Aberrant Ossification in Basophilic Ghost Cells of Calcifying Odontogenic Cyst Resembling to Pilomatricoma

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Although the mechanism of the abnormal calcification in the calcifying odontogenic cyst (COC) was not elucidated so far, it has been known that the ghost cells are closely related to the calcification, producing dystrophic globular cementum-like materials, comparable to pilomatricoma in epithelium1). Here, we presented a case of COC occurred in left maxillary canine area of 23 years old female, exhibiting a collection of aberrant ossification admixed with basophilic ghost cells in comparison with seven cases of COC. In the polarizing microscope observation with Masson trichrome stain the present case clearly disclosed the typical birefringence of bony tissue, stained red in von Gieson stain, indicating the collagenous backbone. Some ghost cells showed the features of interdigitating epithelial attachments, empty spaces of nuclei, and reticular basophilic cytoplasms, which were similar to the basophilic ghost cells of philomatricoma. The present case demonstrated the aberrant ossification by basophilic ghost cells in COC similar to the ossification of pilomatricoma.

Key words: Calcifying odontogenic cyst, Ossification, Ghost cells, Pilomatricoma

Ⅰ. Introduction

The calcifying odontogenic cyst (COC) was first described as a distinct entity by Gorlin et al in 19622). COCs are lesions of odontogenic origin that may arise as primary lesions from the remains of the dental epithelium or secondarily from the lining epithelium of pre-existing lesions3), and be broadly classified into two main entities: cysts and neoplasms. The COC is also characterized to be an oral analog of pilomatricoma4,5). The World Health Organization (WHO) classified COC as an odontogenic tumor in 1992 based on the "monistic" concept, that all COCs are neoplastic in nature, even though the majority are cystic in architecture and appear to be non-neoplastic. Current thinking favors strongly the "dualistic" concept that COC contains two entities: a cyst and a neoplasm6).

The COC may occur in association with other odontogenic tumors, the most common is the odontoma, occurring in about 24% of the cases7), exhibiting a greater
amount of luminal and mural dentinoid as well as luminal ghost cells. Histopathologically, COCs showed thin or thick lining epithelium with ghost cells, COCs were classified according to proliferative type or nonproliferative type lining epithelium, the presence or absence of ameloblastomatous appearance, and the presence or absence of odontoma in the cyst walls. The cytoplasms of ghost cells in COC were usually eosinophilic and demonstrated distinct immunolocalization of the enamel-related proteins. Dentinogenic ghost cell tumor accompanied with COC was described in terms of its clinical, histological, immunohistochemical, lectin binding and biophysical properties. The lesion comprising dentinogenic ghost cell tumor and COC contained odontogenic epithelium with ghost cells, eosinophilic amorphous materials and osteodentin. Some of the eosinophilic materials had undergone transformation into osteodentin.

The present case of COC demonstrated the aberrant ossification closely associated with basophilic ghost cells, which were histologically different from the ordinary ghost cells of COCs so far but resembled to the ghost cells of pilomatricoma. The biological status of the basophilic ghost cells of COC was discussed in comparison with other seven cases of COC.

### II. Materials and methods

Totally eight cases of COC filed in the Department of Oral Pathology, Gangneung-Wonju National University Dental Hospital (GWNUDH) were examined using histochemical stainings and polarizing microscope observation. The surgically removed specimens were fixed in 10% neutral formalin, decalcified in 5% nitric acid, and followed by routine histological procedures for the microsections in 4 \( \mu \)m thickness. The microsections were stained with hematoxylin and eosin, Masson trichrome, and von Gieson methods, and observed under light and polarizing microscope. The usage of the biopsy specimens filed in the Department of Oral Pathology was approved by Life Ethic Committee (GWNUDH-IRB2009-16-3). All of the eight cases showed the clinical and radiological features of COC, and their microsections were also reviewed and agreed by two pathologists.

### Table 1. The calcifying odontogenic cysts observed in this study

<table>
<thead>
<tr>
<th>cases</th>
<th>gender</th>
<th>age</th>
<th>location</th>
<th>pathological findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>S1999-115</td>
<td>M</td>
<td>21</td>
<td>upper incisors</td>
<td>arisen from tooth follicle tissue a few calcification and many keratinizing ghost cells</td>
</tr>
<tr>
<td>S2000-30</td>
<td>F</td>
<td>13</td>
<td>left maxillary molars</td>
<td>arisen from ameloblastic fibro-odontoma diffuse calcification and many keratinizing ghost cells</td>
</tr>
<tr>
<td>S2001-87</td>
<td>F</td>
<td>25</td>
<td>left maxillary molars</td>
<td>with diffuse calcification and a few keratinizing ghost cells</td>
</tr>
<tr>
<td>S2001-109</td>
<td>M</td>
<td>22</td>
<td>upper incisors</td>
<td>with diffuse calcification and many keratinizing ghost cells</td>
</tr>
<tr>
<td>S2002-65</td>
<td>F</td>
<td>37</td>
<td>upper incisors</td>
<td>with diffuse calcification and many keratinizing ghost cells</td>
</tr>
<tr>
<td>S2006-200</td>
<td>M</td>
<td>12</td>
<td>upper incisors</td>
<td>arisen from hamartoma-like odontoma diffuse calcification and a few keratinizing ghost cells</td>
</tr>
<tr>
<td>S2009-31</td>
<td>F</td>
<td>23</td>
<td>left maxillary incisors</td>
<td>with ossification by basophilic ghost cells (present case)</td>
</tr>
<tr>
<td>S2009-58</td>
<td>M</td>
<td>12</td>
<td>lower incisors</td>
<td>with a few calcification and many keratinizing ghost cells</td>
</tr>
</tbody>
</table>

Abbreviation: COC: calcifying odontogenic cyst
III. Results

Eight cases of COC occurred in four females and four males, and their mean age was 20.6 years old. Among them seven cases were found in maxilla and only one case in mandible, and more six cases were located at incisor area while two cases at molar area. However, a case from 23 years old female showed aberrant ossification admixed with many basophilic ghost cells contrast to the other cases. This case was found during her routine dental check, disclosing a reverse pear shape cystic radiolucency in left maxillary canine area of pantomogram (Fig. 1). The lesion was asymptomatic and showed a diffuse radiopacity in the luminal space, thereby it was clinically diagnosed as a COC. Histologically, the cystic lesion showed many lumps of ossified tissue, in which a lot of basophilic ghost cells were diffusely distributed (Fig. 2A). Compared to the other seven cases of COC, this case showed no cementum-like granular dystrophic calcification. In the polarizing microscope observation, Masson trichrome stain showed the typical birefringence of bony architecture in the ossified materials of COC (Fig. 2B), which was strongly stained red by von Gieson method (Fig. 2C). The ossification was directly associated with basophilic ghost cells, which were much enlarged and devoid of nuclei. The histological feature of the ghost cells in the present COC was different from the eosinophilic keratinizing ghost cells found in the other seven cases compared in this study.

The ghost cells showed the complete degradation of nuclear materials and cytoplasmic organelles, retaining only the reticular matrix of cell membrane and cytoplasmic skeletal networks which were stained basophilic by hematoxylin (Fig. 2 A1, A2, A3). Some ghost cells still disclosed the features of interdigitating epithelial attachments, and also the degradation of nuclear chromatin or nucleoli was found in their empty nuclear spaces (Fig. 2 A4, A5, A6). However, the ordinary odontogenic cyst epithelium was not found in the luminal surface of the present COC, and the cyst wall was relatively thin and gradually replaced by ossifying cellular fibrous tissue. And the degenerating ghost cells did not show any feature of neoplastic growth.

Fig. 1. Pantomogram showed a reverse pear shaped cyst in left maxillary canine area. The cyst lumen contained slight homogenous radiopacity.
IV. Discussion

The calcifying odontogenic cyst (COC) is a rare lesion without specific clinical characteristics. Its diagnosis essentially depends on the histopathological presence of calcification and ghost cells associated in the cyst wall\(^ {2,11}\). The ghost cells and cornifying cells have an altered distribution of their cytokeratins demonstrated by the absence of antibody stainings against cytokeratins\(^ {12}\), therefore, the coexistence of cornifying and ghost cells may imply the great potential of odontogenic epithelium to form different epithelial lesions\(^ {7,13-15}\). It is also known that the amorphous calcification in COC is closely related to the odontogenic epithelium, which is able to produce odontogenic tumors, i.e., odontoma and ameloblastic fibro-odontoma. The present case of COC showed aberrant ossification rather than the dystrophic calcification as usual. The eosinophilic osteoid materials were continuously deposited in the vicinity of the basophilic ghost cells, and eventually the ghost cells were replaced by the bony tissue. Its bony architecture was confirmed by the typical palisading birefringence in polarizing microscope observation. However, the present study clearly demonstrated the mesenchymal ossification originated from ghost cells of COC similar to pilomatricoma.

The basophilic ghost cells of the present COC showed the loss of entire nuclear materials and cytoplasmic organelles, only the reticular structure of cell membrane and cytoplasmic skeletal networks were remained. Therefore, the basophilic ghost cells of the present COC were basically different from the eosinophilic keratinizing ghost cells found in the other seven cases of COC examined in this study. However, it was presumed that the pathological features of the basophilic ghost cells were similar to the ghost cells of pilomatricoma\(^ {4,16-18}\), a benign tumor arisen from the skin appendage tissue. These findings may indicate the variable cytodifferentiation of COC epithelium, including the presence of melanin and melanocytes in the COC\(^ {19}\).

The present COC showed aberrant ossification closely associated with basophilic ghost cells. The basophilic ghost cells were not proliferative rather gradually degenerative and replaced by the ossification similar to the findings of pilomaticoma.

V. References


