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Case Report

A case of solitary fibrous tumor of the chin

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ABSTRACT

We report a case of solitary fibrous tumor (SFT) of the chin. A 50-year-old female presented to our hospital in August 2010 with swelling of the chin. Magnetic resonance imaging (MRI) showed a well-demarcated, homogenous lesion of the chin, with low signal intensity on T1-weighted images and moderate signal intensity on T2-weighted images. A benign tumor was suspected based on MRI findings, and subsequently, surgical excision was performed under general anesthesia. Histopathological studies showed that the tumor was composed of interstitial collagenous fibers, microvasculature, and proliferating spindle cells that resemble fibroblasts. The tumor cells were distributed irregularly and inhomogeneously. Immunohistochemical studies revealed that the tumor cells showed positive staining for CD34, vimentin, and bcl-2, but negative for CK AE1/AE3, CD99, SMA, and S100 protein. These findings led to the diagnosis of SFT. No recurrence or metastasis had occurred after a follow-up period of 5 years.

1. Introduction

Klemperer and Rabin reported the occurrence of a solitary fibrous tumor (SFT) in loose connective tissue in the pleura and described it as the first report of SFT [1].

At first, SFT was considered to occur only in the pleura, and benign and malignant SFT were classified as subtype of mesothelioma being mesothelial neoplasm in 1995 WHO classification [2].

However, SFT has been reported to occur in various parts of the body, such as orbit, nasal cavity, and meninges. Therefore, according to the 2002 WHO classification, SFT was stated as occurring in various soft tissues [3].

There are a few cases of SFT occurrence in the oral region, which is particularly rare in the chin. The present report describes a case of SFT of the chin and reviews the characteristics and nature of this neoplasm.

2. Case report

A 50-year-old female was referred to our hospital by her family dentist for swelling of the chin in August 2010. The patient had been suffering from phyma in the same region for five years; however, during the previous month, the patient felt that the phyma have rapidly enlarged, and she consulted her family dentist. The family dentist referred her to our hospital for the detailed examination. The clinical examination showed a slight asymptomatic swelling on the left side of the chin. There was no intraoral swelling in the same region and the anterior teeth were positive of electric vital test (Fig. 1). No significant physical abnormalities such as spontaneous pain, tenderness, or paresthesia, were present on the left side of the chin, and no lymphadenopathy was found. No other relevant medical or family histories were present. A magnetic resonance imaging examination showed a well-demarcated, homogenous lesion on the left side of the chin, with low signal intensity on T1-weighted images and intermediate signal intensity on T2-weighted images (Fig. 2). A benign tumor was suspected based on these findings. Intraoral excision of the tumor was performed under general anesthesia in November 2010. A part of the tumor was adhered to the surrounding tissue; the tumor was excised along with the surrounding healthy tissue. The patient's postoperative course was uneventful. No recurrence or metastasis was observed during the follow-up period of five years.

☆ Asian AOMS: 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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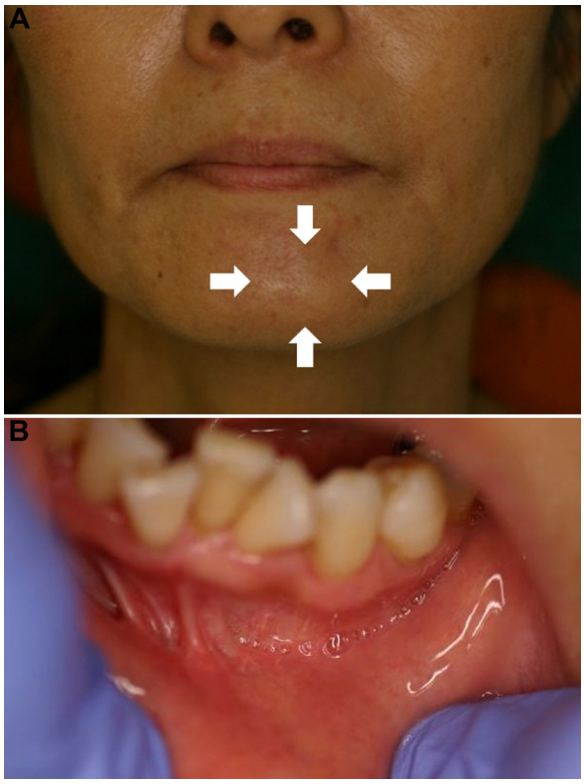

Fig. 1. Extra- and intra-oral view at initial visit. The clinical examination showed a slight asymptomatic swelling on the left side of the chin. (Arrow) There was no intraoral swelling in the same region.

2.1. Histopathological findings

Macroscopic studies showed that the tumor was a well-circumscribed, solid mass, with fibrous encapsulation measuring 15 mm × 14 mm × 13 mm.

Histopathological studies revealed that the tumor was composed of interstitial collagenous fibers, microvasculature, and proliferating spindle cells resembling fibroblast cells. We can see a hemangiopericytoma-like structure, which is characterized by cells surrounding thick-walled, branching "staghorn" blood vessels. The tumor cells were distributed irregularly and were inhomogeneous. The cellular atypia was poor, with almost no mitoses recognized [0/10 high-power fields (HPF)] (Fig. 3).

Immunohistochemical findings revealed that the tumor cells showed diffuse positive staining for CD34, vimentin, and bcl-2, but were negative for CD99, CK AE1/AE3, SMA, and S100 protein. These findings led to the diagnosis of SFT (Fig. 4).

3. Discussion

Previously, the diagnostic criteria of extrapleural SFT were unclear, and they were often misdiagnosed as hemangiopericytoma (HPC), which is a similar disease. In 1997, Chan established the disease concept and diagnostic criteria of SFT [4]. The 2002 WHO classification defined SFT as a ubiquitous mesenchymal tumor of a probable fibroblastic type that shows a prominent hemangiopericytoma-like branching vascular pattern [3]. There are two histological features of SFT: the arrangement of cells in short, ill-defined fascicles, randomly arranged in what has been described as a patternless pattern; and a hemangiopericytoma-like structure, which is characterized by cells surrounding thick-walled, branching "staghorn" blood vessels [3]. The present case report was considered to be typical, as it exhibited the above histological features. Malignant SFTs are usually hypercellular lesions, showing at least focally moderate to marked cytological atypia, tumor necrosis, numerous mitoses (≥4/10 HPF), and/or infiltrative margins [5]. In this case, mitosis was 0/10 HPF and no other malignant findings were observed. O'Regan et al. reported 21 cases of SFT of the oral cavity, and reported that SFT was observed in middle-aged adults 20–60 years of age, with no gender predilection [6]. The buccal mucosa was described as the most common site of occurrence in the oral cavity [7]. The case presented here was particularly rare, as there have been no previously reported cases of SFT in the chin.

The relation between trauma and the occurrence of SFT has been reported [8]. Alawi et al. [7], who reported the connection between trauma and SFT, supported the inference of Bucala et al. [9], who said that CD34-positive fibroblast-like cells multiply in the site of injury and function as a restoration mediator cell. The buccal mucosa, where biting wounds are most common, is the most common region of SFT occurrence in the oral cavity, suggesting an association with trauma. However, no relation between trauma and the lesion is clarified in this case because the patient indicated no history of trauma.

Generally, treatment of SFT is tumor excision, as a clear border is observed. However, in cases of confirmed malignant findings, such as infiltration, the inclusion of healthy surrounding tissue is necessary. The recurrence rate of SFT is low, at 0–8.3%, and that of SFT in the oral cavity only is still lower (2.6%) [10–12]. There are varied opinions regarding residual tumors following surgery. England et al. [13] reported that the presence of residual tumor tissue is the

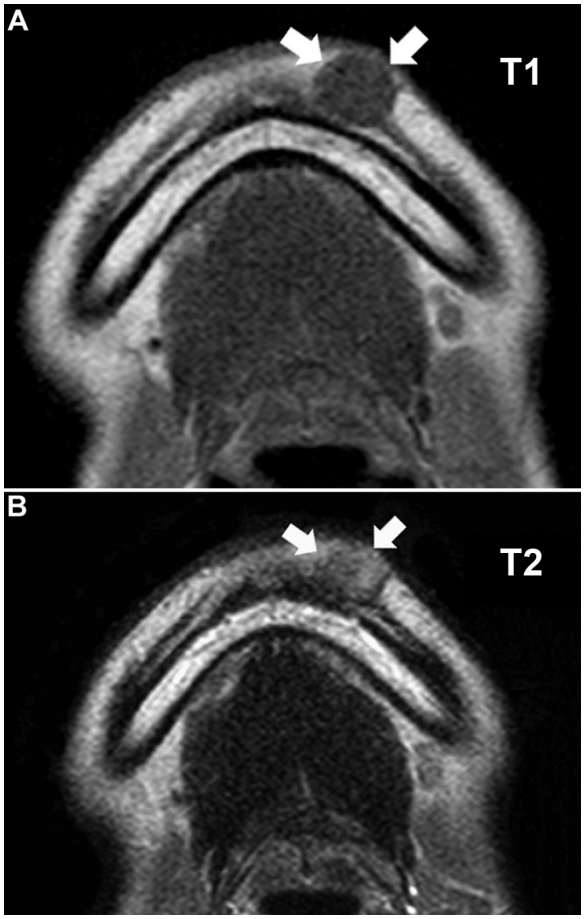

Fig. 2. Magnetic resonance imaging of initial visit. A magnetic resonance imaging examination showed a well-demarcated homogenous lesion of the left chin with a low signal intensity on T1-weighted images and a moderate signal intensity on T2-weighted images (arrow).

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