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Case report

Glandular odontogenic cyst arising in the posterior region of the mandible

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ABSTRACT

Glandular odontogenic cyst (GOC) is a rare, benign, cystic lesion of the jaws, which occurs mostly in the mandible, especially in the anterior region. The histogenesis of GOC remains uncertain and the World Health Organization classified the lesion as a developmental odontogenic epithelial cyst in 1992. In this article, we describe a rare case of GOC arising in the posterior region of the mandible in a 64-year-old male and also present the clinical, radiological and histopathological features of GOC. Radiological examination revealed a solitary well-defined unilocular radiolucent lesion in the left mandibular posterior region. The lesion was clinically diagnosed as radicular cyst associated with the left first molar. The cystic lesion was enucleated from the jaw bone in one piece and tooth extraction was performed under general anesthesia. The surgical specimen was investigated histopathologically and was diagnosed as GOC. Due to the high recurrence rate of GOC, complete removal of the cystic lesion and careful clinical and radiological follow-up are important. In the present case, a panoramic radiograph was taken after 19 months of follow-up, which revealed complete healing of the involved site with no evidence of recurrence.

1. Introduction

Glandular odontogenic cyst (GOC) is a rare, benign, cystic lesion of the jaws, which was first described in Gardner et al. [1] as a distinct clinicopathologic entity. In 1992, the World Health Organization classified the lesion as a developmental odontogenic epithelial cyst under the terms "glandular odontogenic cyst" or "sialo-odontogenic cyst" [2].

GOC occurs mostly in middle-aged people and the most common site of occurrence is the mandible, especially in the anterior region, presenting as an asymptomatic slow growing swelling [3,4]. Radiographically, GOC typically presents as a solitary well-defined unilocular or multilocular radiolucency [3,5]. Histopathologically, GOC presents certain characteristic features that include non-keratinized stratified squamous epithelium, the superficial layer of the epithelium consisted of eosinophilic cuboidal cells, localized plaque-like thickenings of epithelium, intraepithelial duct-like structures, mucous cells and absence of inflammation in the underlying connective tissue [6,7]. For the treatment of GOC, several surgical methods including enucleation, curettage, and a more aggressive surgery have been used [8,9].

In this article, we describe a rare case of GOC arising in the posterior region of the mandible in a 64-year-old male and also reviewed the literature.

2. Case report

A 64-year-old male was referred to National Defense Medical College Hospital, with a chief complaint of swelling of the left mandible. The patient reported that he had occasionally noticed such symptom for several years. The patient's medical and family histories were unremarkable. An intraoral examination revealed diffuse swelling and fistula of the buccal gingiva surrounding the left first molar of the mandible. A panoramic radiograph showed a solitary well-defined 20 × 30-mm unilocular radiolucency present in the root apex region extending from the left second premolar to the left first molar of the mandible (Fig. 1). There was no apparent root resorption of the involved teeth. Computed tomographic examination was performed and revealed a well-defined 20 × 10 × 16-mm unilocular bone absorption in the left mandibular posterior region at the level of the root apex and the mandibular canal was displaced inferiorly (Fig. 2). Following a clinical diagnosis of radicular cyst associated with the left first molar of the mandible, the involved tooth was extracted and the cystic lesion was enucleated from the jaw bone in one piece under general anesthesia (Fig. 3). In addition, an apicectomy was performed on the second premolar and due to a severe periodontitis, the second molar was extracted. The surgical specimen was submitted for histopathological examination to confirm the diagnosis. Histopathological examination revealed a cystic lesion with

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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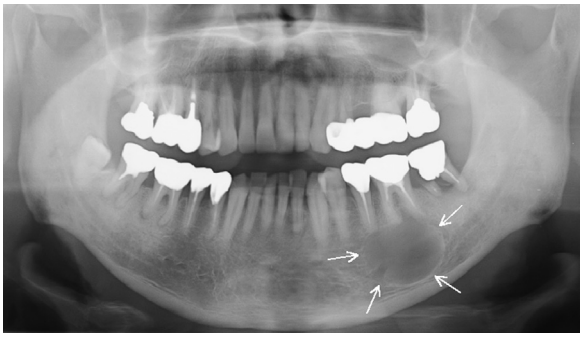

Fig. 1. Panoramic radiograph showing a well-defined unilocular radiolucent lesion (arrow) in the left posterior mandible.

luminal epithelium and surrounding connective tissue. The cystic wall was lined by non-keratinized, stratified squamous epithelium associated with plaque-like epithelial thickenings and the superficial layer of the epithelium consisted of cuboidal eosinophilic cells (Fig. 4a and b). Mucous cells and duct-like structures were noted in the lining epithelium (Fig. 4c). The underlying connective tissue consisted of densely fibrous tissue with very few inflammatory cells. Occasionally, epithelial islands were noted in the underlying connective tissue (Fig. 4d). Based on the above-mentioned histopathological features, a definitive diagnosis of GOC was made. A panoramic radiograph was taken after 19 months of follow-up, which revealed complete healing of the involved site with no evidence of recurrence (Fig. 5).

3. Discussion

GOC is a rare developmental cyst of the jaws comprising only 0.2% of all odontogenic cysts [10]. The most recent published review of the literature on GOC, reported by Kaplan et al. [11], identified 111 reported cases in 48 articles. According to the article, a total of 57% of the cases have been reported in males and 43% in females, the mean age at diagnosis was 45.7 years and GOC can occur within wide age range of 14–75 years [11]. Location of GOC is the most often in the mandible, especially in the anterior region, whereas 30% of GOC is located on the maxilla [11]. In the present case, the lesion was found in the posterior region of the mandible in a 64-year-old male.

Radiographically, GOC typically presents as a well-defined unilocular or multilocular radiolucent lesion with well-defined borders and it does not display any specific or pathognomonic features [3,5]. Computed tomography (CT) scan is useful for diagnosis and surgical planning of GOC because it provides accurate information, particularly in evaluating the locularity and expansion of the lesion. However, based on the radiographic appearance, the diagnosis of GOC is practically difficult because there are other lesions having similar radiographic appearance, such as unicystic or multicystic ameloblastoma, odontogenic keratocyst, simple bone cyst, lateral epithelial cyst, botryoid odontogenic cyst, lateral epithelial cyst and central mucoepidermoid tumor [5]. In the present case, a unilocular radiolucent lesion with a well-defined border was observed and the cystic lesion was located below the apex of a nonvital tooth. Based on the radiographic appearance, we diagnosed the lesion as radicular cyst.

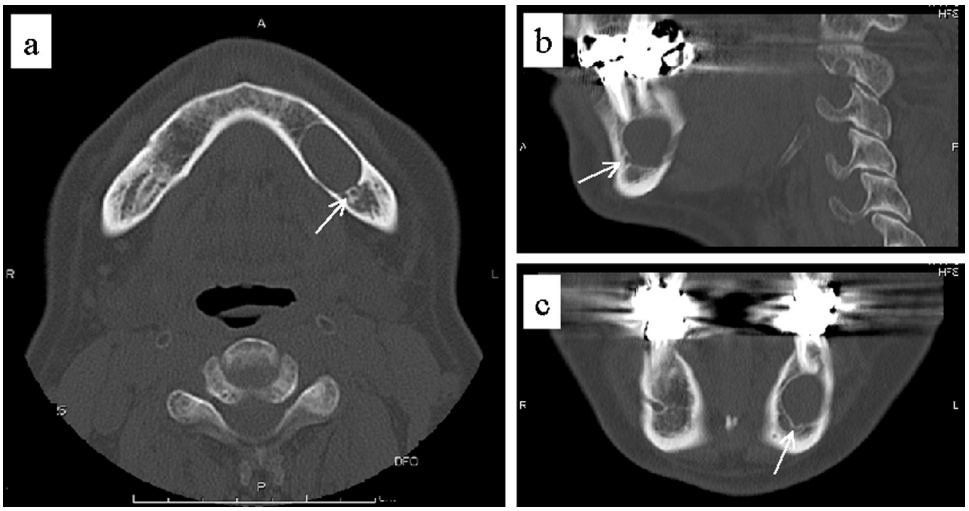

Fig. 2. Computed tomographic axial (a), sagittal (b), and coronal (c) scans revealing a well-defined unilocular bone absorption in the left posterior mandible at level of the root apex and inferior displacement of the mandibular canal (arrow).

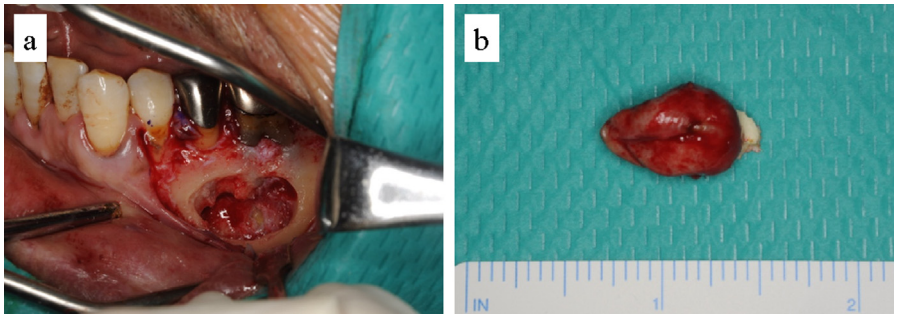

Fig. 3. Intraoperative photographs showing the cystic lesion in the jaw bone (a) and enucleated specimen (b).