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Clinical Report

Epithelium-poor Odontogenic Fibroma with an Unusual Progress

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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

Introduction

Odontogenic fibroma (OF) is a rare benign tumor which is originally derived from odontogenic ectodermal mesenchymal tissue[1]. It is divided into central OF (COF) and peripheral OF (POF) based on the affected region[2]. COF occurs in the jaw bone, and usually shows a well-defined radiolucent area in the X-ray findings[3] and sometimes induces bone swelling[4-6]. On the other hand, POF arises in extra-bone regions.

Several large-scale retrospective studies about OF have been reported in various nations (Table 1). The incidence of OF has been reported to be 0.3-5.3% of all odontogenic tumors[7-11]. COF and POF have been reported to occur 1.4-5.6%[10-12] and 2.1-8.1%[10-13] of all odontogenic tumors, respectively. The incidence of POF may be more than that of COF according to these previous reports. Buchner et al.[14] reported that the incidence rates of COF and POF are 1.5 and 2.1% of 1,088 central odontogenic tumor (COT) cases, respectively. They also estimated similar incidence rate of COF and POF (0.02%) of all oral lesions[14]. On the other hand, Delay et al.[12] reported that COF and POF occurred 0.06% and 0.09% of all oral lesions. According to these data, both of COF and POF is a rare disease therefore the details have been still unknown.

From the findings of histopathological features, OF is classified as an epithelium-rich (WHO) type or an epithelium-poor (simple) type depending on the amount of the odontogenic epithelium[1]. In the previous reports, WHO type is the most frequently detected, while reports describing the simple type OF were found occasionally[4, 6, 10, 15-22], and all of them were intraosseous lesions, i.e. no simple type POF has been reported. We herein report a simple type COF with a unique type of growth apparently like POF.

Materials and Methods

This study received ethical approval from the institution. The aim of this study was to reveal the origin of OF apparently like POF. A 52-year-old Japanese male was referred to the Department of Oral and Maxillofacial Surgery, Nagasaki University Hospital. His chief complaint was a sharp pain of the right posterior region of the maxilla when he wore his upper denture. His upper right

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

Reference	Country	OT	OF	COF	POF
Siriwardena[7]	Sri Lanka	1,677	7 (0.4%)		
Jing[8]	China	1,642	5 (0.3%)		
Adebaya[9]	Nigeria	318	7 (1.2%)		
Mosqueda-Taylar[10]	Mexico	349	16 (4.6%)	5 (1.4%)	11 (3.1%)
Ladeinde[11]	Nigeria	319	17 (5.3%)	10 (3.1%)	7 (2.2%)
Delay[12]	Canada	445		25 (5.6%)	36 (8.1%)
Aladdini[13]	Iran	380			18 (4.7%)
Buchner[14]	United States	1,088 (COT)		*16 (1.5%)	*23 (2.1%)

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

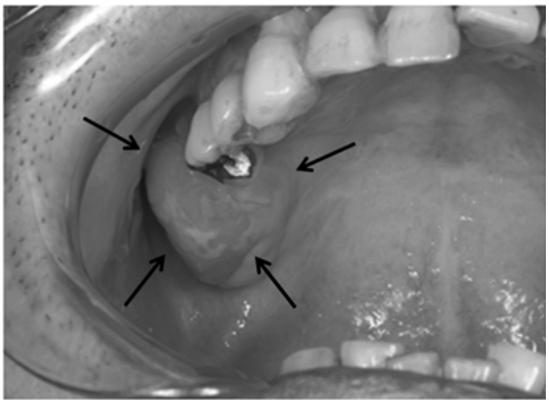

Figure 1. Intraoral findings at the first visit. A pedunculated mass was observed around the molar to the region of the maxillary tuberosity on the right side of the maxilla (arrows). The mass was red and included an ulcerated region.

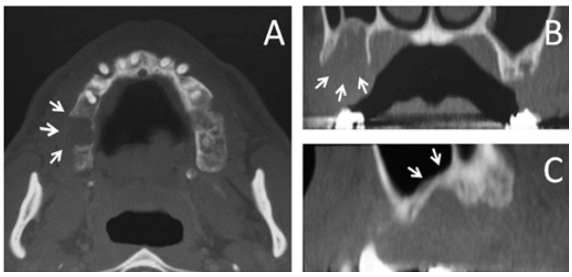

Figure 3. The postoperative CT findings. A: An axial section. The buccal cortical bone was ruptured at the right maxillary molar region (arrows). B: A coronal section. The extraction socket was not full of regenerated bone (arrows). C: A sagittal section. The maxillary sinus floor was elevated in the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

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

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).

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 was elevated in that region, and the cortical bone was rough around the right maxillary tuberosity in the coronal and sagittal sections. There was no finding of bone absorption at a bottom of the right maxillary sinus, and no inflammation was detected around

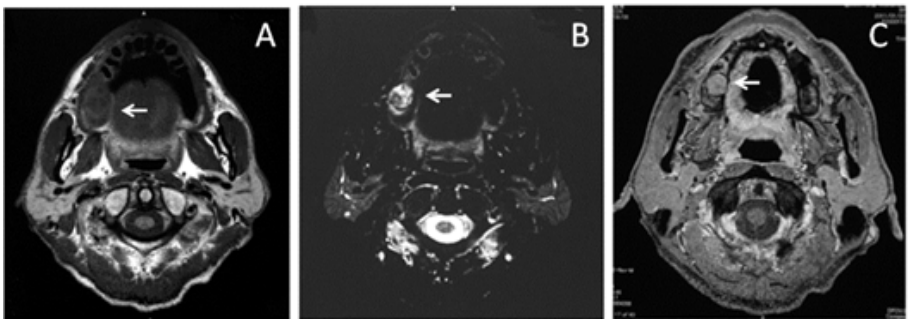

Figure 4. The postoperative MRI findings. A well-defined mass, which showed low signal intensity in T1 images (A) and moderate signal intensity in T2 images (B) was detected. The lesion was enhanced by an imaging agent (C). The internal heterogeneity of the legion was observed in all images.

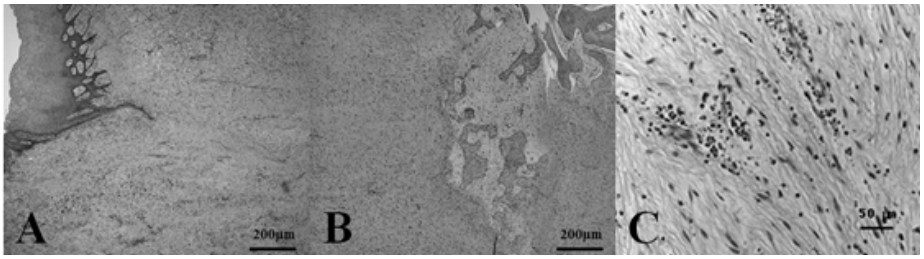

Figure 5. The histopathological findings (H-E staining).

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 (simple type). The tumor resection was performed under general anesthesia with a 5 mm safety margin. There was no adhesion between the tumor and the bone. The surface of exposed bone after resection was smooth, and irregular bone absorption was not found. The excision specimen was solid, with a white division surface. Over the 18-month follow-up period since the surgical treatment was performed, there has been no finding of recurrence, and the patient has not had any symptoms while wearing his dentures.

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 tumor[19, 23, 24]. POF commonly occurs in the posterior region of the mandible and anterior region of the maxilla[15, 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. 1) There was no bone regeneration in the extraction socket. 2)

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. Gardner[27] advocated that cases of OF, which originated as COF and grew out extra-ossoeously, are POF. Based on his suggestion, the lesion in our present case was finally diagnosed to be a POF arising in the right posterior edentulous region of the maxilla.

The histopathological findings of this lesion showed similar features to that of odontogenic ectodermal mesenchymal tissue, but did not include the odontogenic epithelium. In this respect, it was considered to be a simple type POF[25-27, 28-31]. There are some case reports which described simple type COF[4, 6, 10, 15-21, 32]. On the other hand, all of the POF reports that described the histopathological diagnosis were of the WHO type[3, 12, 13, 23, 26, 27, 30, 33-38], and this is the first report referring to a simple type POF in the English literature. The histopathological findings of OF showed the proliferation of elliptical and spindle-shaped cells, and fibrous connective tissue containing a small amount of odontogenic ectodermal mesenchymal tissue like a mucoid matrix (Fig. 5).

In addition to these characteristic findings, it is easy to diagnose a lesion as OF if odontogenic epithelium is observed in the lesion. However, in cases where the OF has little or no odontogenic epithelium, like the present case, it is important to distinguish the condition from odontogenic myxoma (OM) which also contains limited or no odontogenic epithelium[39]. There was a myxomatous portion in the central region of the tumor, but uniformly-spread abundant myxoid component, which is the typical feature of OM[39], was not observed in the present resected specimen, and OM could be ruled out. An intra-osseous fibrogenic tumor, desmoplastic fibroma (DF) was also one of the candidates of this tumor. In the present specimen, dense hyalinized collagen fibers, which are abundantly seen in DF, were rarely observed in this case, and DF was ruled out. In addition, non-ossifying fibroma, which is characterized by storiform arrangement of spindle cells with multinucleated osteoclast-like cells, was also ruled out because of no similar findings in this case. In the present case, it was also necessary to distinguish this lesion from neurogenic tumors (NTs) based on the cellular morphology, because OF and NT have similarly shaped cells. An additional immunohistological study showed that S-100 staining was negative in this lesion. This suggested that this disease was not a NT. According to the above histological findings, the present patient was finally diagnosed to have a simple type POF.

Based on the initial diagnosis, the tumor resection was performed for COF, and the prognosis has been relatively good. However, the recurrence rates of POF after surgery have been reported to be 0-50%[3, 13, 14, 24-27]. Lin et al.[3] alleged that the reason for this wide range of recurrence rate might be caused by the differences in ethnicity, the size of the tumors and the small number of cases examined. Since there have been too few reports which described the prognosis of POF to provide a consensus about the prognosis of POF, a further epidemiological examination to acquire more information about POF is needed.

Ritwik et al.[24] reported that the basal cell layer budding of the surface squamous epithelium was observed in 27 of 29 recurrent cases (93.1%). They therefore concluded that the basal cell layer budding of the surface squamous epithelium was one of the risk factors for recurrence after resection of POF. They also reported that various amounts of calcific substances were observed in 16 cases of the 29 recurrent cases (55.2%). This finding may also be associated with the recurrence of the disease, and additional examinations are necessary to fully elucidate the risk factors for recurrence. In the present case, there was no finding of basal cell layer budding of the surface squamous epithelium, and no calcific substance was found. However, the lesion was considered to be overgrown from COF, and was finally diagnosed to be a POF, so there is a possibility that the risk of recurrence may be high. Therefore, it is important to observe the patient long-term in order to detect any recurrences as early as possible. It is believed that 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.

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