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Case report

Central myofibroma of the maxilla: A case report

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ABSTRACT

Myofibroma is a benign tumor characterized by the proliferation of spindle cells originating from myofibroblasts. This tumor, common in infants, rarely occurs in the oral cavity of adults. Owing to its clinical and pathological characteristics, myofibroma has often been misdiagnosed, leading to inappropriate treatment choices. We report here a rare case of a myofibroma occurring in the maxillary bone of a 57-year-old woman. A rapidly growing mass was observed in the anterior maxilla. Imaging examinations revealed a radiolucent, tumor-like solitary lesion. Nuclear medicine scans revealed hyperintense accumulations at the anterior maxilla. Myofibroma was strongly suggested and no malignant finding was observed on biopsy. Since the lesion was solitary, it was excised completely by surgery. The histopathological findings indicated that the lesion mainly comprised bundles of spindle cells with a central hemangiopericytoma-like vascular pattern. Immunohistochemical staining showed the lesion to be positive for vimentin, α-smooth muscle actin and S-100 and negative for desmin. Thus, a definitive diagnosis of myofibroma was made. No recurrence was observed at the 2-year postoperative follow-up. The present case suggests that pathological examination including immunohistochemical analysis is essential for definitive diagnosis and for avoiding misdiagnosis and the consequent unnecessary therapies.

1. Introduction

Myofibroma is a benign tumor characterized by the proliferation of spindle cells originating from myofibroblasts. This tumor occurs rarely, with an incidence of 2–4 cases/million. It is mostly observed in the skin, bones, lungs, and gastrointestinal tract of infants: 89% cases occur in patients before the age of 2 years, and of these, 54% are present at birth [1]. Myofibroma can occur as solitary or multiple lesions. When it occurs at multiple sites, it is termed as myofibromatosis [2], and myofibromatosis is also known as infantile myofibromatosis because majority of these tumors occur in newborns and infants [3]. Myofibromatosis involving multiple visceral organs has a poor prognosis because of severe cardiorespiratory or gastrointestinal complications, which are often difficult to manage and can lead to a poor general condition. Thus, it is important that adequate examinations be performed to rule out the presence of multiple lesions [4]. Solitary infantile myofibroma often regresses spontaneously, probably due to the tendency of tumor cells to undergo apoptosis (programmed cell death). However, the tumor is less likely to regress in older patients; therefore, previous studies have suggested surgical excision of solitary myofibromas in adults [5]. In addition, although recurrence is uncommon, it can be treated by re-excision [6].

Myofibroma of the oral cavity, with an incidence rate of 2%, is very rare [7]. In the oral cavity, myofibroma has been reported to develop in the mandible, tongue, buccal mucosa, hard palate, oral floor, and gingiva. However, a myofibroma in the maxillary bone has not yet been reported (Table 1) [8–17]; thus, the present report is the first to describe the characteristics of a myofibroma in the maxilla.

Owing to the low incidence, information about myofibroma in the oral cavity is very limited. In this report, we aim to clarify the clinical characteristics of a myofibroma in the oral cavity to aid in the appropriate treatment of future cases.

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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Table 1
Previously reported cases of myofibroma in the oral cavity of adults.

Authors	Clinical presentation			Treatment	Prognosis
	Age (y)	Gender	Lesion site		
Beham et al. [8]	41	F	Tongue	Excision	NED
Ugar et al. [9]	21	M	Mandible	Excision	NED
Vered et al. [10]	41	F	Buccal mucosa	Excision	NED
Brierley et al. [11]	43	F	Mandible	Excision	NED
Lyons et al. [12]	28	F	Mandible	Excision	NED
Sapelli et al. [13]	28	M	Lip	Excision	NED
Andreadis et al. [14]	56	F	Buccal mucosa	Excision	NED
Ramadorai et al. [15]	32	F	Mandible	Excision	NED
Brasileiro et al. [16]	23	M	Tongue	Excision	NED
Oliver et al. [17]	34	F	Mandible	Excision	NED
Present case	57	F	Maxilla	Excision	NED

NED, no evidence of disease.

2. Case report

A 57-year-old woman visited our department in April 2011 with a complaint of a painless swelling in the anterior maxilla. She first became aware of a slight swelling in the maxilla in January 2011. On realizing that the mass was rapidly growing causing deformation of the face, she visited a general hospital. She was referred to our department for a detailed examination. She had hyperlipidemia and no remarkable family history.

On extra oral examination, a diffuse swelling was observed in the anterior maxilla with asymmetry in the wings of the nose (Fig. 1A). A semispherical mass of elastic hardness, measuring 32 mm × 19 mm, covered by a normal mucous membrane was found in the root apex region of the maxillary incisors. The teeth adjacent to the lesion were vital and did not show any unusual mobility, but mesial inclination and extrusion were apparent (Fig. 1B). Panoramic radiographic examination revealed a radiolucent lesion in the maxillary bone with root dehiscence of the incisor, and extrusion and mesial inclination were found in the right upper incisor (Fig. 2A). A radiolucent mass, measuring 20 mm × 10 mm × 15 mm, with well-defined borders and extending from the alveolar part of the anterior maxilla to the floor of the nasal cavity was observed on the computed tomography images (Fig. 2B and C). On magnetic resonance imaging, a mass-like lesion was observed in the anterior maxilla. This lesion was well defined and hypointense on T1-weighted images (Fig. 2D), and well defined, internally heterogeneous, and hyperintense on T2-weighted images (Fig. 2E). Intense accumulation at the anterior maxilla, consistent with the tumor focus, was observed on nuclear medicine scanning. Tumor and bone scintigraphies, however, showed no other abnormal accumulation (Fig. 2F).

On the basis of the clinical examination, the swelling was diagnosed as a maxillary tumor and the patient underwent a biopsy. Biopsy data revealed that the lesion mainly comprised proliferative spindle cell bundles with rich fibrous tissues. No malignant finding was observed. Thus, myofibroma was strongly suggested and fibromatosis was suspected. Whole-body imaging examinations confirmed that the lesion was solitary; hence, the tumor was surgically excised after extraction of the upper incisors under general anesthesia in May 2011. When the buccal/lingual mucous membrane was retracted, the encapsulated tumor mass was directly exposed because the alveolar and maxillary bone was destroyed by the development of the tumor (Fig. 3). Since the incisors were in close contact with the tumor and their roots were absorbed by the tumor-derived pressure, we decided to extract the teeth. The tumor mass was easily removed from the surrounding tissues. It was completely excised along with the incisors as a lump.

The resected tumor was a solid 24 mm × 14 mm × 20 mm encapsulated mass with well-defined borders, elastic hardness, and a yellowish-white cut surface (Fig. 4A). Root absorption of the upper incisors was observed (Fig. 4B).

Histopathological analysis of the resected specimen revealed spindle cells that exhibited biphasic sequences with dense and coarse sections (Fig. 5A). The tumor cells were present in the maxillary bone with no trabecular structure. This finding strongly suggested that the tumor in our case originated from the maxillary bone cells. Some hemangiopericytoma-like vascular patterns with capillary vessel hyperplasia were observed in the center of the tumor (Fig. 5B). No atypical cells were observed. Immunohistochemical analysis revealed that the tumor cells were positive for vimentin, α-smooth muscle actin (α-SMA), and S-100, and negative for desmin (Fig. 6A–D and Table 2). Thus, the tumor was histologically diagnosed as a myofibroma of the maxilla.

At the 2-year postoperative follow-up, no local recurrence or occurrence of myofibroma at other sites was observed.

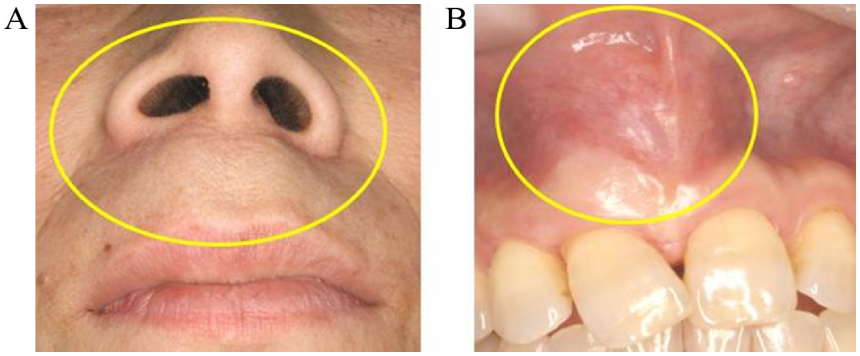

Fig. 1. Photographs at the initial examination. Extraoral photograph shows diffuse swelling in the anterior maxilla and deformation of the right nostril (A). A semispherical mass of elastic hardness covered with a normal mucous membrane was observed in the maxilla, along with mesial inclination and extrusion of the right upper incisor (B).