Kaohsiung J Med Sci 16: 53 — 58, 2000

# GLANDULAR ODONTOGENIC CYST : A CASE REPORT

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**Glandular odontogenic cyst (GOC) is an extremely rare lesion occurring in the jawbones. Sialo-odontogenic cyst was first described as a multicystic lesion resembling a botryoid odontogenic cyst (BOC) or a central mucoepidermoid carcinoma by Padayachee in 1987. In 1988, Gardner used the term "glandular odontogenic cyst" and considered it as a histologic variant of BOC. Most authors agreed that GOC was odontogenic because of the concurrent ball-like epithelial structure, ameloblastoma, squamous odontogenic tumor-like proliferation in its wall, or hyaline bodies in the epithelium lining. Recently, immunohistochemical studies of the cytokeratin profiles have also supported this concept. Its aggressive behavior and the recurrent tendency make it important. A new case of GOC in a 59-year-old male presented as a multilocular radiolucency in the anterior region of the mandible, invading the marrow space by epithelial islands is described with other clinicopathologic features and the literature is briefly discussed.**

**Key words: glandular odontogenic cyst (GOC), odontogenic cyst, mucoepidermoid carcinoma**

(*Kaohsiung J Med Sci* **16**: 53 — 58, 2000)

Glandular odontogenic cyst (GOC) is an extremely rare lesion occurring in the jawbones. Different names have been used for this lesion. Sialo-odontogenic cyst was first described as a multicystic lesion resembling a botryoid odontogenic cyst (BOC) or central mucoepidermoid carcinoma (MEC) by Padayachee and van Wyk in 1987 [1]. Gardner *et al.* first used the term "glandular odontogenic cyst" because they believed that its epithelium lining was odontogenic and considered it a histologic variant of BOC [2]. The term mucoepidermoid odontogenic cyst or mucus-producing cyst was used by Sadeghi in 1991 due to the microscopic findings of mucus-producing cells, and squamous cells [3]. The term polymorphous odontogenic cyst was used by High et al. in 1996 because of its varied histological appearances [4]. The World Health Organization named GOC as an independent pathologic entity and classified it as a developmental odontogenic epithelial cyst [5]. Most authors agree that GOC is odontogenic because of a concurrent ball-like epithelial structure, ameloblastoma, and squamous odontogenic tumor(SOT)- like proliferation in the cyst wall [2,6-8]. Recently, immunohistochemical studies of the cytokeratin profiles have also supported this concept [9,10].

Clinically, it shows a non-specific swelling in the jawbone and more than half of the cases show multilocular radiolucency. Anterior region of the mandible is the most frequent site of occurrence. A slight predilection for males has been found with a mean age of about 50 years old. Its aggressive behavior and the recurrent tendency make it important. Histopathologically, GOC is lined by nonkeratinized stratified squamous epithe-

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Received: November 29, 1999 Accepted: December 24, 1999
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lium of relatively uniform thickness, which contains glandular structures filled by mucin-like material. The superficial layer of the epithelium consists of eosinophilic cuboidal cells that are sometimes ciliated. Epithelial spheres consisting of swirled epithelial cells have been occasionally found.

In this article, a new case of GOC in a 59-year-old Taiwanese male, presenting as a multilocular radiolucency in the anterior region of the mandible, is described with its clinicopathologic features and the literature is briefly discussed.

## CASE REPORT

A 59-year-old male, Taiwanese farmer came to our dental clinic with the chief complaint of a slowly growing swelling mass for about six months. He found this mass and visited a local dentist for treatment six months prior to visiting our clinic. The dentist performed endodontic treatment for his lower anterior teeth and arranged an operation for this lesion. Finally the patient wanted to be treated at a large hospital, so he visited the dental department of Chang-Gung Memorial Hospital, Kaohsiung, and then was referred to our dental clinic for further treatment with the clinical impression of an ameloblastoma. It appeared as a large swelling measuring 8.0x4.0 cm with a bluish color over the labial aspect of the anterior region of the mandible (Fig. 1). In panoramic radiography, a large multilocular radiolucency from the lower left first premolar to the lower right canine was found (Fig. 2). The patient denied any systemic diseases and neither heart disease nor diabetic mellitus was found. He did not smoke but he chewed betel quid for about ten years and drank moderately for a long time. The clinical impression of an ameloblastoma was made and he was referred to the Department of Oral Surgery for incisional biopsy. A glandular odontogenic cyst was diagnosed and then enucleation was performed four weeks later. During the operation the mandibular lesion showed multiple separated cavities, each of which contained thin fibrous capsules and semi-clear fluid. Trimming of bone of each cavity and iodoform gauze packing were performed. After discharge, an obturator was made. The specimens containing more than 10 soft tissue and thin bone fragments measuring up to 2.2 x 1.0 cm in maximal diameter were sent for microscopic examination. The pathologic diagnosis was a glandular odontogenic cyst.

Histopathologically, it consisted of multiple thin-walled cysts. The epithelium lining varied in thickness from double layer of cuboidal to nonkeratinized stratified squamous epithelium. The epithelial surface consisted of eosinophilic cuboidal, ciliated, and mucus- or goblet-like cells (Fig. 3). Papillary epithelial processes projecting into the lumen were found occasionally but swirl-like epithelial thickening was not found. Within the thick epithelium, there were glandular or cyst-like spaces lined by cells similar to those seen in the epithelial surface (Fig. 4). Mucicarmine staining was positive in the goblet-like cells and glandular spaces (Fig. 5). Most parts of the underlying connective tissue was having mild or even free of inflammatory cell infiltration. Some epithelial islands resembling squamous epithelium containing glandular structures were growing into the surrounding marrow spaces (Fig. 6).

## DISCUSSION

GOC is an extremely rare condition involving the jawbones. Review of the literature and adding the present case yield a total of 64 cases reported in the world. Ramer et al. excluded Wang's seven cases of GOC reported in 1994 in their study because microscopic findings and individual data of these cases were unavailable [11,12]. Hence to date, only 56 cases of histologically confirmed GOC have been reported in the world literature and were included in our analysis [1-4,6-29]. Most of the cases occurred in the mandible (46 case, 82.14%) rather than in the maxilla (10 cases, 17.86%), especially in the anterior region of both jaws. All maxillary cases and 34 of the 46 (73.91%) mandibular cases occurred in the anterior region. Radiographically, nearly 50% of the cases showed a multilocular radiolucency (27/56). Over half (53.57%) of the cases of GOC occurred in the 5th and 6th decades (age of 41-50:16 cases, age of 51-60:14 cases) with a mean age of 49.55 years; male: 48.09 and female: 55.15. A slight prevalence for males with a ratio of 32:24 (1.3:1), and easy recurrence after simple curettage or enucleation with a recurrent rate of 21% were noted. Histopathological findings in the present case fulfilled the diagnostic criteria of GOC originally described by Gardner *et al.* [2] Treatment of GOC included curettage, enucleation, partial or total resection with or without cryotherapy according to most of the authors [3,8,9]. The significant evidence of a locally invasive pattern in previous studies and also in the current case was that epithelial islands invading to the marrow spaces [7,8,21]. In view of the high recurrent rate of this cyst and its aggressive behavior, a surgical removal of the cyst with a rim of uninvolved bone is necessary to pre-

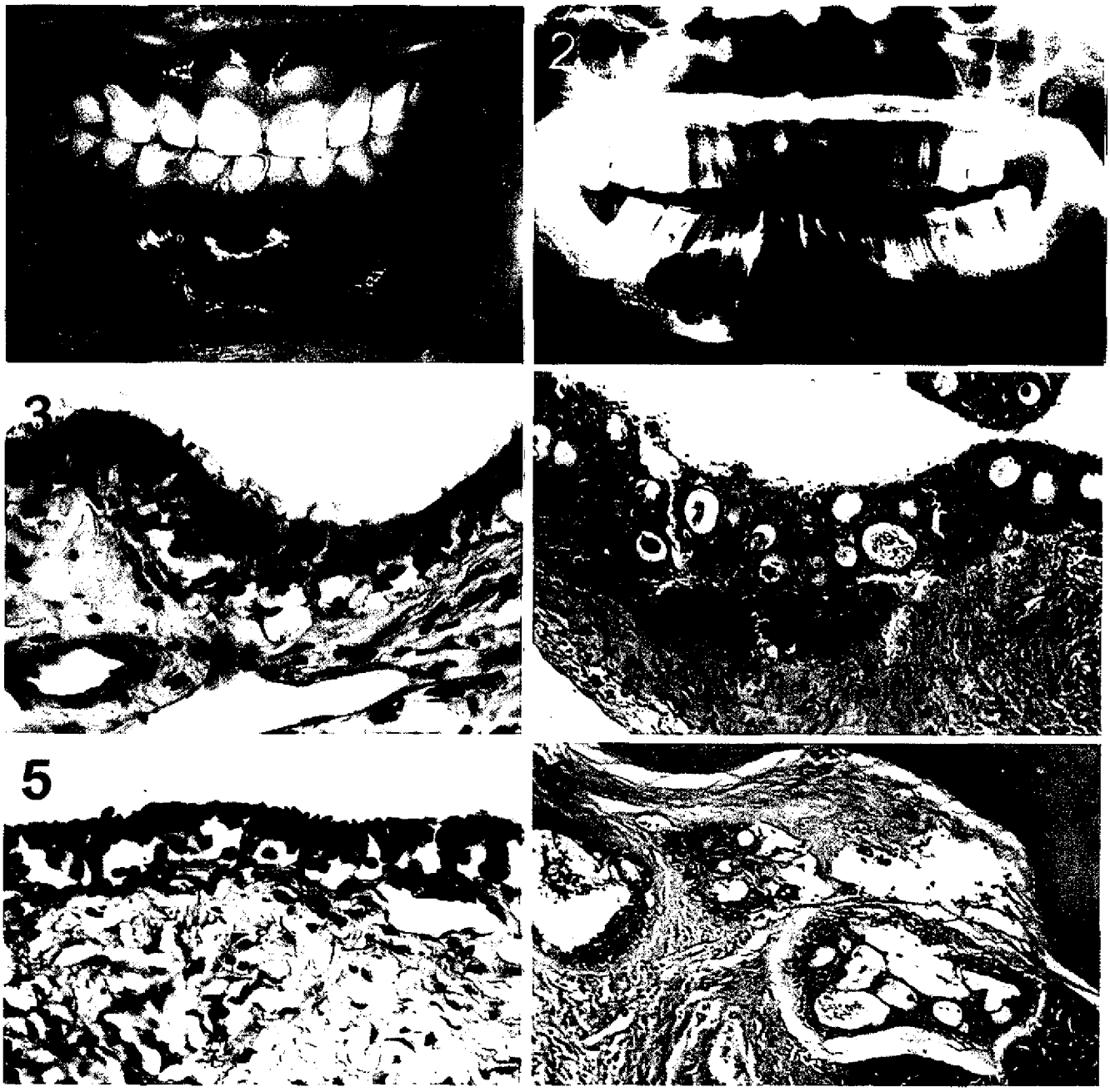


Fig. 1. Multiple separated swellings with dark bluish color over the labial aspect of the anterior mandible. Fig.2. Radiographically, a large multilocular radiolucency from the lower left first premolar to right canine. Fig 3. The surface epithelium consisted of eosinophilic, low cuboidal, ciliated cells.(x 50) Fig.4. Within the non-keratinized squamous epithelium, there were glandular spaces lined by cuboidal cells.(x 20) Fig.5. Mucicarmine staining was positive in goblet-like cells.(x 50) Fig.6. Epithelial islands resembling non-keratinized squamous epithelium containing glandular structures were growing into the surrounding marrow space. (x 20)

vent further recurrence [10].

GOC might clinicopathologically resemble both BOC and the central MEC of jaws, but they were concluded to be different. Some authors suggested that GOC might be a variant of BOC [2,10,13]. In contrast, other authors did not consider them as a same entity [1,3,22]. They thought there were some dissimilarities, such as mucous and ciliated epithelial cells and mucin pools in cystic spaces were either not or rarely found, in BOC. However, the presence of mucous cells in the

epithelial lining of odontogenic cysts should not be considered as a support of the concept of central MEC and intrabone salivary glandular origin, because it might originate from the pluripotentiality of oral epithelium or be due to mucous metaplasia from odontogenic epithelium [3,30,31]. Therefore, it is important to differentiate GOC from central MEC, particularly the low-grade cystic type [21]. However, based on clinical, radiographical, behavioural and histomorphological characteristics, a distinction among GOC, BOC, and MEC seems necessary [19]. The immunohistochemical findings of negative reaction for CK 8 and CK 18 is a potentially important finding that may help differentiate GOC from MEC [10,32]. The presence of CK 13 and CK 19 in Koppang and Matthews' study was thought to be a useful marker for identifying odontogenic epithelium [9,33]. GOC simultaneously occurring with SOT-like proliferation, ameloblastoma, and ghost cell calcifications in previous studies strongly suggests its odontogenic origin [2,6,11]. Furthermore, Ide *et al.* recently reported a case of GOC presenting as a dentigerous cyst and containing hyaline bodies which had been considered exclusive to odontogenic cysts [23]. These findings also support the evidence that GOC is odontogenic in nature.

## ACKNOWLEDGMENTS

The authors are grateful to Dr. Fong-Chang Chen for the financial support of this manuscript.

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# 管狀齒源性囊腫：病例報告

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管狀齒源性囊腫(Glandular odontogenic cyst)是一發生在顎骨罕見之囊腫。在1987 Padayachee首先用Sialo-odontogenic cyst發表病例並敘述它為：多囊性病變、與葡萄狀齒源性囊腫 (botryoid odontogenic cyst, BOC)、或骨內粘液類上皮細胞癌相似。1988年Gardner將此病變命名為"管狀齒源性囊腫"。因為他們相信其上皮襯裏為齒源性，並認為此囊腫是一種葡萄狀齒源性囊腫的組織學上的變体。大部分的學者同意管狀齒源性囊腫為齒源性的，因為它的上皮襯裏可見球狀上皮構造(ball-like epithelial structure)，或同時與造釉細胞瘤(ameloblastoma)存在，或在其壁上有鱗狀齒源性腫瘤樣增生(squamous odontogenic tumor-like proliferation)，或含有玻璃樣體(hyaline bodies)等等。最近的cytokeratin profiles免疫組織化學研究也支持此一說法。其具有侵犯性及容易復發傾向使其變為重要。本篇報告發表一59歲男性，在下顎前牙區呈一多房性放線透射區、囊腫上皮侵犯附近骨髓腔、並敘述其臨床及組織病理學所見及文獻回顧。

(高雄醫誌 **16**: 53 — 58, 2000)

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收文日期：88年11月29日接受刊載：88年12月24日
索取抽印本處：林正仲　高雄市十全一路100號　高雄醫學大學口腔醫學院牙醫學系口腔病理科