Abstract: Odontogenic fibroma (OF) is a relatively rare benign tumor derived from odontogenic ectodermal mesenchymal tissue. It is divided into central (COF) or peripheral OF (POF) based on the affected area. Regarding its pathological features, OF can also be classified as epithelium-rich (WHO type) or epithelium-poor (simple type), depending on the amount of odontogenic epithelium in the tumor. There is limited information available about the latter type because of its low incidence. We report case of simple type COF apparently like POF. A 52-year-old Japanese male was suffering from tenderness at the right posterior maxilla during occlusion with his removable partial denture. The lesion was diagnosed as a simple type OF arising at the edentulous region around the right molar site of the maxilla. A tumor resection was performed, and there was no evidence of recurrence at his 18-month follow-up examination. In addition, we provide a review of the literature with the most up-to-date information about this lesion so that it can be diagnosed correctly.

Keywords: Peripheral odontogenic fibroma, Central odontogenic fibroma, Epithelium-poor type, Maxilla
second molar had been extracted by his family dentist eight months prior to his first visit because of severe periodontitis. The right first molar at the maxilla had also already been extracted, but the date was unclear. Subsequently, he had worn a newly fabricated partial denture for the missing teeth. Since he had complained of pain at the posterior region of right side of the maxilla during eating since the initial setting of the new denture, his dentist had modified it frequently. However, his symptoms had not been improved. A mass was observed at the region coincident with the painful area in the maxilla seven months after tooth extraction, so his doctor suggested that he visit a specialist. He did not have any remarkable past medical or family histories. His facial appearance was symmetrical, and the lymph nodes were not swollen. A well-circumscribed, elastic-hard, pedunculated mass was observed around the right posterior region of the maxilla. The size of the mass was 24 x 20 x 6 mm (buccolingual x mesiodistal x height) and the surface was smooth and showed redness with partial ulceration (Fig. 1). The routine examinations such as X-ray, CT and MRI examinations were performed before the treatment.

Results
The alveolar bone at the lesion, where the upper right second molar was extracted eight months prior to initial visit, was defective, and the bone crest, which was more posterior than the bone defect, was rough, instead of demonstrating a smooth line as normally seen on orthopantomography (OPG) (Fig. 2). In the computed tomography (CT) findings (Fig. 3), the buccal cortical bone was noted to be destroyed in the upper right molar region in the transverse section. The bottom of the right maxillary sinus floor was elevated in the affected region (arrows). The results of orthopantomography at the first visit. A rough line was observed at the alveolar crest around the right maxillary molar region (arrows).

Table 1. A List of the Previous Retrospective Studies of Odontogenic Tumors and Fibromas

<table>
<thead>
<tr>
<th>Reference</th>
<th>Country</th>
<th>OT</th>
<th>OF</th>
<th>COF</th>
<th>POF</th>
</tr>
</thead>
<tbody>
<tr>
<td>Siriwardena(7)</td>
<td>Sri Lanka</td>
<td>1,677</td>
<td>7</td>
<td>(0.4%)</td>
<td></td>
</tr>
<tr>
<td>Jing(8)</td>
<td>China</td>
<td>1,642</td>
<td>5</td>
<td>(0.3%)</td>
<td></td>
</tr>
<tr>
<td>Adebaya(9)</td>
<td>Nigeria</td>
<td>318</td>
<td>7</td>
<td>(1.2%)</td>
<td></td>
</tr>
<tr>
<td>Mosqueda-Taylor(10)</td>
<td>Mexico</td>
<td>349</td>
<td>16</td>
<td>(4.6%)</td>
<td>5</td>
</tr>
<tr>
<td>Ladeinde(11)</td>
<td>Nigeria</td>
<td>319</td>
<td>17</td>
<td>(5.3%)</td>
<td>10</td>
</tr>
<tr>
<td>Delay(12)</td>
<td>Canada</td>
<td>445</td>
<td>17</td>
<td>(5.6%)</td>
<td>25</td>
</tr>
<tr>
<td>Aladdini(13)</td>
<td>Iran</td>
<td>380</td>
<td>17</td>
<td>(5.4%)</td>
<td>18</td>
</tr>
<tr>
<td>Buchner(14)</td>
<td>United States</td>
<td>1,088(COT)</td>
<td>*16</td>
<td>(1.5%)</td>
<td>*23</td>
</tr>
</tbody>
</table>

COF; central odontogenic fibroma, COT; central odontogenic tumor, OF; odontogenic fibroma, OT; odontogenic tumor, POF; peripheral odontogenic fibroma, *: The incidence of COT

Figure 1. Intraoral findings at the first visit. A pedunculated mass was observed around the molar to the region of the maxillary tuberosity on the right side of the maxilla (arrows). The mass was red and included an ulcerated region.

Figure 2. The results of orthopantomography at the first visit. A rough line was observed at the alveolar crest around the right maxillary molar region (arrows).

Figure 3. The postoperative CT findings. A: An axial section. The buccal cortical bone was ruptured at the right maxillary molar region (arrows). B: A coronal section. The extraction socket was not full of regenerated bone (arrows). C: A sagittal section. The maxillary sinus floor was elevated in the affected region (arrows).
Seigo Ohba et al.: Ohba Epithelium-poor Odontogenic Fibroma

the lesion. The tooth extraction socket was filled with soft tissue, not with regenerated bone. Magnetic resonance imaging (MRI) (Fig. 4) detected a well-defined lesion which showed a low signal that was equivalent to muscle in T1-weighted images (Fig. 4A), and the immediate signal in T2-weighted images (Fig. 4B) in the right posterior region of the maxilla. The internal signal of the lesion was uneven. Moreover, the inside of the lesion was inhomogeneously enhanced using contrast media (Fig. 4C), which was the same as the finding in T2-weighted images. These findings suggested that the lesion was a fibrous tumor.

The mass mainly consisted of fibrous connective tissue which contained high- and low-cellular areas in the histopathological findings (Figs. 5A, B, C). There were elliptical and spindle-shaped cells in the lesion. The fibrous connective tissue included some basophilic matrix and was similar to dental sac. Although the histopathological findings were similar to those of the odontogenic ectomesenchymal tissue, odontogenic epithelium was not observed in the lesion. In the central region, a portion of low cellular areas with less fibrous connective tissue and with myxoid component was also included (Fig. 5B, C). Based on these histological findings, the lesion was diagnosed as an odontogenic fibroma. However, it was initially unclear whether the lesion was a POF or a COF that first grew extra-osseously, passing through the extraction socket. Based on the following findings, this lesion was considered to be a COF which originally existed around the apex of the right second molar at the maxilla, and grew out extra-osseously by passing through the extraction socket after tooth extraction, resulting in its POF-like appearance.

1) There was no bone regeneration in the extraction socket. 2) The present tumor apparently liked peripheral tumor. Although POF is a very rare disease, it has been reported that POF, along with peripheral ameloblastoma, is a common type of peripheral odontogenic tumor19, 23, 24). POF commonly occurs in the posterior region of the mandible and anterior region of the maxilla15, 25-27), and usually occurs around the periodontal tissue, including teeth. It is seldom observed in an edentulous region. Lin et al.3) reported that the incidence of POF in edentulous regions was 4% (1/25 cases) and Ritwik et al.25) calculated that it was 1.3% (2/151 cases). Therefore, a POF in an edentulous posterior region of the maxilla, as was the lesion in the present case, is sometimes hard to diagnose correctly.

The mass existed at the extraction socket of the upper right second molar and it continued from the intra- and extra-osseous parts in this case to form a hemispherical mass on the alveolar ridge. The histopathological diagnosis of the present case was odontogenic fibroma. However, it was initially unclear whether the lesion was a POF or a COF that first grew extra-osseously, passing through the extraction socket. Based on the following findings, this lesion was considered to be a COF which originally existed around the apex of the right second molar at the maxilla, and grew out extra-osseously by passing through the extraction socket after tooth extraction, resulting in its POF-like appearance.

Discussion

The present tumor apparently liked peripheral tumor. Although POF is a very rare disease, it has been reported that POF, along with peripheral ameloblastoma, is a common type of peripheral odontogenic tumor19, 23, 24). POF commonly occurs in the posterior region of the mandible and anterior region of the maxilla15, 25-27), and usually occurs around the periodontal tissue, including teeth. It is seldom observed in an edentulous region. Lin et al.3) reported that the incidence of POF in edentulous regions was 4% (1/25 cases) and Ritwik et al.25) calculated that it was 1.3% (2/151 cases). Therefore, a POF in an edentulous posterior region of the maxilla, as was the lesion in the present case, is sometimes hard to diagnose correctly.

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The maxillary sinus floor was not destroyed, but was elevated. 3) The tumor was continued from the side of the maxillary sinus to the submucosa in the histopathological findings. 4) There were small findings of inflammation, proliferation of fibroblasts and neoangiogenesis, which are usually observed during the process of wound healing after tooth extraction. 5) The extra-osseous region of the mass appeared soon after tooth extraction. Finally, the tumor may have grown from the maxillary bone and passed through the extraction socket. Finally, the tumor may have served as a trigger to enhance the growth of this OF originally occurred in the maxillary jaw bone. The tooth extraction may have served as a trigger to enhance the growth of the tumor, and it may have grown from the maxillary bone and passed through the extraction socket. Finally, the tumor may have become a POF in this case. Since there is limited information published about POF, especially with this unique growth process and of the simple type, a long-term follow-up will be required.

References


35. Brannon BR. Central odontogenic fibroma, myxoma


