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Case Report

Solitary fibrous tumor composing benign and malignant components in the floor of the mouth: A case report

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ABSTRACT

Solitary fibrous tumor (SFT) is a rare mesenchymal neoplasm arising mostly in the pleura. We describe a case of SFT composed of benign and malignant components in the left floor of the mouth of an 88-year-old man presenting with a growing mass. A biopsy was diagnostically inconclusive because immunohistochemical staining for CD34 was negative. Postoperatively, the resected tumor was found to comprise 2 components of different cellularity and atypia. Immunohistochemically, tumor cells in both components were positive for vimentin, bcl-2 and STAT6 but showed opposing immunoreactivity to CD34. The malignant lesion with hypercellularity and atypia contained several small necrotic foci was immunohistochemically negative for CD34, and exhibited a high Ki-67 labeling index. On the other hand, the benign lesion of hypocellularity and no atypia was positive for CD34. Taken together, these findings indicated a rare case of SFT with distinctive histological features of benign and malignant components.

1. Introduction

Solitary fibrous tumor (SFT) is a rare spindle cell neoplasm of mesenchymal origin first reported by Klemperer and Rabin [1] in 1931 as arising mostly in the pleura. After the eight cases of SFTs reported as arising at extrapleural sites [2], others have been described in various tissues and organs including the oral cavity [3–5], suggesting that SFTs could develop at any extrapleural site. Those that develop in the oral cavity arise most commonly in the buccal region including the mucosa, then in the tongue and the lips [3,4]; however, only 2 cases have been reported in the floor of the mouth [6,7], and 39 cases in the oral cavity, with a male to female ratio and age distribution of 18:21 and 20–83 years (mean, 51 years), respectively [8].

Most SFTs are benign, but malignant ones and their histopathological features have been reported [9–12]. Pathologically, SFTs are characterized by a haphazard growth pattern (the so-called "patternless pattern") of short spindle cells with scant cytoplasm, a bland cytologic appearance separated by strands of rope-like collagen, and the presence of abundant blood vessels in a hemangiopericytoma-like pattern [13]. The diagnosis of SFT is dependent on microscopic features and immunohistochemical staining for CD34 and bcl-2 [6].

Here, we describe a rare case of SFT arising in the floor of the mouth, comprising benign and malignant components, and exhibiting different immunohistochemical reactivity to CD34 and Ki-67.

2. Case report

An 88-year-old man referred to the Department of Oral and Maxillofacial Surgery, Gunma University Hospital with chief complainment of a growing mass in the floor of the mouth over the previous 5 months. At first examination, a well-demarcated, elastic-hard, and movable tumor (50 mm × 40 mm × 30 mm) was noticed in the left floor of the mouth (Fig. 1A). X-ray images showed no destruction of the mandibular bones. Contrast-enhanced computed tomography (CT) revealed a tumor mass with a heterogeneous internal structure, and compressive resorption of the left mandible (Fig. 1B). ^{18}F-fluorodeoxyglucous-positron emission tomography (FDG-PET) revealed mild FDG uptake in the tumor

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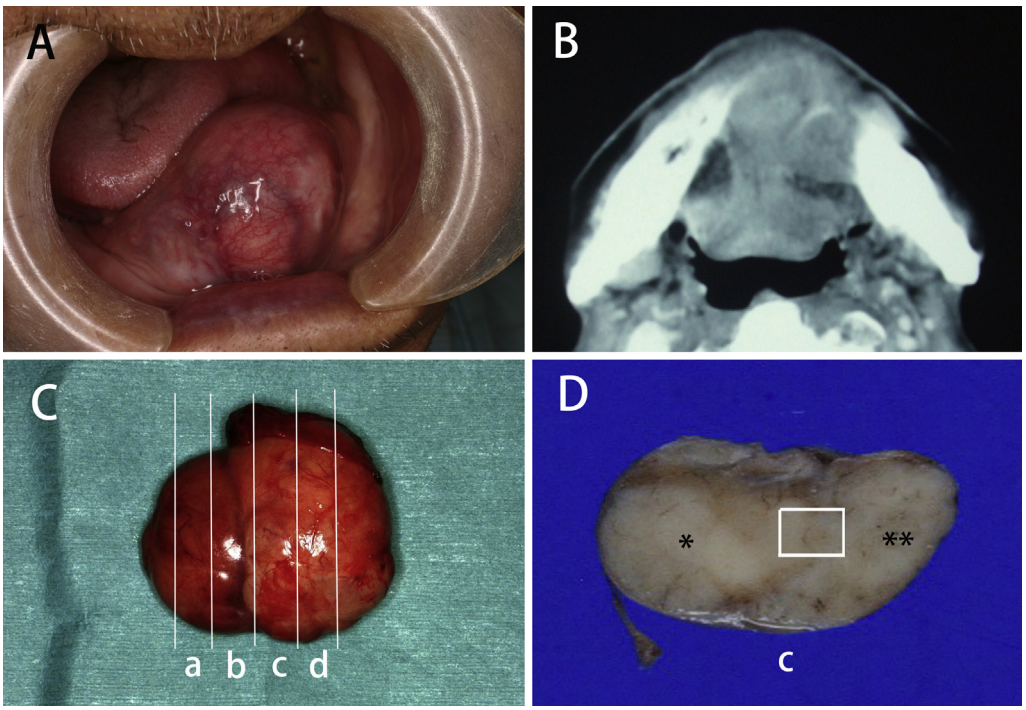

Fig. 1. (A) Clinical photograph shows the lesion (50 mm × 40 mm × 30 mm) in the floor of the mouth. (B) Axial computed tomography (CT) shows the mass of the lesion growing in the left side of the floor of the mouth. The lesion is divided into two components. (C) Macroscopic view shows the tumor comprising two lesions. The lines indicate the cutting lines, and the pieces named a, b, c, d. (D) The cut surface of the piece of 'c' in the panel C. The tumor exhibits macroscopically whitish (*) and macroscopically gray (**) solid area. The square area is corresponding to Fig. 2.

mass. No palpable regional lymphadenopathy was detected, and laboratory data were within normal limits.

Pathological analysis of a preoperative biopsy led to suspicion of a fibrous tumor of low-grade malignancy, especially that immunohistochemical staining was negative for α-smooth muscle actin (α-SMA), S-100, CD68, and CD34, but positive for vimentin. The surgically resected tumor measuring 50 mm × 45 mm × 20 mm was an encapsulated spherical mass with a groove on the surface, and comprised two different lesions (Fig. 1C). The cut surface of the tumor displayed gray and whitish solid areas (Fig. 1D) of different cellularity, and the border line was relatively clear. The macroscopically gray area showed hypocellularity (Fig. 2A, right), and the macroscopically whitish area showed hypercellularity (Fig. 2A, left). Histological examination of the macroscopically gray solid area revealed neoplastic spindle-shaped cells with small ovoid nuclei growing at a low density among abundant collagen fibers. The cells showed a patternless pattern and a storiform pattern (Fig. 3A and B), and about two mitotic cells per 10 high power fields (HPFs) were observed in this area. The macroscopically whitish solid area, on the other hand, disclosed atypical spindle-shaped cells proliferating irregularly in bundles at high cell density in a patternless pattern, and contained fewer collagen fibers than those in the macroscopically gray solid area (Fig. 3C and D). The tumor cells grew in an invasive manner to the fibrous area, and sometimes showed hemangiopericytoma-like structures. The tumor contained small, scattered necrotic foci and about eight mitotic cells per 10 HPFs.

Immunohistochemical staining of the tumor cells in the macroscopically gray solid area was strongly positive for vimentin, CD34, bcl-2 and STAT6, but negative for S-100 and α-SMA. The Ki-67 labeling index was 3.9% of tumor cells (Fig. 2B, right). While tumor cells in the macroscopically whitish solid area were strongly positive for vimentin and STAT6, and weakly positive for bcl-2, they were negative for CD34, S-100, and α-SMA. The Ki-67 labeling index was 10.5% of the tumor cells (Fig. 2B, left).

The typical features of SFTs (patternless pattern, storiform pattern) and the immunohistochemical positivity for CD34 in the macroscopically gray part of the tumor (Fig. 2C) and for bcl-2 and STAT6 in the whole tumor (Fig. 2D), led to the diagnosis of SFT. Moreover, the macroscopically whitish solid area, negative for CD34, was thought to be SFT of low-grade malignancy because of hypercellularity, atypical tumor cells, the more mitotic cells, necrosis and the increased expression of Ki-67. This area appeared to have progressed to tumor cells from the benign SFT. Therefore, based on the histopathological analysis, the whole tumor was diagnosed as SFT composed of benign and malignant components.

In the present case, the patient died of pancreatitis that was not related with SFT at the other hospital 6 months postoperatively, although no recurrence or metastasis of SFT was observed during the follow-up period.

3. Discussion

SFTs are usually immunohistochemically positive for CD34, a surface marker of vascular endothelial cells and hematopoietic stem cells; therefore, they are uniquely mesenchymal, mostly benign, spindle cell tumors characterized by positive CD34 immunoreactivity. CD34 positive dendritic and spindle cells that are not related to vessels are residents of deep submucosa of normal oral tissue [14]. Therefore, SFTs at oral sites may develop from these CD34 positive cells. In the present case, the tumor was strongly positive for CD34, bcl-2 and STAT6 in the macroscopically gray solid area, but was negative for CD34 and positive for bcl-2 and STAT6 in the macroscopically whitish solid area. Since the biopsy specimen might have been taken from the macroscopically whitish solid area, it is likely that the immunohistological result was negative for CD34, leading to the confusion in the diagnosis. Recently it was found that STAT6 has highly sensitivity and it can be specific immunohistochemical marker for SFT [15]. In resected specimen,