

# An Unusual Odontogenic Cyst with Diverse Histologic Features

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An unusual odontogenic cyst, which was originally believed to be a clinical dentigerous cyst associated with an impacted mandibular third molar, was found histologically to demonstrate the characteristics of a glandular odontogenic cyst with para- and orthokeratinization. These histologic diversities were interpreted as a reflection of the pluripotentiality of the epithelial remnants of the mandibular third molars or dentigerous cyst epithelium. It is possible that it has the capacity to induce the formation of cysts in both squamous and glandular epithelium.

**Key Words:** Glandular odontogenic cyst, odontogenic keratocyst, dentigerous cyst, mandible

## INTRODUCTION

Considerable variation in the nature of the epithelial lining of cysts of the jaws has been observed.<sup>1</sup> Although the luminal surface of odontogenic cysts is usually lined by squamous epithelium, ciliated or mucous cells are occasionally found in the linings.<sup>2</sup> Over the years there have been sporadic reports of unusual cystic lesions of the jaws, which have not been readily classified under conventional headings.<sup>3</sup> Of these, the glandular odontogenic cyst (GOC) is a recently recognized developmental odontogenic cyst, with diverse histologic features.<sup>4</sup> Herein, an unusual odontogenic cyst, presenting as a dentigerous cyst and showing diverse histologic features of cili-

ated, glandular and squamous epithelium with keratinization, is reported.

## CASE REPORT

A 66-year-old Korean woman presented with a painful swelling of the right mandible that had been present for 5 months. The patient's medical history was not otherwise significant. An oral examination disclosed a hard bony swelling in the right mandible and a panoramic radiograph revealed a well-defined unilocular radiolucency around the impacted third molar, with a thin sclerotic margin. There was a root resorption of the right mandibular first molar tooth.

Computed tomogram demonstrated a well-demarcated cystic lesion, with bucco-lingual expansion of the right mandible, including an impacted third molar tooth (Fig. 1). A provisional diagnosis of an odontogenic keratocyst (OKC) or a unicystic ameloblastoma was made, and a cyst enucleation performed. The postoperative course was uneventful, and there was no recurrence during the 1-year follow-up period.

Microscopically, the cyst was multilocular, with thin or thick fibrous septa. It was lined by epithelium, with varying morphologic features and a smooth epithelial to connective interface devoid of inflammation. The epithelium was predominantly thin and non-keratinized squamous, with cuboidal or ciliated epithelium and an epithelial plaque of swirl-like arrangement (Fig. 2A). In addition, there were papillary projections, microcysts or duct-like structures, along with clusters of goblet cells (Fig.

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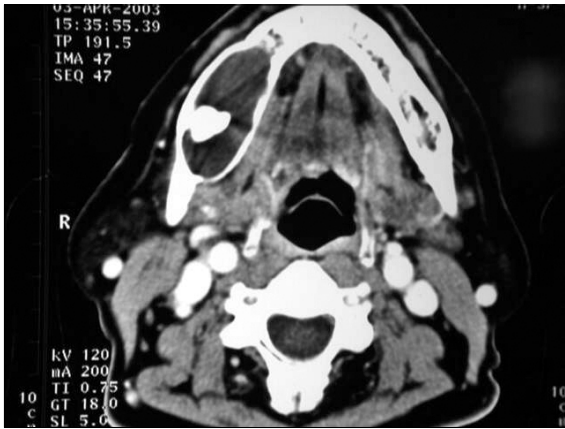


Fig. 1. Computed tomogram demonstrating a well demarcated cystic lesion with buccolingual expansion of the right mandible. A third molar tooth is impacted within the lesion.

2B). The mucus cells reacted positively to mucicarmine stain. Small satellite cysts, which showed similar features, appeared also to be entrapped in the connective tissue wall. In addition to the glandular structures, some areas of the lining epithelium were composed of a uniform layer of stratified squamous epithelium, with a hyperchromatic, palisaded basal cell layer and a corrugated parakeratotic surface (Fig. 3A). A focal area of prominent keratohyaline granules beneath the orthokeratotic surface was also observed (Fig. 3B).

### DISCUSSION

The cyst described in this report mainly exhi

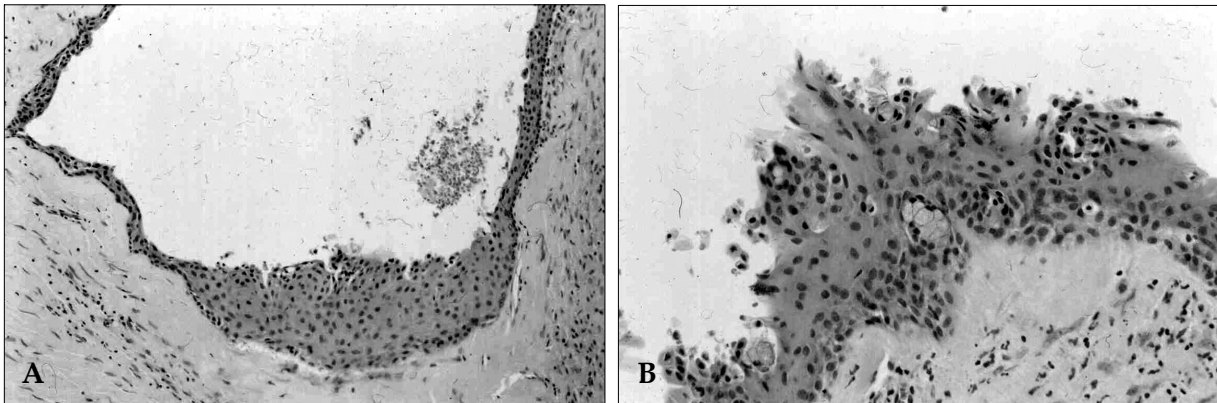


Fig. 2. A. Photomicrograph showing a non-keratinized squamous epithelium with cuboidal surface cells and an epithelial plaque of swirl-like arrangement (H-E,  $\times 100$ ). B. The cyst lining showing papillary projections, duct-like structures and clusters of goblet cells, resembling a glandular acinus (H-E,  $\times 200$ ).

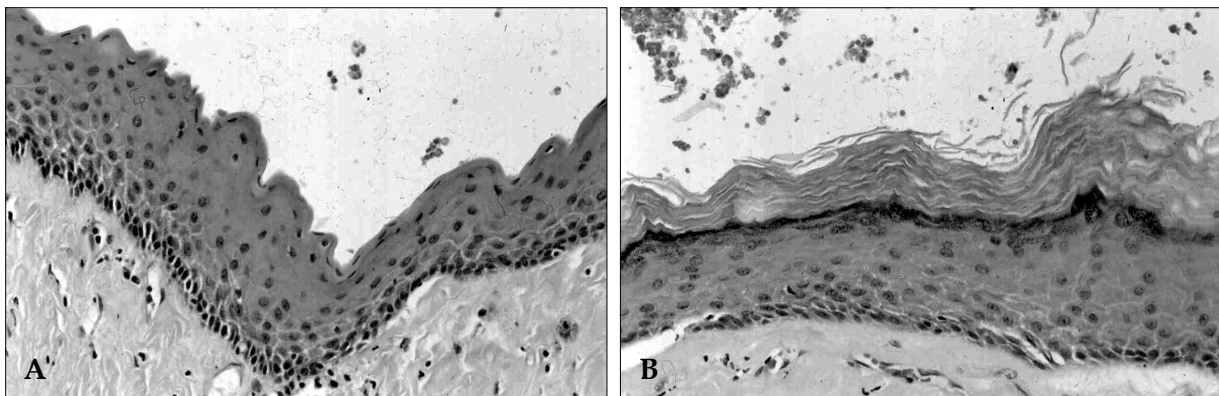


Fig. 3. A. The lining epithelium is composed of a uniform layer of stratified squamous epithelium, with a hyperchromatic and palisaded basal cell layer. There is a corrugated parakeratotic surface, showing typical features of odontogenic keratocysts (H-E,  $\times 200$ ). B. The epithelial lining showing prominent keratohyaline granules beneath the orthokeratotic surface (H-E,  $\times 200$ ).

bited the characteristic histopathological features of a glandular odontogenic cyst. However, the most interesting aspect of this lesion was its association with an impacted tooth, presenting as a dentigerous cyst, and the presence of a prominent histological component of an OKC. While the presence of a narrow cuboidal or reduced enamel epithelium-like lining in part of the lesion associated with an impacted tooth crown, was suggestive of a dentigerous cyst. Nevertheless, its extensive intramedullary growth, root resorption of the adjacent tooth, the presence of significant glandular elements as well as the parakeratotic and orthokeratotic surface of its lining, make it less acceptable as a dentigerous cyst. Based upon the histopathological findings, this peculiar case was regarded to be a GOC, with features of an OKC.

The possible pathogenic mechanisms would seem to be either a collision of two separate lesions or a transformation of one lesion to another. The collision of two separate cysts appears unlikely in this case, as both the GOC's lining epithelium showed direct continuity to that of the OKC. It is more likely that this was a single process that manifested two distinct types of odontogenic cyst.

Therefore, the main question is, does the lining epithelium of a GOC show para- and orthokeratinization? The epithelial lining of a GOC may be able to induce an ameloblastomatous and squamous odontogenic tumor-like proliferation in the connective tissue wall.<sup>4,5</sup> To our knowledge, there have been no reports of a histological transformation or differentiation to a para- or orthokeratotic epithelium. High et al.<sup>3</sup> reported a similar case. In contrast to the present case, one small area of keratinization was observed, but the associated basal layer was cuboidal without any polarity of the nuclei. Alternatively, it is possible that the OKC had undergone glandular metaplasia. It was previously suggested that the epithelial lining of the odontogenic cysts can undergo metaplasia from a stratified squamous to a more highly differentiated ciliated columnar or glandular type.<sup>2,6</sup> However, only four cases of OKC with respiratory epithelium have previously been reported.<sup>1,7,8</sup> In contrast to maxillary OKCs, where the metaplasia can be related to the anatomi-

cal proximity to the respiratory tracts; however, the mandibular OKC was barely resolvable.<sup>1,7,8</sup>

Considerable variation in the nature of the epithelial lining of cysts of the jaws has been observed.<sup>1</sup> It has been previously suggested that epithelial inclusions in the region of the mandibular third molars might represent pluripotentiality.<sup>9,10</sup> In addition, the pluripotentiality of the epithelium in mandibular dentigerous cysts was further emphasized by Gorlin.<sup>11</sup> In view of the multipotentiality of the odontogenic epithelium around the mandibular third molars, it is possible that it would have the capacity of inducing the formation of cysts with both the squamous and glandular epithelia. The possible pathogenic mechanism of this case would appear to be a reflection of the pluripotential character of the odontogenic epithelium. Therefore, this case indicates that the multipotential odontogenic epithelial tissue has the ability to develop diverse differentiation. The relationship, if any, of both GOC and OKC should become clearer as more examples of each are examined. The prognosis and biological behavior in this case will probably be as expected for a GOC.

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