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A huge glandular odontogenic cyst occurring at posterior mandible

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ABSTRACT

The glandular odontogenic cyst is a rare lesion described in 1987. It generally occurs at anterior region of mandible in adults over the age of 40 and has a slight tendency to recur. Histopathologically, a cystic cavity lined by a nonkeratinized, stratified squamous, or cuboidal epithelium varying in thickness is found including a superficial layer with glandular or pseudoglandular structures. A 21-year-old male visited Dankook University Dental Hospital with a chief complaint of swelling of the left posterior mandible. Radiographically, a huge multilocular radiolucent lesion involving impacted 3rd molar at the posterior mandible was observed. Buccolingual cortical expansion with partial perforation of buccal cortical bone was also shown. Histopathologically, this lesion was lined by stratified squamous epithelium with glandular structures in areas of plaque-like thickening. The final diagnosis was made as a glandular odontogenic cyst. (***Korean J Oral Maxillofac Radiol 2004; 34 : 209-13***)

KEY WORDS : Mandible; Odontogenic Cysts; Radiography, Panoramic; Tomography, X-ray Computed

The glandular odontogenic cyst is a new entity included in the most recent edition of the WHO's 'Histological Typing of Odontogenic Tumours'.[1] It is defined as 'a cyst arising in the tooth-bearing areas of the jaws and characterized by an epithelial lining with cuboidal or columnar cells both at the surface and lining crypts or cyst-like spaces within the thickness of the epithelium'.[1,2] They are rare and the first description was introduced in 1987 by Padayachee and Van Wyk[3] with two multilocular mandibular cysts. At the time of first description, this cyst was proposed as 'sialo-odontogenic cyst', but it was finally named as glandular odontogenic cyst by Gardner,[4] which is now the preferred term because the possible salivary gland origin of these cysts has not yet been established. In 1992 the World Health Organization accepted glandular odontogenic cyst as a distinct pathological entity and classified it as a developmental odontogenic cyst.[5]

There are some agreements that have been reached on the aggressive, somewhat neoplastic nature of glandular dontogenic cyst and their tendency to recur.[4,6] The prevalence of this lesion was reported to be 0.012% to 1.3%.[2,7] Reported studies help to understand general characteristics of the lesion. It is a slow growing lesion and the age range was reported to be 14 to 85 years, with a mean age of 49 years and the majority of the patients were older than 40 years old.[8] The male to female ratio is equal or slightly higher in males and most of the lesion were observed at the anterior mandible through premolar area.[9] The most predominant clinical finding was the presence of a painless swelling.[2-9] Previous report presented fine-needle aspiration, electrophoresis, and exfoliative cytologic examination of the cyst contents might help differentiate glandular odontogenic cysts from other odontogenic cysts.[10] And the recurrence rate after treatment was reported to be about 25% to 55%.[11] The lesion is localized and occurred uni-, or bilaterally. Its size is small to moderate. Radiographically, the lesion is round to oval shape and it has well-defined and sometimes smooth or scalloped margin. And it shows uni- or multilocular radiolucency. The adjacent teeth usually respond positively to vitality test but, slight displacement of teeth and loss of lamina dura, can also be seen.[2-4,6,8,9] Expansion and thinning of cortex can be observed with perforation, which rarely occurred, on radiographs.[11]

The main histopathologic features are as follows: a cystic cavity lined by a nonkeratinized, stratified, squamous epithelium varying in thickness; localized palquelike thickenings of the epithelium; the presence of variable numbers of mucus-secreting cells in the surface layer of the epithelium, sometimes forming cryptlike invaginations or glandlike areas within the layer of epithelium; a subepithelial fibrous tissue tendency; the presence of multiple cysts, some of which are within adjacent bone marrow spaces; and an absence of

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inflammation.[10]

The differential diagnosis of a slowly growing radiolucent lesion of the mandible includes cysts (lateral periodontal, botryoid odontogenic cyst, simple bone cyst, and odontogenic keratocyst), ameloblastoma, and central mucoepidermoid carcinoma.[4,12]

Several methods of treatment of glandular odontogenic cysts include currettage, enucleation, and local block excision. In view of high recurrence rate, which is still on controversial, associated with conservative treatment of these cysts and their invasive potential, some suggest local en bloc excision with primary reconstruction.[10] And others recommend that conservative surgery followed by meticulous long-term review is the treatment of choice.[2]

The occurrence of the glandular odontogenic cyst is considered to be uncommon. Total fifty-five cases have been reported in the literature reviews that have been published so far.[1-17] In this article, we report a case of glandular odontogenic cyst occurring at posterior mandible to add to more knowledge about this rare entity.

Case report

A 21-year-old male patient was referred to Dankook University Dental Hospital for the management of a painless swelling in lower left cheek. He complained of the swelling having grown since 6 months ago. Clinical examination showed a bony hard swelling at buccal side of left posterior mandible with facial asymmetry.

Panoramic radiographic examination showed huge, multilocular radiolucent lesion with partially scalloped border involving left mandibular body and ramus (Fig. 1). Displacement of the impacted lower left third molar and external root resorption of #35, 36, 37 could be seen. Computed tomographic axial and frontal views showed bony expansion of the lingual and buccal cortex, with thinning and partial perforation and extension of the lesion into the buccal soft tissues (Fig. 2A, B, C, D). Provisional diagnosis, based on the clinical and radiographic examinations, was an ameloblastoma.

Through the incisional biopsy at the department of oral and maxillofacial surgery, this lesion was reported as glandular odontogenic cyst. Enucleation of the lesion was performed and mandible was reconstructed with metal plate under general anesthsia. During the operation, excisional biopsy was made and it was submitted for histopathologic analysis. Histopathologic findings were that the lesion was lined by nonkeratinized stratified squamous epithelium and thick plaquelike area within epithelium lining, and glandular cell could be observed (Fig. 3). The final diagnosis was made as glandular odontogenic cyst. The patient is on follow-up periodic check and there has not been any radiographic evidence of recurrence on follow-up radiograph after 2 years of operation (Fig. 4).

Discussion

This case does not fulfill the general clinico-radiographic appearance of the glandular odontogenic cyst reported previously, however, there were some reports to support this case as a glandular odontogenic cyst. Toida and co-workers[8] in their review of the literature mentioned that the lesion lacked specific features making distinction from ameloblastoma and odontogenic keratocyst radiographically. And more aggressive

Fig. 1. Panoramic radiograph shows multilocular radiolucent lesion with partially scalloped border involving left mandibular body and ramus.

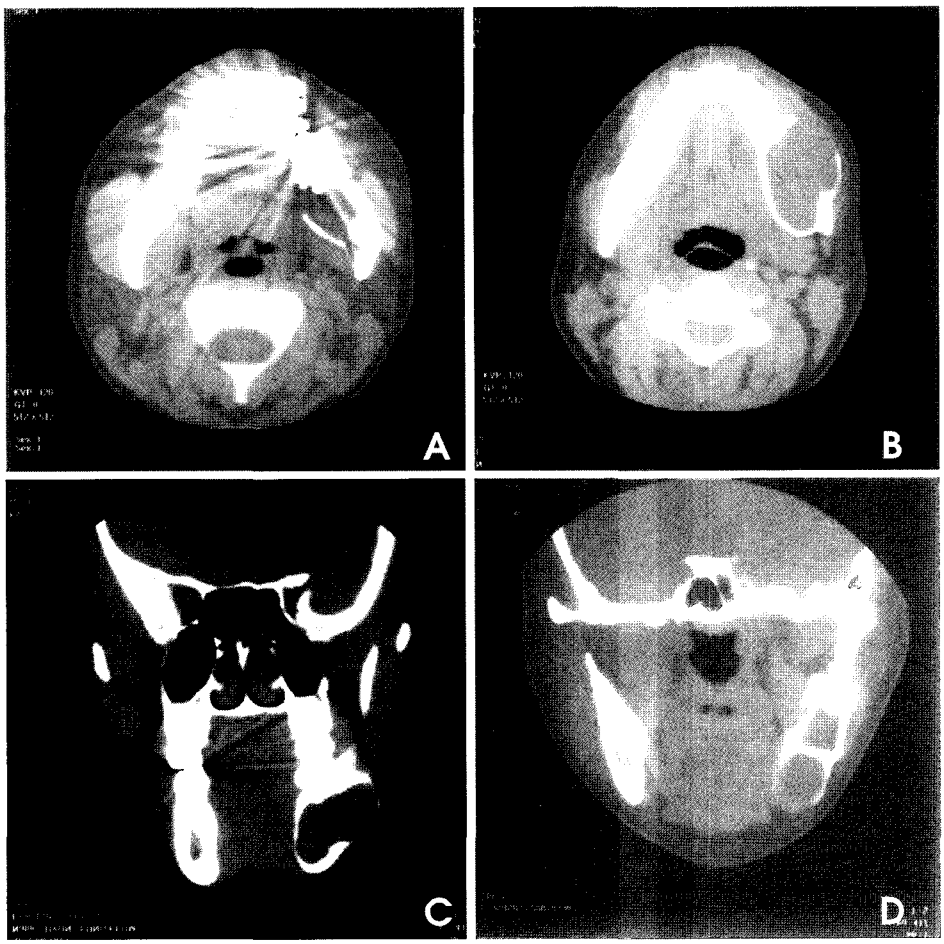

Fig. 2. A, B, C, D. Axial and frontal computed tomographic images show bony expansion of the lingual and buccal cortex, with thinning and partial perforation and extension of the lesion into the buccal soft tissues.

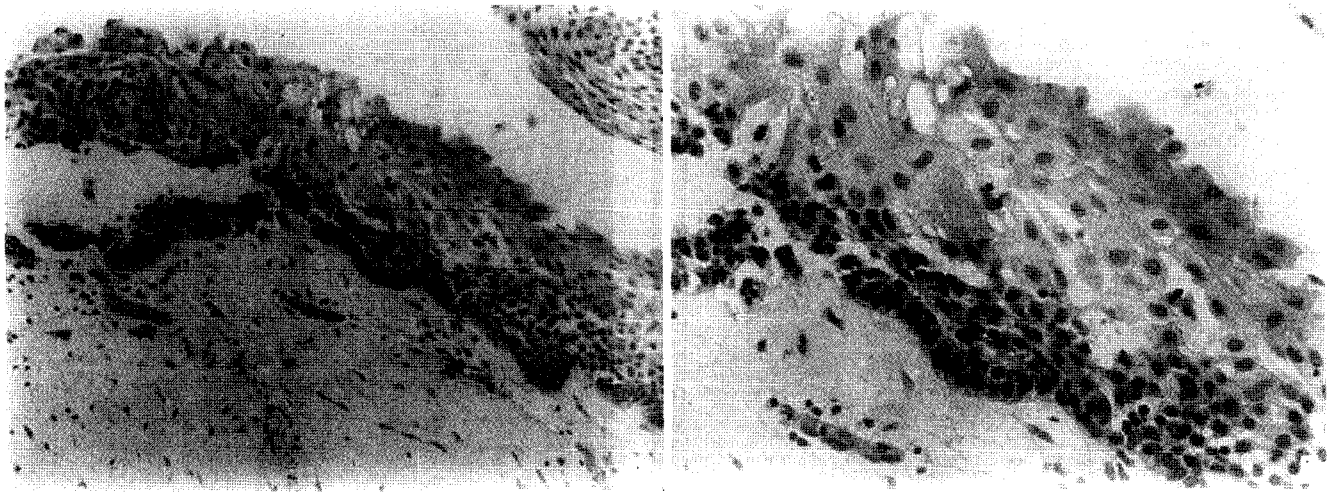

Fig. 3. A, B. Nonkeratinized stratified squamous epithelium, thick plaque-like area within epithelium lining, and glandular cells are shown (H&E stain, A. ×200, B. ×400).

surgical removal rather than simple curettage was sug-gested and careful follow-up was strongly recommended. The cysts might reach large dimension, often associated with expansion. And radiological findings were reported to be non-specific. In 1994 Takeda[17] reported a glandular odontogenic cyst in the mandibular third molar region. Noffke and Rau-benheimer[5]

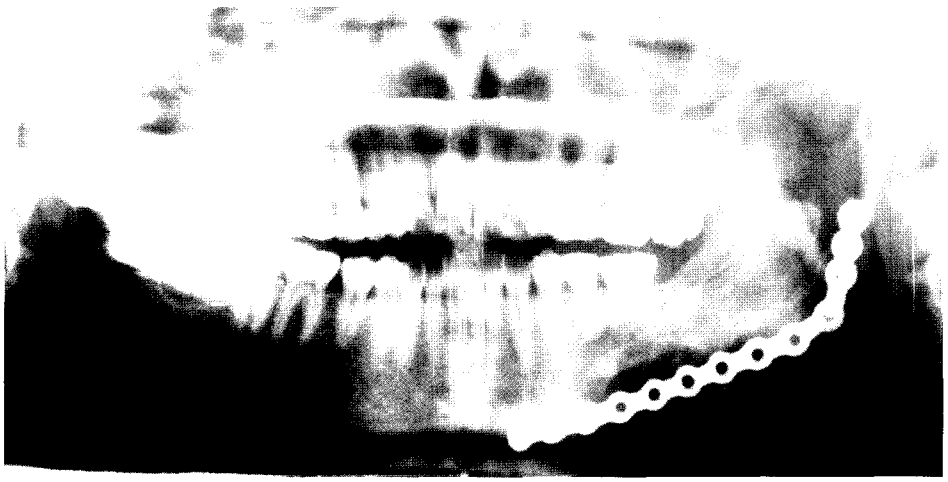

Fig. 4. Follow-up panoramic radiograph after 2 years of operation does not show any evidence of recurrence.

noted in their review that mean age (35 years) was a decade younger than generally reported mainly due to singnificantly younger average age of 24 years at presentation of their male patients. And this tendency is in agreement with the literature where males are generally reported to be affected at a younger age. One of their cases occurred in the molar area of the mandible and extended into the ramus.[5] And they reported that all glandular odontogenic cysts measured in excess of 6 cm showed bone expansion with perforation, a feature supporting their aggressive expansile behaviour. Ertas, et al.[14] reported another case involving large area of mandible. The clinical and the radiological aspects of our case were partially different from others reported for glandular odontogenic cyst. However, the typical histopathological findings, that is, a cystic cavity lined by a nonkeratinized, stratified, squamous epithelium varing in thickness was found with localized plaque-like thickening of the epithelium. Variable number of glandular cells in the surface layer of the epithelium were present.

Several methods of treatment of glandular odontogenic cyst are curettage, enucleation, local block excision, and marginal resection.[17] In this case, the treatment was a careful and complete enucleation. Although there was no recurrence at the time of 2 years after enucleation, it is imperative the patient be followed carefully for a long time.

The glandular odontogenic cyst remains a rare lesion but should be considered in the diffential diagnosis of unilocular and multilocular radiolucencies of the jaws. And we suggest computed tomographic examination to distinguish it from other odontogenic cysts, tumors, and mucoepidermoid carcinoma. It is important that new cases be documented and followed carefully so that more accurate conclusions can be drawn concerning optimal treatment.

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