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염증성 치근단낭종의 소견을 보이는 이형성 치성낭종

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〈Abstract〉

Dysplastic Odontogenic Cyst with the Manifestation of Inflammatory Radicular Cyst

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This report describes a case of odontogenic cyst with keratinization and dysplastic change of lining epithelium, which showed the manifestation of inflammatory radicular cyst, clinically. A 28-year-old man complained of dull pain in the right mandibular molar region. Radiographically a well-defined oval cystic lesion with non-vital teeth, a common finding in radicular cyst, was observed. Microscopically, the lining epithelium of the cyst demonstrated both keratinization and severe epithelial dysplasia. Atypical findings such as hyperchromatic nuclei, increase of N/C ratio and drop shaped rege ridge were observed in the lining epithelium. However, definite invasion into fibrous connective tissue was not found. Immunohistochemically, the dysplastic lining epithelium was highly positive for proliferative marker, Ki-67. Based on the dysplastic changes of lining epithelium, this periapical lesion would be considered to be signs of malignant change. From this case, we conclude that definitive diagnosis by microscopical examination should be made, even if the periapical lesion would be clinically considered as inflammatory radicular cyst.

Key words : Epithelial dysplasia, Odontogenic keratocyst, Radicular cyst.

I. INTRODUCTION

Squamous cell carcinomas (SCC) rarely arises from the epithelial lining of odontogenic cyst, termed as primary intraosseous squamous cell carcinoma (PISCC), mainly derived from long-standing odontogenic cysts¹⁻⁴⁾. The purpose of this report is to describe a rare case of keratinization and dysplastic changes of lining epithelium in odontogenic cyst which is periapically located. Proliferating activity of this lesion was measured and the relationship between keratinization and malignant transformation was discussed.

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* We would like to thank Ms Yurika Kawahara for her assistance, and Associate Professor Jeremy Williams (Laboratory of Dental Information, Tokyo Dental College) for his help with English. This work was supported by the Priority Research Centers Program through the National Research Foundation of Korea (NRF) funded by the Ministry of Education, Science and Technology (2010-0029702).

Received: May 6, 2011; Revised: May 17, 2011; Accepted: May 27, 2011

II. CASE REPORT

A 28-year-old man visited a dental office complaining of a dull pain in the right mandible. An intraoral examination disclosed a tender swelling of the buccal vestibule, encompassing the right mandibular first premolar to the second molar, and discharge of pus from the buccal vestibule (Fig. 1A). The clinical facial examination revealed no noticeable abnormalities, and the patient's medical history was unremarkable. A panoramic radiograph examination revealed a relatively well-defined oval radiolucent lesion at the right mandible, together with root resorption at the first and second premolars and first molar of the right mandible, resembling an odontogenic tumor (Fig. 1B). Computerized tomography revealed relatively well-defined oval lesion without perforation of mandibular bone (Fig. 1C). The teeth were non-vital, as a result of an earlier pulpotomy at a local clinic. In order to confirm that the lesion is periapical periodontitis or odontogenic tumor, an incisional biopsy was performed. Histopathological examination revealed severe inflammatory cell infiltration with a small fragment of the lining epithelium, and a histopathological diagnosis of radicular cyst was reached (Fig. 2A). Although root canal treatment had been carried out, the lesion was refractory and the signs and symptoms continued to persist. Finally, both surgical enucleation of the lesion and root-end surgery of the 3 teeth involved were performed. On histopathological examination, the lining epithelium showed thickened stratified squamous epithelium with partly orthokeratinization with definite granular cell layer and partly parakeratinization (Fig. 2B). Characteristically, epithelial dysplasia such as hyperchromatic nuclei, increase of N/C ratio, loss of polarization and dropping off of bulbous rete ridge were observed in the lining epithelium (Fig. 2C). However, definite invasion of the epithelial lining into fibrous con-

nective tissue was not seen. Immunohistochemically, the dysplastic lining epithelial cells were positive for proliferative marker Ki-67 (Fig. 2D), and the labeling index was 13.3% (± 6.7). Pathologically, the lesion was diagnosed as inflammatory radicular cyst with keratinization with severe dysplasia. The patient has been under regular review for 7 years, with no sign of recurrence.

III. DISCUSSION

The incidence of malignant change in odontogenic cysts ranges from 0.3 to 3%⁵⁻¹¹. Dysplastic lining was seen in one fourth of PISCC from odontogenic cyst in a review by Eversole *et al.*¹², and dysplastic transitional area has been demonstrated by Browne and Gough preceding the development of carcinoma¹³. Long-standing chronic inflammation has been proposed as the principle predisposing factor of malignant transformation in the lining epithelium of the cyst¹². Several reports emphasized that keratinization of the lining epithelium is more often associated with high risk of malignant transformation^{6,7,13}. Browne and Gough investigated that keratin metaplasia, followed by epithelial hyperplasia and epithelial dysplasia of cyst epithelia, were the significant events in the development of PISCCs from odontogenic cysts¹³.

Furthermore, Schwimmer *et al.* reviewed 56 cysts associated with PISCC derived from odontogenic cyst and reported 17.8% were keratinized residual radicular cyst⁶. In this case, both orthokeratinization and parakeratinization of lining epithelium were also seen. However, whether keratinization in inflammatory odontogenic cyst can be a marker for malignancy potential should be validated by further study, in that malignant transformation can occur in non-keratinizing odontogenic cysts as well and furthermore orthokeratinized epithelium can be also found

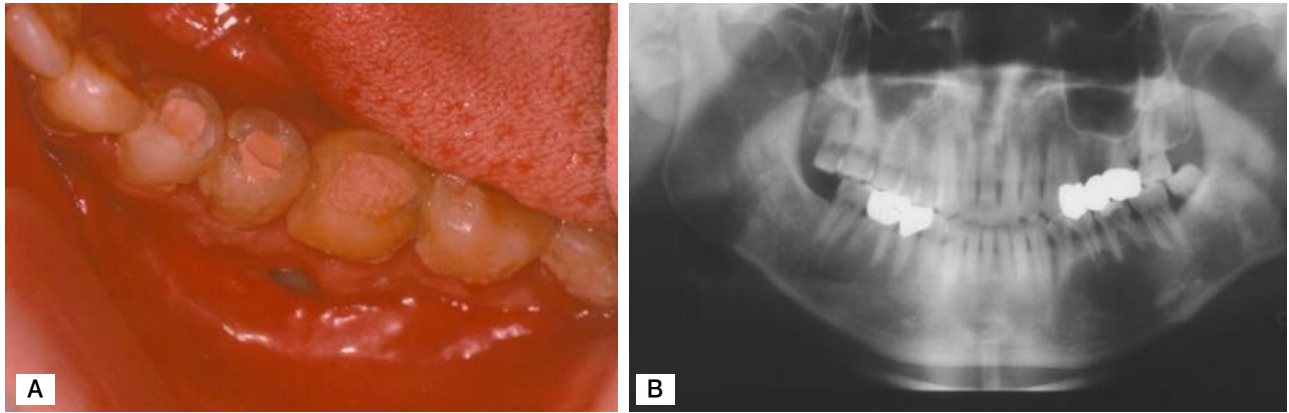


Fig. 1. (A) Intraoral photograph: Pus drainage, swelling and tenderness are examined in buccal vestibule from the right mandibular second premolar to the second molar. (B) Panoramic view showing relatively well-defined radiolucent lesion in the right mandible. (C) Computerized tomography revealed relatively well-defined oval lesion without perforation of cortical plate.

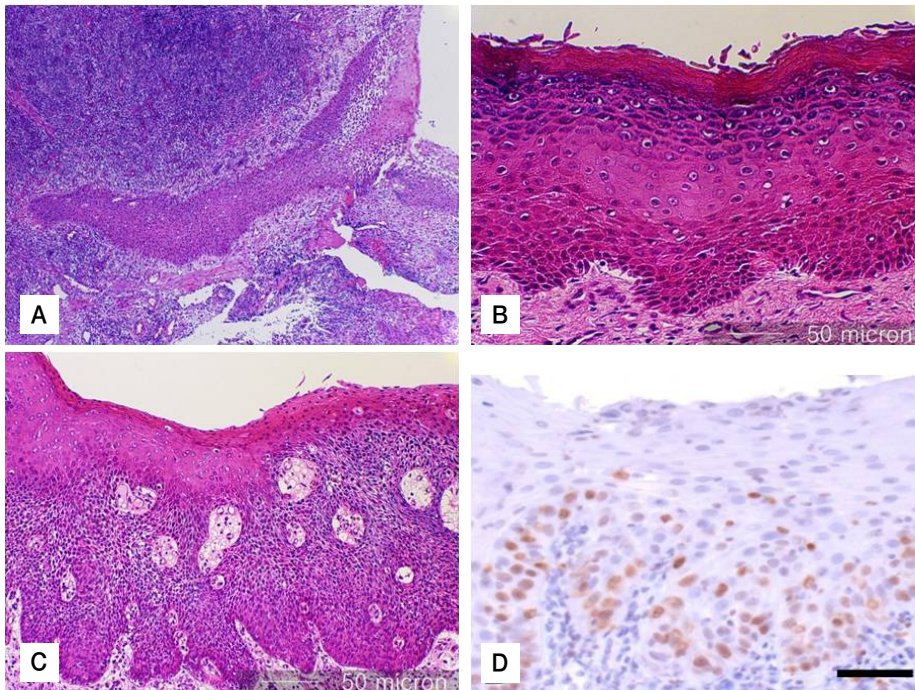


Fig. 2. (A) Incisional biopsied tissue: Small fragment of lining epithelium with severe inflammation observed in underlying connective tissue wall; epithelium revealed neither epithelial atypia nor dysplasia in the specimen. (Hematoxylin-eosin stain, original magnification; $\times 25$) (B) Enucleated tissue: Lining epithelium was orthokeratinized with granular layer, (C) Severe epithelial dysplasia showing bulbous rete ridge was seen. Cellular atypism such as nuclear hyperchromatism and pleomorphism, increased N/C was found at higher magnification (Inset). (D) Immunohistochemically, Ki-67 positive cells were seen in basal cell layer and suprabasal layer of the lining epithelium. (original magnification; $\times 100$)

in odontogenic cysts without malignant transformation, even it is rare^{14,15}.

To evaluate the malignant potential of this lesion, we measured proliferating activity of this lesion by immunohistochemical staining of Ki-67, which is commonly used for evaluation of cell proliferative activity. Ki-67-positive cells were observed in dysplastic lining epithelium in this case. Some reports have showed proliferative activity in lining epithelia of odontogenic cysts¹⁶⁻¹⁹. Suzuki *et al.*(2005) reported that Ki-67 labeling index (positive cells / total cells) ranged from 2.62 to 3.94 in lining epithelium of radicular cyst¹⁶. Gadbaill *et al.*(2009) reported that Ki-67 labeling index ranged from 2.87 to 7.13, and mentioned Ki-67 expression was related to elongated rete pegs and increased thickness of lining epithelium¹⁷. These earlier reports suggested that proliferative activity might be related to degree of inflammatory reaction^{16,17}. In this case, Ki-67 labeling index in basal cell layer and suprabasal layer were higher than that of earlier reports^{16,17}. This unusual proliferation in lining epithelium suggests that this lesion harbors the potential to transform to malignancy.

In conclusion, we strongly suggest that definitive diagnosis by microscopical examination should be made, even if the periapical lesion would be clinically considered as inflammatory radicular cyst.

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