J Oral Pathol Med (2005) 34: 190–2

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CASE REPORT

Benign fibrous histiocytoma of the mandible

Mitsunobu Kishino[1,2], Shumei Murakami[3], Satoru Toyosawa[1], Atsutoshi Nakatani[3], Yuzo Ogawa[1], Takeshi Ishida[2], Naokuni Ijuhin[1]

[1]*Department of Oral Pathology, Osaka University Graduate School of Dentistry, Osaka, Japan;* [2]*Clinical Laboratory, Osaka University Dental Hospital, Osaka, Japan;* [3]*Department of Oral and Maxillofacial Radiology, Osaka University Graduate School of Dentistry, Osaka, Japan*

A very rare case of benign fibrous histiocytoma of the mandible is presented. A 49-year-old woman was admitted because of left buccal swelling and pain. Panoramic radiograph showed well-demarcated soap-bubble appearance without sclerotic rim in the left mandibular bone. A yellowish-white and partly brown solid tumor was noted in the excised mandibular bone specimen. The tumor histologically consisted of spindle cells, in which areas showing a storiform pattern and other areas composed of histiocytic cells with erythrophagocytosis and foam cells were mixed. Immunohistochemically, the tumor cells were positive for vimentin, CD68, α-1-antichymotrypsin and α-1-antitrypsin. From these findings the tumor was diagnosed as a primary BFH of the mandible. No recurrence has been noted 2 years and 11 months after surgery.
J Oral Pathol Med (2005) **34**: 190–2

Keywords: benign fibrous histiocytoma; bone; mandible

A 49-year-old woman was admitted to our hospital because of left buccal swelling and spontaneous pain. Panoramic radiograph showed well-demarcated soap-bubble appearance with an irregular margin, between the molar region and condyle in the left mandible (Fig. 1). Bone target CT showed a slight bucco-lingual bulge with the thin cortical bone (Fig. 2). She underwent incisional biopsy of the mass, which revealed benign fibrohistiocytic tumor. The jawbone excision ranging from the left mandibular molar region to the condyle was performed. Although the excised mandibular bone showed a slight bucco-lingual bulge between the left mandibular second molar and the angular region, no exposure of the tumor out of the cortical bone was found. The cut surface was yellowish-white solid mass, showing hemorrhage in some areas. The mandibular cortical bone became thin overall (Fig. 3).

Tissues obtained by surgery were fixed in 10% buffered formalin and then decalcified in 10% formic acid solution for 1 week. Histological sections from paraffin embedded blocks were stained with hematoxylin and eosin (H&E). Microscopic examination showed

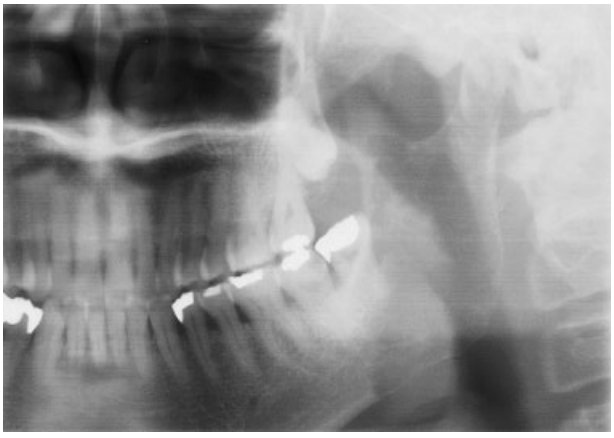

Figure 1 Panoramic radiograph. Well-demarcated soap-bubble appearance is noted between the left molar region and the condyle.

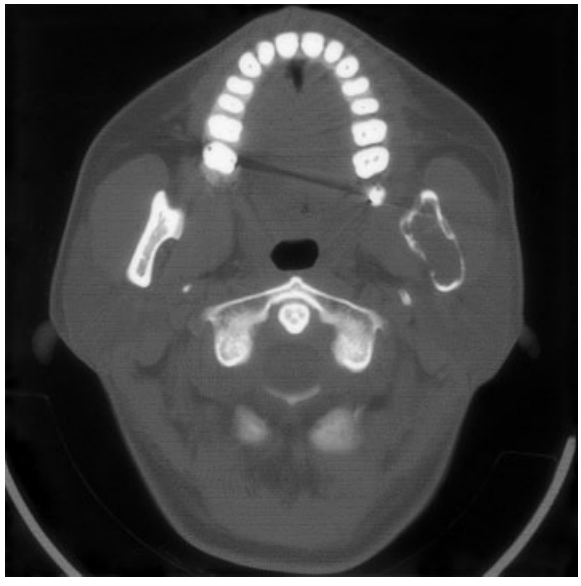

Figure 2 Bone target CT. The left mandible shows a slight bucco-lingual bulge and thin cortical bone.

Correspondence: Mitsunobu Kishino, Department of Oral Pathology, Osaka University Graduate School of Dentistry, 1-8 Yamadaoka, Suita, Osaka 565-0871, Japan. Tel.: +81 6 6879 2892. Fax: +81 6 6879 2895. E-mail: mkishino@dent.osaka-u.ac.jp
Accepted for publication August 12, 2004

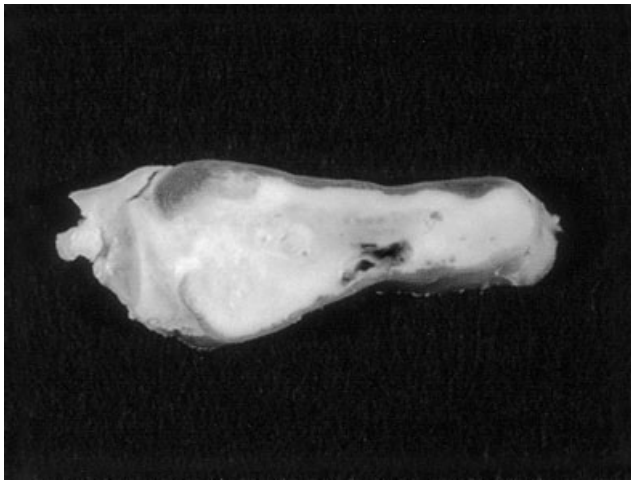

Figure 3 Cut surface of the central area of the tumor (after fixation). The tumor consists of yellowish-white solid soft tissue. Although the cortical bone is thin overall, it is not destroyed.

that spindle-shaped tumor cells were arranged in a storiform pattern (Fig. 4a). Furthermore, the proliferation of histiocytic cells was observed, and foam cells and erythrophagocytic cells were detected (Fig. 4b). Multinucleated giant cells were not detected. The tumor cells showed few mitotic figures and slight pleomorphism with no nuclear atypia. Reactive new bone formation was not present at the periphery of the tumor.

Immunohistochemical stain was performed on the formalin-fixed paraffin-embedded sections of the biopsy specimen by the labeling streptavidin–biotin-peroxidase complex (LSAB) method. The tumor cells were all positive for vimentin (V9, 1:200, Dako, Glostrup, Denmark), and most of them were positive for α-1-antitrypsin (polyclonal, 1:2000, Dako) (Fig. 5a) and α-1-antichymotrypsin (polyclonal, 1:2000, Dako) (Fig. 5b), and partially positive for CD68 (KP1, 1:100, Dako) (Fig. 5c). However, the tumor cells were negative for cytokeratin (AE1/AE3, 1:100, Novocastra, Newcastle,

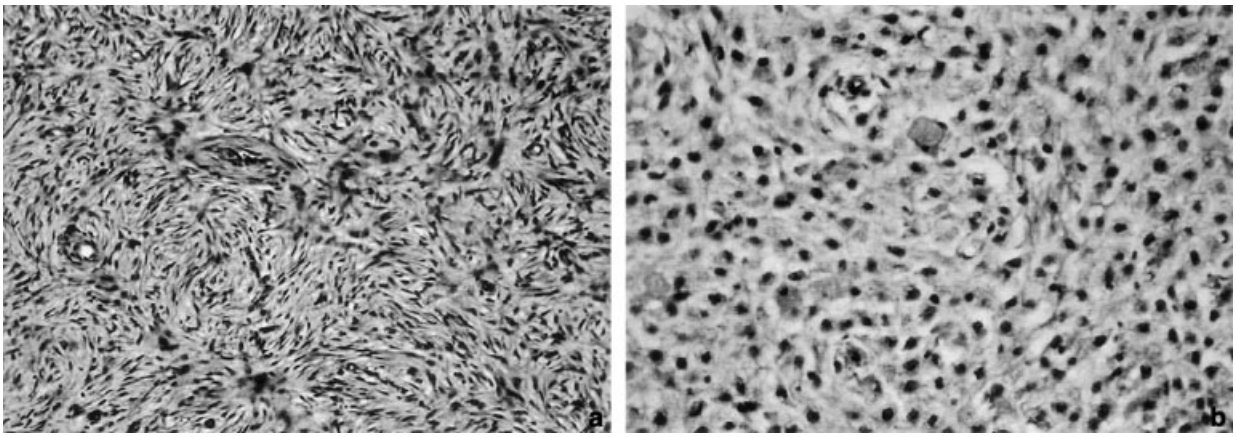

Figure 4 Histological findings of the tumor. (a) The tumor is composed of proliferating spindle cells showing a storiform pattern arrangement (H&E; magnification: ×100). (b) Areas showing the proliferation of histiocytic cells including foam cells and erythrophagocytic cells (H&E; magnification: ×200).

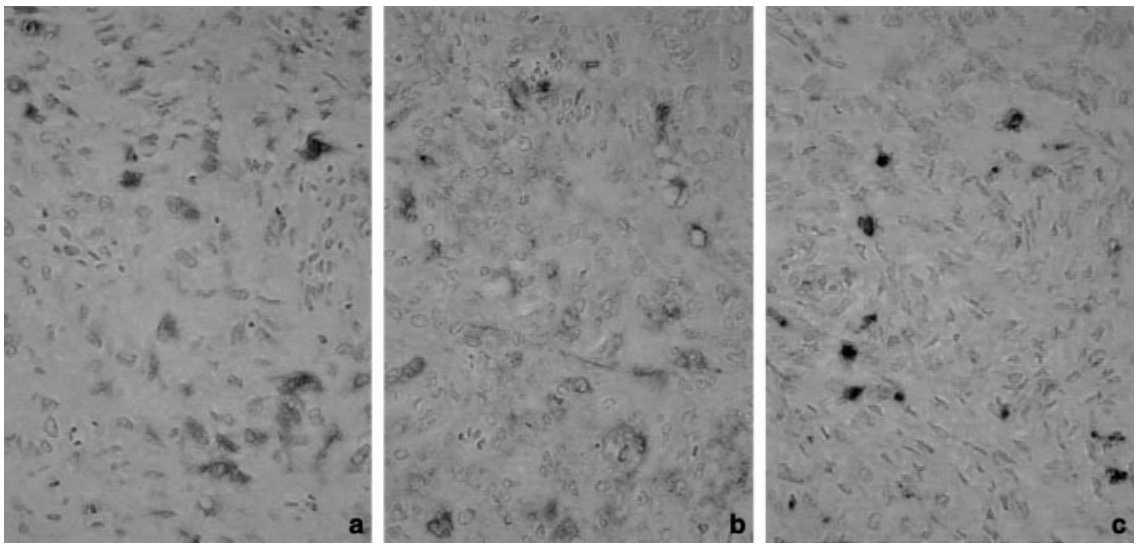

Figure 5 Immunohistochemical findings of the tumor (magnification: ×200). (a) Most of the tumor cells are positive for α-1-antitrypsin and (b) α-1-antichymotrypsin. (c) Partially positive for CD68.

UK), epithelial membrane antigen (E29, 1:100, Dako), α-smooth muscle actin (1A4, 1:200, Dako), desmin (D33, 1:100, Dako), S-100 protein (polyclonal, prediluted, Dako), or CD34 (QB-END/10, 1:50, Novocastra). These findings indicated a histiocytic origin of the tumor.

Comments

Primary benign fibrous histiocytoma (BFH) in the bone is rare with less than 100 reported cases according to *World Health Organization Classification of Tumours* (1). Among those cases, frequent occurrence sites were the pelvic bone, femur and tibia. Histological characteristics of BFH in bones are similar to those of BFH occurring in soft tissues. The tumor consists of proliferating fibrohistiocytic cells showing a storiform pattern arrangement, and various numbers of foam cells and giant cells (1, 2). Histological findings of BFH are indistinguishable from those of non-ossifying fibroma (NOF), and, therefore, both should be differentiated based on the clinical radiographic setting (1, 2). Clinically, BFH in bone frequently occurs in adults and causes pain, without bone fractures (3). NOF is centered in the metaphysis of the long bones and radiologically show a lytic lesion with a sharply circumscribed sclerotic rim, whereas BFH occurs in non-long bones, or lack of metaphyseal involvement even if in a long bone (2). Furthermore, radiographic sclerotic rim was generally seen in NOF, but not in BFH. Our case in this study was clinically a painful lesion occurring in the adult non-long bone (mandible) without bone fracture. Radiologically, the lesion showed well-demarcated soap-bubble appearance without sclerotic rim. Histological findings, which revealed the proliferation of fibrohistiocytic cells with erythrophagocytosis and foamy appearance, were consistent with those of BFH. These findings indicated that this tumor was diagnosed as a primary BFH of the mandibular bone.

BFH occurring in the jaw bone is very rare, and there have been only one English report of a maxillary case (4), and three mandibular cases (5–7). The three mandibular cases involved large lesions occurring in the posterior mandible. Remagen et al. (5) described the enormous 'blow-out' lesion of the left half of the mandible, and White and Makar (6) reported a large bucco-lingual bulging lesion ranging from the molar region to the ramus. Recently, Heo et al. (7) appeared as a well-defined multilocular radiolucency involving from the left mandibular body to the condylar head that caused buccal and lingual expansion with thinning of the cortex. Histologically, these tumors were composed of spindle-shaped cells showing a whorled appearance and histiocytes with formy cytoplasm. These clinical and histological findings of the three mandibular cases were similar to the findings of our case in this study. Harsanyi and Larsson (8) described seven 'xanthomatous lesion of the mandible'. We do not consider that their cases represent BFHs from the clinical, histological and radiographic features. Ertas and Buyukkurt (9) reported a case of BFH involving the mandible. It is difficult to evaluate this case adequately because of unconvincing photomicrograph.

As BFH does not show aggressive behavior without histological findings such as nuclear atypia, pleomorphism, or atypical mitosis, it can be diagnosed as benign (2). Although fibrous capsules of this tumor was not identified in the histological sections, histological examination revealed the clear boundary between this lesion and surrounding tissues, and no malignant findings such as abnormal mitosis or cellular atypia. These findings indicated that it was benign. Furthermore, differential diagnosis from giant cell tumor and giant cell reparative granuloma was easy, as multinucleated giant cells were not detected in this case.

No recurrence has been noted 2 years and 11 months after jaw bone excision. Three of the eight cases in the series reported by Clarke et al. (2) recurred locally. As curettage and bone grafting were performed in the three recurrent cases, it is considered that recurrence can be prevented by completely excising the tumor.

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