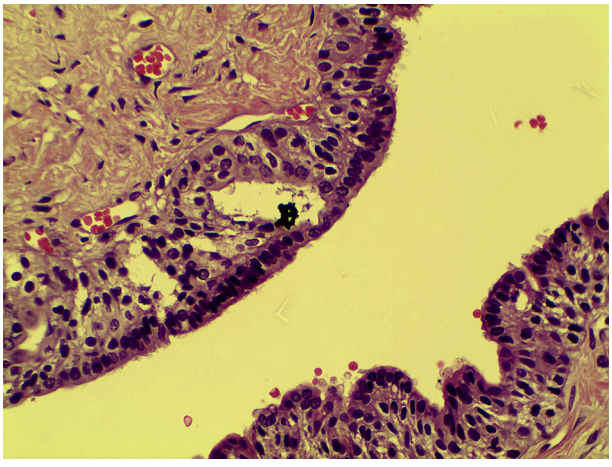

Fig. 3. Photomicrograph showing cuboidal to columnar epithelium and cilia on surface and microcyst formation (hematoxylin and eosin stain, original magnification × 80).