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(Citation)

The Kobe journal of the medical sciences, 54(3):169-173

(Issue Date)

2008-06

(Resource Type)

departmental bulletin paper

(Version)

Version of Record

(JaLCOI)

<https://doi.org/10.24546/81000774>

(URL)

<https://hdl.handle.net/20.500.14094/81000774>



Myofibroma of the Mandible

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Received 25 April 2008/ Accepted 26 May 2008

Key words: Myofibroma, Myofibromatosis, Mandible, FDG-PET

This article is reporting a case of myofibroma involving the mandible of a 12-year-old boy. The patient did not have any swelling nor any redness but of partial mental nerve hypoesthesia. A panoramic radiograph demonstrated a globe like radiolucent lesion in the left angle of the mandible that was an increased uptake spot by FDG-PET. Incisional biopsies indicated myofibroma, therefore the tumor was totally resected under general anesthesia. After surgery there was no complication except for a change for the worse of unilateral mental nerve hypoesthesia. At 29 months postoperative, no evidence of recurrence could be found.

INTRODUCTION

Myofibroma and myofibromatosis are a benign fibroblastic and myofibroblastic tumor in the soft tissue, bone, or internal organs affecting all ages. Although Williams and Schrum first named this lesion congenital fibrosarcoma in 1951 (1), Stout featured its multifocal growth as a benign tumor and amended the term to congenital generalized fibromatosis in 1954 (2). This lesion was also reported as multiple vascular leiomyomas of newborn (3), or multiple mesenchymal hamartomas (4), while Chung and Enzinger used the term of infantile myofibromatosis in 1981 because of its affecting infants and young children (5). On the other hand, adult counterpart of this lesion was reported by Daimaru et al in 1989, renamed as “myofibromatosis” (6). In the same year, solitary case of this tumor was delineated by Smith et al and was termed “myofibroma” (7). Afterwards, the terms of “myofibromatosis” and “myofibroma” were adopted by the World Health Organization (8). Myofibroma involves predominantly the head and neck region (36%) or the trunk, however cases of the jaws are rare. The purpose of this article is to report a case of myofibroma involving the mandible, and to describe its clinical, histologic and immunohistochemical features.

CASE REPORT

A 12-year-old boy was referred to our hospital because of partial mental nerve hypoesthesia after his facial injury. Physical examination revealed neither any swelling nor any redness in the intra and extra oral regions, and no trismus (Fig.1).



Fig. 1. A photograph at the first visit. There is neither any swelling nor any redness.



Fig. 2. A panoramic radiograph revealing a globe like radiolucent lesion in the left angle of the mandible (arrows).

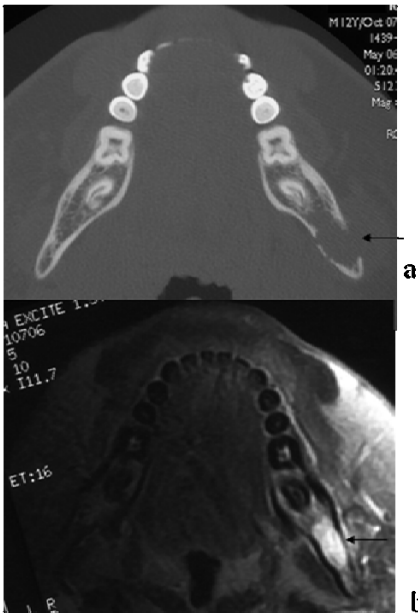


Fig.3. a; CT scan. A unilocular low-density area with partial resorption of the cortical bone is detected (arrow).
b; MRI on T2-weighted spin-echo. The lesion is shown as high signal intensity (arrow).

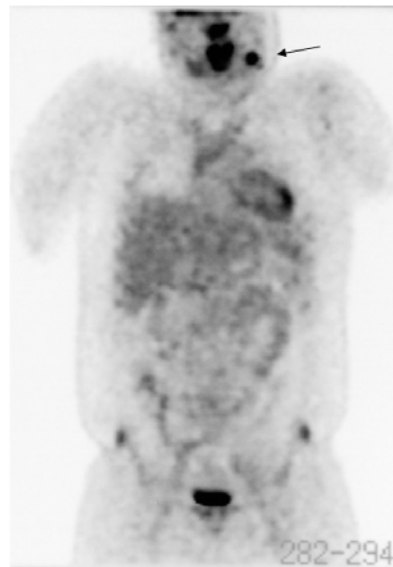


Fig.4. The lesion is revealed as an increased uptake spot by a whole-body scan of fluorodeoxyglucose positron emission tomography (FDG-PET) (arrow).

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Although a panoramic radiograph did not demonstrate any bone fracture, a globe like radiolucent lesion was detected in the left angle of the mandible (Fig.2). Computed tomography (CT) showed the lesion as a unilocular low-density area with partial resorption of the cortical bone (Fig.3a). This lesion was low signal intensity on T1-weighted spin-echo and high signal intensity on T2-weighted spin-echo by Magnetic resonance imaging (MRI) (Fig.3b), and was shown as an increased uptake spot by a whole-body scan of fluorodeoxyglucose positron emission tomography (FDG-PET) (Fig.4).

An incisional biopsy was performed that indicated fibrous dysplasia. However, 3 months later the radiolucent area extended rapidly like sarcoma, and a secondary incisional biopsy was re-performed which revealed definitely to be myofibroma. Surgical resection was thus performed under general anesthesia. The tumor was yellowish and segmented form without capsule. Although the inferior alveolar nerve was involved by the tumor, the tumor was easily removed completely from the mandible. There was no postoperative complication except for unilateral mental nerve hypoesthesia. At 29 months postoperative, no evidence of recurrence could be found.

Histologically, the tumor was composed of spindle-shaped cells with little atypia and few mitosis which formed whirling or multinodular pattern with surrounding fascicular alignment in a myxoid background (Fig.5a). In some areas a hemangiopericytoma-like cellular pattern with abundant vascular components was observed (Fig.5b). Immunohistochemically, spindle-shaped cells showed positive reaction for vimentin and smooth muscle actin, whereas negative for desmin, S-100 protein and CD34.

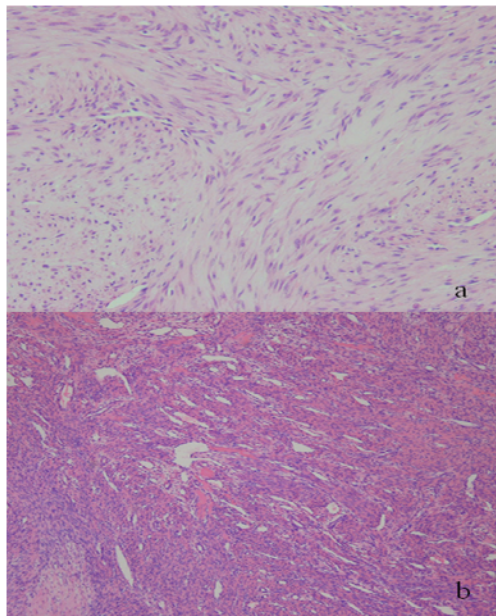


Fig. 5. a; Interlacing fascicular arrangement of myofibroblast closely resembling smooth muscle tumor. (HE stain; original magnification x100.)
b; Prominent vascular pattern mimicking the appearance of hemangiopericytoma. (HE stain; original magnification x40.)

DISCUSSION

Myofibroma has been reported to occur at any age from birth to 84 years of age (9), and most often in the dermis and subcutis, with the head and neck accounting for a majority of the lesions, including the scalp, forehead, orbit, parotid and oral cavity (10). Other locations include the trunk, extremities, skeleton and viscera. Some reported cases have suggested the possibility of a familial pattern of inheritance (9), while the exact etiology has been yet unknown, and the case presented here showed no familial history. In cases of the oral myofibroma, mandible has a majority as the location. Practically, in a report of 79 cases of the oral myofibroma, 30 cases were affecting mandibulae of which 12 cases were located centrally with only partial or poorly developed sclerotic borders around a radiolucent area (9). These intrabony cases were painless, that prevented the patients from noticing until X-ray examination or distinct bone expansion (11,12). Examinations with pathological and immunohistochemical methods are indispensable for diagnosis, however 2 of 79 reviewed cases were reported to have accelerated growth after incisional biopsy like our case (9).

Recently, a whole-body scan of FDG-PET has been used for a diagnosis in patients with cancer. An important role of FDG as a tumor-seeking agent has been established for various malignant tumors, including colorectal, lung, and head and neck cancers. FDG-PET is a functional imaging modality of tumor tissue representing the increased metabolic rate and glucose consumption characterizing malignant cells (13). On the contrary, some cases of FDG uptake in benign leiomyoma and myoma in uterus were reported (14, 15). The reason for the accumulation of FDG in leiomyoma was thought to be the existence of higher levels of growth factors and receptors to be related in proliferation of smooth muscle cells due to the metabolic needs (15). As for the present case, the exact mechanism for the accumulation in myofibroma is not known because the use of FDG-PET imaging to detect a myofibroma has not been studied up to date.

Initial histologic interpretation of this lesion was variously reported as sarcoma, fibromatosis, myofibromatosis, leiomyoma and so forth (9). Foss *et al* emphasized clinical and histological overlap between myofibroma and myofibrosarcoma, whereas it is useful to evaluate desmin expression in discrimination because myofibroma has been uniformly negative in recent studies (9).

Treatment for the typical solitary lesion is an excisional biopsy or simple excision. Local recurrence has been reported in 7% to 31% of excised cases (16), however those recurrences are thought to be mostly caused by multicentric tumor feature or insufficient excision. Chemotherapy or radiation has been seldom used except for a few cases with recurrence or nonresectable lesion (17).

The prognosis of this tumor is mostly excellent in solitary cases. On the other hand, the cases with multicentric visceral lesions, as myofibromatosis, are likely to show an aggressive and sometimes fatal outcome.

In the case presented here, the intrabony lesion was unexpectedly found by his partial mental nerve hypoesthesia after the facial injury. Fortunately, this early detection made it possible to totally excise without any oral functional sequela.

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