

Bilateral Glandular Odontogenic Cyst of Mandible: A Rare Occurrence

İsmail Akkaş · Orçun Toptaş · Fatih Özkan ·
Fahri Yılmaz

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Abstract Glandular odontogenic cysts (GOCs) of the jaw are rare with well-defined limits radiologically, unusual histopathological features and a high recurrence rate. The radiographic appearance of GOCs vary and are not pathognomonic. Definitive diagnosis of the GOC is established only by histopathological examinations. Histologically, GOC is characterized by a thin nonkeratinized squamous epithelial lining, with papillary projections, nodular thickenings, mucous (goblet) cells with intraepithelial mucous pools and intraepithelial glandular, microcystic or duct-like structures. We present an unusual case of a bilateral GOC in the mandible. This case report is also the first documented case of bilateral GOC in the mandible.

Keywords Glandular odontogenic cyst · Enucleation · Carnoy's solution · Mandible

Introduction

Padayachee and Van Wyk [1] presented multilocular cystic lesions that were similar to botryoid odontogenic cysts but with a glandular ingredient and suggested the name “sialo odontogenic cyst”. In 1988, Gardner et al. [2] reported eight other cases and called the lesions glandular odontogenic cysts (GOCs) because there was a mucin structure in the cyst epithelium that had no finding of salivary gland

origin. Glandular odontogenic cysts are usually misdiagnosed with other lesions such as botryoid odontogenic cysts and low-grade central mucoepidermoid carcinomas due to microscopic similarities [3, 4].

The GOC has a higher prevalence in middle-aged people [4]. Distribution by gender showed a slightly male predilection [4]. It may be found in both jaws. However, lesions are more common in the mandible [5], especially the anterior mandible [6].

Patients may complain of pain, swelling or discharge, but sometimes GOCs can be asymptomatic and discovered on routine radiographs, when a tooth has failed to erupt, teeth are missing, tilted or otherwise out of alignment [7–9]. GOCs have benign but locally aggressive behavior with a significant recurrence rate [4, 7, 10].

Radiographically, the lesion appears as a well-defined, uni or multilocular radiolucent cystic area [8, 9]. Loss of cortical integrity and root resorption may occur [4], and an impacted tooth sometimes can be observed in a cyst cavity [11, 12]. Computed tomography (CT) is recommended for diagnosis, surgical planning and follow-up [13].

Histologically, GOC is mostly characterized by a thin nonkeratinized squamous epithelial lining, with papillary projections, nodular thickenings, mucous (goblet) cells with intraepithelial mucous pools and intraepithelial glandular, microcystic or duct-like structures. It also includes superficial layer of cuboidal or columnar cells within the lining [4].

Enucleation with peripheral curettage or marginal excision is the most common treatment modality, although some authors claim marginal resection is a more reliable treatment due to the tendency of the cyst to recur after enucleation with curettage [3]. The aim of this study is to present an unusual case of a bilateral GOC in the body of the mandible. This report is also the first documented case of a bilateral GOC in the mandible.

İ. Akkaş · O. Toptaş (✉) · F. Özkan
Department of Oral and Maxillofacial Surgery, Faculty of
Dentistry, Abant İzzet Baysal University, Bolu, Turkey
e-mail: otostas@ibu.edu.tr

F. Yılmaz
Department of Pathology, Faculty of Medicine, Abant İzzet
Baysal University, Bolu, Turkey

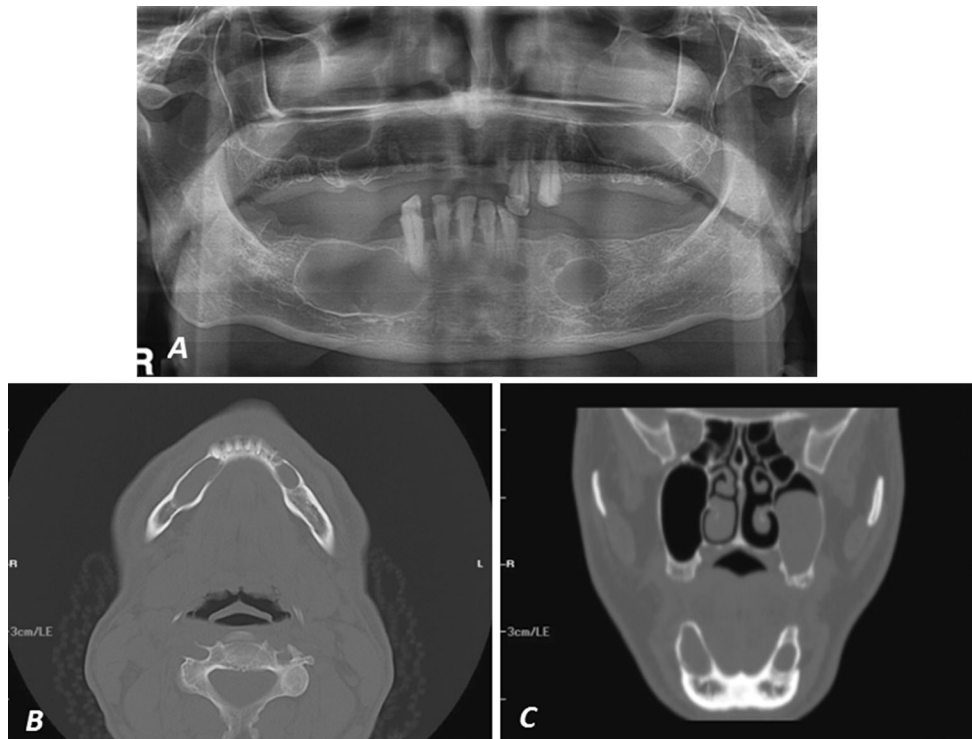


Fig. 1 **A** Panoramic view of the patient revealed bilateral radiolucent lesion on the body of the mandible. **B, C** Axial and coronal view of lesion by computerized tomography, respectively

Case Report

A 40-year-old man was admitted to the School of Dentistry, University of Abant Izzet Baysal, Bolu, Turkey, with a complaint of partial edentulism. The patient's medical history was insignificant. Intraoral examination revealed no evidence associated with radiolucent lesion. The overlying mucosa was of normal color and appearance. The right mandibular canine had no vitality loss. There was no complaint of lower lip numbness bilaterally. The extraoral examination revealed no significant findings.

A panoramic radiograph demonstrated a well-defined, unilocular radiolucency present in both sides of the body of mandible (Fig. 1A). There was no evidence of resorption of the root or displacement of the tooth. Axial and coronal CT showed a large, hypodense unilocular lesion with a cystic pattern and well-defined borders in the right mandibular body and a smaller unilocular radiolucent area in the left premolar region of the mandible. Both lesions were surrounded by a thin and intact cortical bone layer, medially and laterally (Fig. 1B, C).

The lesions were enucleated via an intraoral approach under local anesthesia. Peripheral osteotomy was performed to a depth of approximately 2 mm. Carnoy's solution was applied to the bone cavity for 3 min using ribbon gauze soaked with Carnoy's solution. The gross

specimen was composed of an elastic, brownish-cream cystic mass. The wound was closed primarily on both sides. The excised specimens were sent for histopathological investigation.

Microscopically, the supporting fibrous connective tissue wall of the cyst was lined with non-keratinized stratified squamous epithelium. Nodular areas were fully filled with mucous cells and mucous cysts. Within the thickness of the epithelium, areas exhibiting intra-epithelial gland-like structures with mucin secreted cells (goblet cells) were observed. These cells were positive with periodic acid-Schiff (PAS) and Alcian blue stain (Fig. 2).

The patient was diagnosed with GOC and was followed up to 36 months postoperatively in our clinic. The healing process was uneventful, and there was no evidence of recurrence (Fig. 3).

Discussion

GOC is a rare developmental odontogenic cyst with a low incidence rate of about 0.012 % [14] to 1.3 % of all jaw cysts [5, 15]. GOC has no pathognomonic panoramic view [5]; therefore, most clinicians often suppose that a radiolucent lesion may be a dentigerous cyst or a radicular cyst at the patient's initial visit [11]. In addition, Manor et al.

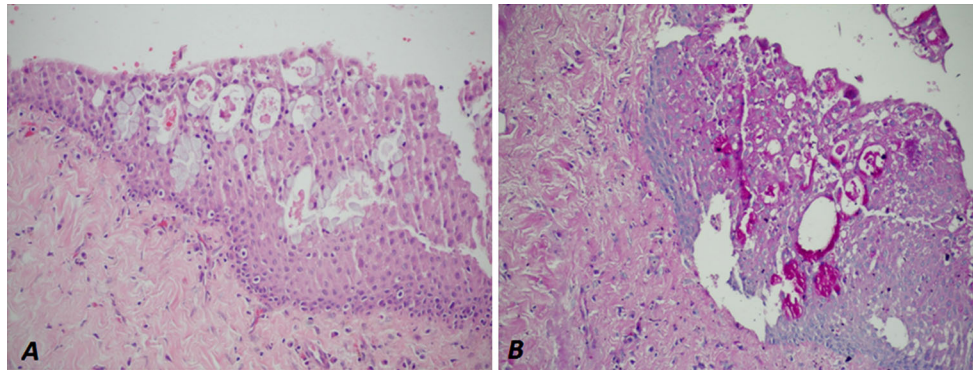


Fig. 2 **A** Cystic line is composed of stratified squamous epithelium, and contains goblet cells, glandular structures and intraepithelial cysts (H&E; original magnification $\times 100$). **B** PAS positive goblet cells (PAS stain; original magnification $\times 100$)



Fig. 3 The healing process was uneventful, and there was no evidence of recurrence after 36 months of follow-up

[5] reported that the number of multilocular and unilocular lesions was almost equal in the literature. Multilocular lesions must be separated clinically and histopathologically from kerotocystic odontogenic tumors, central mucoepidermoid tumors, central giant cell granulomas, and ameloblastomas [3].

Glandular odontogenic cysts are found in the mandible, in approximately a 4-to-1 ratio with a predilection for the anterior areas of the mandible [5]. Bilateral occurrence of GOC has been reported only once previously in the literature [16]. In the previous case, the lesions were bilateral and present in the posterior maxilla. In our case, the lesions were also bilateral but present in the body of the mandible. Therefore, our case is the first report of bilateral occurrence in the mandible in the scientific literature.

GOC is characterized by locally aggressive behavior. Surgical excision with peripheral osseous curettage or ostectomy, curettage with adjuvant Carnoy's solution or cryotherapy is the conservative method of management

[17, 18]. This more aggressive approach (marginal resection to partial jaw resection) for a cystic lesion is justified because of the high recurrence rate associated with GOC [3, 8, 19]. Long-term follow-up of patients with GOC treated with curettage or enucleation should be carefully carried out, due to reported recurrent cases up to 7 years [4]. Kaplan et al. [4] recommended that patients diagnosed with GOC should be followed up for at least 3 years and up to 7 years in cases treated with curettage or enucleation. In the present case, there was no recurrence during the 3-year follow-up period for both sides.

The reason for the high recurrence rates, as supported by the fact that 14–86 % of cases recur following curettage [10], remains unclear but may be related to one of several possibilities. Micro cysts or small dental lamina remnants in the bone adjacent to the primary lesion may contribute to recurrence [7]. The thin connective tissue wall of the cyst may lead to incomplete removal. In addition to the histopathological disadvantages, the lesion's increased tendency

to recur is directly associated with the size and the locularity of the lesion. Most patients who have recurrent disease have large multilocular lesions with cortical perforations [19]. Therefore, an aggressive surgical approach is important to prevent recurrences [17, 19]. In GOCs with cortical perforation, resection of the overlying mucosa was also recommended [10]. In the present case, both lesions had no cortical perforations, and conservative treatment associated with peripheral ostectomy and cauterization with Carnoy's solution was performed. The patient did not accept any further treatment, such as marginal or en-bloc resection with mandibular reconstruction.

Carnoy's solution was first described in 1933 as a powerful fixative, haemostatic and cauterizing agent for the treatment of cyst and fistulae [20]. It also penetrates cancellous spaces in the bone and devitalizes the left out microcystic structures of some types of cysts. Gosau et al. [21] reported in their retrospective study that enucleation plus the application of Carnoy's solution reduced the recurrence rate of keratocystic odontogenic tumors compared with simple enucleation. Recent studies also showed that the use of Carnoy's solution after enucleation of unicystic ameloblastomas reduces recurrence rates [22, 23]. However, exposure to chloroform during treatment procedures has been associated with cancer and reproductive toxicity [24]. Therefore, the new formula of Carnoy's solution without chloroform is accepted by some authors but further studies which compare using conventional and modified Carnoy's solution are needed to find out the recurrence rate of two treatment modalities [25].

Lateral periodontal cysts, botryoid odontogenic cysts, and central mucoepidermoid carcinomas (multicystic variant) have significant histopathological similarities to GOC [4, 7, 8]. Thus, some histopathologic features should be definitely noted to rule out similar lesions. The most prominent and unique microscopic features are the presence of a superficial layer of cuboidal or columnar epithelial cells occasionally with cilia and glandular or pseudoglandular structures and intraepithelial crypts containing mucin [3]. The GOC is lined with thin nonkeratinized stratified squamous epithelium [3, 7, 9]. Moreover, the multicystic type more than the unicystic type exhibits infiltration of the surrounding tissue and/or daughter cyst formation [3]. The histopathological features of our case are similar to the GOC described by these authors.

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