A dentigerous cyst (DC) and an odontogenic fibroma (OF) in the jaws are well known entities, which have been reviewed extensively. However, the presence of OF components in the wall of dentigerous cyst is a very rare event. A case of DC in the left mandible displaying the foci of an OF-like lesion is described. It is unclear whether the two lesions were just coincidental or were actually related to each other. However, OF-like proliferation may be arisen from secondary epithelial and mesenchymal interactions in a pre-existing DC, although the initiating stimulus could not be identified.

Key words: Dentigerous cyst, Odontogenic Fibroma, Mandible

I. Introduction

The odontogenic fibroma (OF) is a rare neoplasm of odontogenic ectomesenchyme characterized by varying amounts of inactive-looking odontogenic epithelium embedded in a mature, fibrous stroma. Although it was recognized as a distinct pathologic entity, a variety of histopathologic features have been described. Histological variants of OF include granular cell, pleomorphic fibroblast and giant cell granuloma (GCG)-like variants.

Dentigerous cyst (DC) is the most common developmental odontogenic cyst in the jaws. The DC may be co-existed with other pathologies including odontoma and GCG. A DC and an OF in the jaws are well known entities, which have been reviewed extensively. However, the presence of OF components in the wall of odontogenic cyst is a very rare event, with only two reported cases. In this report, we describe a case of DC displaying the foci of an OF-like proliferation in the connective tissue wall.

II. Case Report

A 23-year-old Korean woman presented with a swelling of the left mandible, which was painful and had been present for 3 months. Oral examination disclosed a slight swelling in the left third molar region. Radiographic examination showed a well-defined unilocular radiolucency around the impacted third molar with a thin
sclerotic margin in the posterior portion of the left mandible (Fig. 1). A provisional diagnosis of a DC or a paradental cyst was made and a cyst enucleation was performed. The post-operative course was uneventful, and there was no recurrence during the 5 years follow-up period.

Microscopic examination revealed that the epithelial lining was composed of flattened cuboidal or non-keratinized squamous epithelium (Fig. 2A). In some areas, there were epithelial proliferations and scattered epithelial islands and inflammatory cell infiltration in the cyst wall. In addition, localized foci of a more cellular fibroblastic tissue with collagen fibers arranged in interlacing bundles that containing varying amounts of odontogenic epithelial rests and extensive calcified materials was observed in the connective tissue cyst wall, resembling an OF (Fig. 2B). Some odontogenic epithelium in the form of long strands or small nests showing clear cytoplasm was present throughout the lesion (Fig. 2C). Calcifications composed of cementum-like material, osteoid or dentinoid were present within the cellular fibrous tissue (Fig. 2D).

Fig. 1. Panoramic radiograph of a well-defined unilocular radiolucency around the impacted third molar with a thin sclerotic margin in the posterior portion of the left mandible.

Fig. 2. Photomicrographs of an OF-like proliferation in the cyst wall of a DC. A. The epithelial lining composed of flattened cuboidal or non-keratinized squamous epithelium. Note the localized foci of the cellular fibroblastic tissue containing small odontogenic epithelial rests and numerous calcifications beneath the epithelial lining. B. A more cellular fibroblastic tissue containing varying amounts of odontogenic epithelial rests (arrow) and cementum-like calcifications in the connective tissue cyst wall. C. Odontogenic epithelium showing clear cytoplasm (arrow) within the collagen fibers arranged in interlacing bundles. D. Calcifications composed of cementum-like material, osteoid or dentinoid were present within the cellular fibrous tissue.
III. Discussion

The most interesting aspect of this lesion was the presence of a prominent histological component that resembled an OF in the cyst wall of a DC. Normal dental follicles associated with unerupted teeth are frequently misinterpreted histologically as an OF. However, dental follicular tissues do not possess the cellular fibroblastic connective tissue arranged in interwoven strands that is typical of the OF. Therefore, we interpreted this case as an OF rather than dental follicular tissue in the wall of a DC.

Two cases of an OF associated with odontogenic cyst have been reported. Jones et al. described typical areas of OF associated with a small inflamed cyst lined by epithelium containing hyaline bodies. Cercadillo-Ibarguren et al. reported locally aggressive odontogenic fibroma associated to an inflammatory odontogenic cyst. In the case presented as similar to previously reported cases, the cyst exhibited unique histologic features in stromal components. There was an active proliferation of spindled cells and numerous calcifications in the cyst wall. Since that both epithelial and stromal proliferation was encountered, it can be speculated that the epithelial and mesenchymal cells reciprocally stimulated each other, as it occurs in tooth germ. Additionally, there is a possibility exists that proliferating odontogenic epithelial islands may develop further odontogenic tumours as a result of an inductive effect by the odontogenic epithelium on the adjacent mesenchymal tissue. We do not believe that this lesion is two separate lesions that grew into one another. We thought this is one pathologic process that is manifesting two types of odontogenic lesions. The reason we thought this is correct is based upon the observation that some of the epithelial islands may be present in the fibrous connective tissue wall of DC. Therefore, OF-like proliferation may be arised from secondary epithelial and mesenchymal interactions in a pre-existing DC, although the initiating stimulus could not be identified.

IV. References
