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Solitary Fibrous Tumor of the Buccal Mucosa: A Patient Report

Kazuhiko Tanio, Kazuo Ryoke, Isamu Kodani, Rieko Doi, Yoshiyuki Tanji, Kazuhito Tohashi and Takayuki Ikeda*

*Division of Oral and Maxillofacial Biopathological Surgery, Department of Medicine of Sensory and Motor Organs, School of Medicine, Tottori University Faculty of Medicine, Yonago 683-8504 and *Clinic of Dentistry and Oral Surgery, Tottori Prefectural Central Hospital, Tottori 680-0901 Japan*

Solitary fibrous tumor (SFT) is a soft tissue tumor most frequently localized in the pleura, but it has recently been described in other body sites. We have encountered a rare patient with an SFT of the buccal mucosa. We examined the case clinically, histopathologically and immunohistochemically. Dimension of the resected tumor was 3 by 2.5 by 3 cm. Histological observation revealed that the tumor was composed of spindle- or ovoid-shaped cells with varying amounts of haphazardly arranged collagen bundles. Immunohistochemically, the tumor cells exhibited strong staining with CD34 and bcl-2 but were negative to smooth muscle actin, ckit and S-100 protein. The patient was discharged 7 days after surgery, with no signs of recurrences after 14 months. We reported a rare case of buccal SFT and reviewed 38 cases of intraoral SFT.

Key words: buccal mucosa; CD34; immunohistochemistry; oral cavity; solitary fibrous tumor

Solitary fibrous tumor (SFT) is a rare neoplasm composed of spindle cells which Klemperer and Rabin (1931) first described in the pleura. In early reports, SFT was thought to arise from the mesothelial cell and was once termed localized fibrous mesothelioma (England et al., 1989). However, recent immunohistochemical, ultrastructural and tissue cultural studies have suggested that the origin of this tumor is the mesenchymal tissue (Dervan et al., 1986; Ali et al., 1997). Thereafter, SFTs at almost every anatomic location have been reported (Goodlad et al., 1991), but very rarely in the oral cavity. In the present paper, we present a case of SFT originating from the buccal mucosa.

Abbreviations: MRI, magnetic resonance imaging; SFT, solitary fibrous tumor; SMA, smooth muscle actin; GIST, gastrointestinal stromal tumor

Patient Report

In 2002, a 54-year-old man noticed a mass in the left buccal mucosa. The mass had been asymptomatic and had not increased in size. In 2003, he visited a hospital for cytological examination of the mass, where its cytology was interpreted as benign. In March 2004, he was referred to our clinic for further examination of the mass.

Intraoral examination revealed a hard, elastic and nontender mobile mass which was about 3 cm in size, 0overlayed by apparent normal buccal mucosa in its color and texture (Fig. 1). There was neither swelling of the cervical lymph-nodes nor any significant change in laboratory data. Magnetic resonance imaging (MRI) revealed that the lesion was a well-circumscribed, solid mass measuring

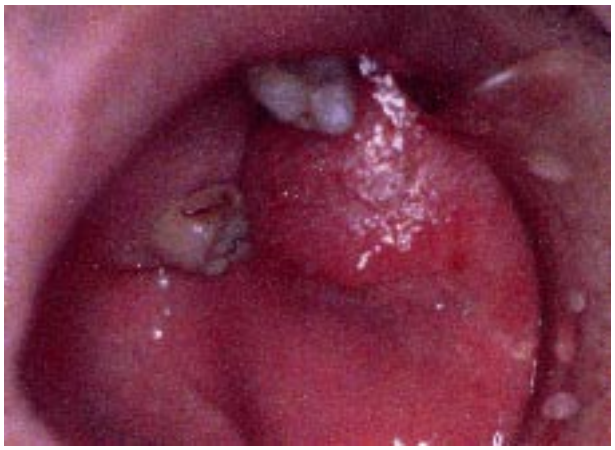

Fig. 1. An elastic hard mobile mass about 3 cm in size could be palpated intraorally.

about 3 cm in the right posterior buccal space. The mass showed a signal intensity homogeneous with that of the muscle on both T1- and T2-weighted images (Fig. 2). Surgical extirpation was performed under general anesthesia in April 2004. The tumor surface was smooth and clearly defined and was easily dissected from the surrounding tissue. The tumor was oval-shaped, and measured 3 by 2.5 by 3 cm. The cut surface of the tumor was solid, firm, grayish-white and homogeneous (Fig. 3).

Histologically, the tumor was well-circumscribed and encapsulated with thin fibrous tissues. The lesion was composed of spindle- or ovoid-shaped cells with a various amount of collagen bundles haphazardly arranged. The tumor showed a partially slight storiform pattern appearance (Fig. 4) and was vascularized, occasionally contained areas of dilated vessels with a stag-horn appearance sometimes seen in hemangiopericytoma (Fig. 5). The mitotic count was 0/10 high-power-field. There was no necrosis.

Through immunohistochemical studies on paraffin-embedded tissue sections, the tumor cells were strongly positive to CD34 and bcl-2, but negative to smooth muscle actin (SMA), c-kit and S-100 protein (Fig. 6). These findings resulted in a diagnosis of SFT.

After surgery, the patient improved progressively and was discharged with no difficulty. Since

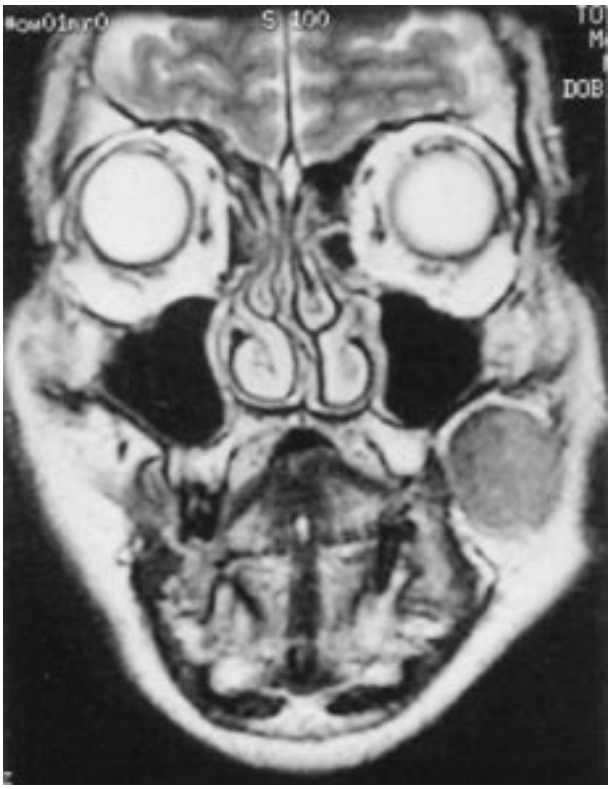

Fig. 2. MRI shows a well-circumscribed mass about 3 cm in diameter. The mass shows a signal intensity homogeneous to that of the muscle on both T1- and T2-weighted images.

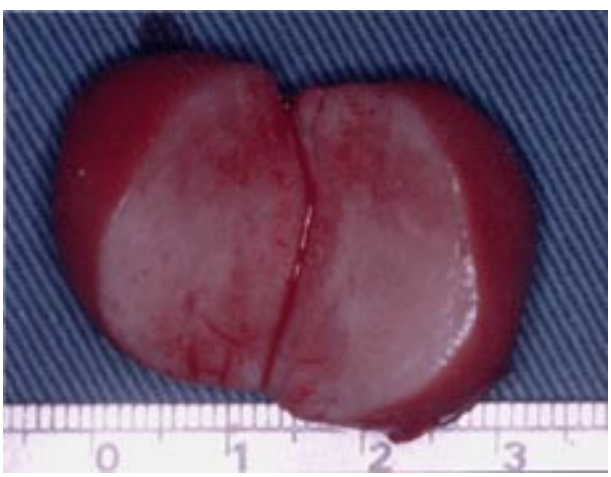

Fig. 3. The cut surface of the tumor is solid, firm, grayish-white and homogeneous.

then, the patient was followed-up for 1 year in the outpatient clinicand has showed no signs of recurrence as of this writing.

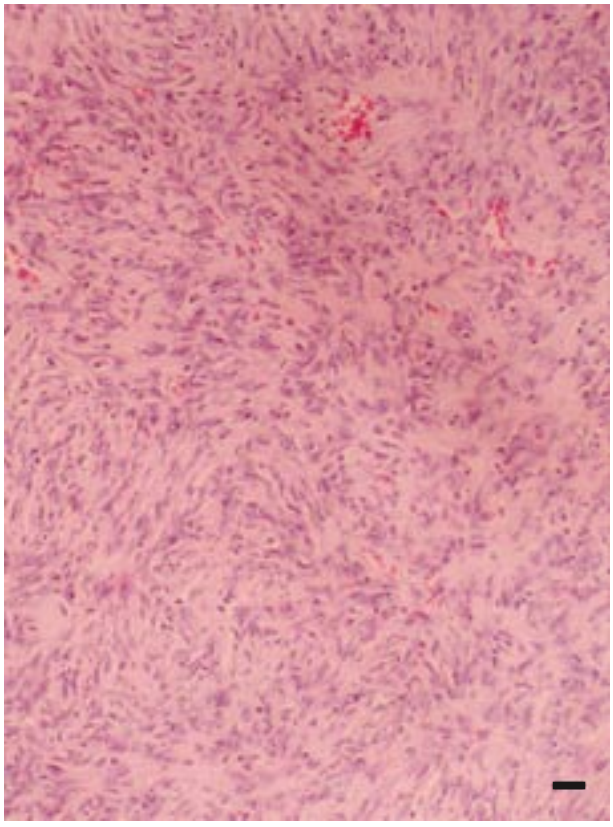

Fig. 4. The tumor shows a partially storiform pattern appearance (hematoxylin and eosin stain). Bar =100 μm.

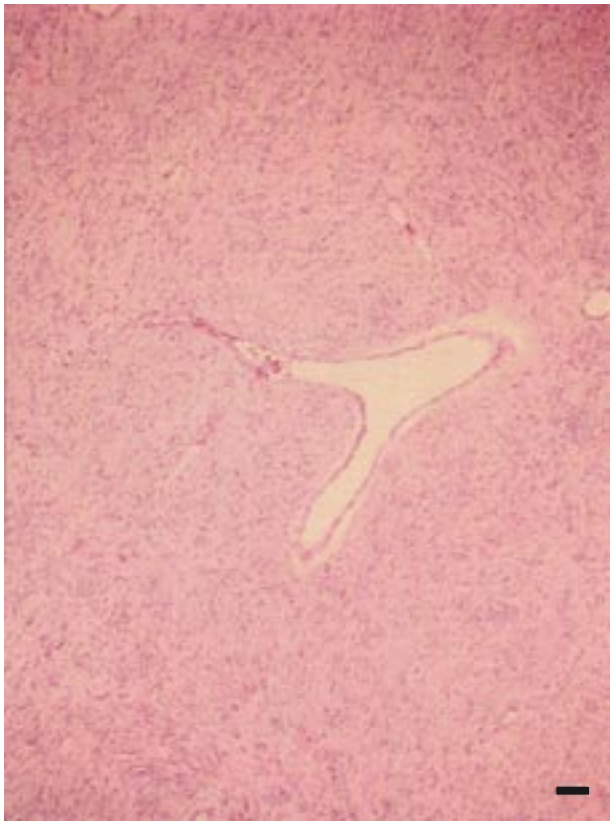

Fig. 5. The SFT tumor is vascularized and partially contains areas of dilated vessels with a stag-horn appearance also occasionally seen in hemangiopericytoma (hematoxylin and eosin stain). Bar = 160 μm.

Discussion

Current advances in immunohistochemical, ultrastructural and tissue cultural techniques have greatly contributed to studies on SFTs. Since Gunhan (1994) first described SFT in the oral cavity, many intraoral SFTs were found and 38 patients were reported in the literature, all of it in English (Gunhan et al., 1994; Suster et al., 1995; Piatteli et al., 1998; Iwai et al., 1999; Kurihara et al., 1999; Perez-Ordonez et al., 1999; Brunnemann et al., 1999; Lukinmaa et al., 2000; Alawi et al., 2001; Hirano et ai., 2001; Kuo et al., 2001; Shin et al., 2001; Harada et al., 2002; Hardisson et al., 2002; Vargas et al., 2002; Shnayder et al., 2003; Yanamoto et al., 2003). The clinicopathological features of these oral SFT patients including the present one are summarized in Table 1 with no significant sex predilection (21 females, 18 males). SFTs develop in adulthood over a wide age range, and most are identified between 40s and 70s (mean 51.3 years) with tumor sizes varing 8 to 45 mm (mean 21.5 mm). Patients with pleural large SFTs have symptoms such as chronic cough, short of breath, osteoarthropathy, chest pain and hypoglycemia (Weiss et al., 2001). In the present series of studies (Table 1), no such symptoms have been identified in patients with intraoral SFT except one. This exception was a female patient with malignant SFT of the tongue, and actually had a history of dysarthria and dysphagia (Shnayder et al., 2003). She was the only case of malignant SFT of the oral cavity, with a 25-year history of an apparently stable tongue mass. However, the mass increased in size, and her symptoms were aggravated for 6 months before admission. Therefore, we should suspect

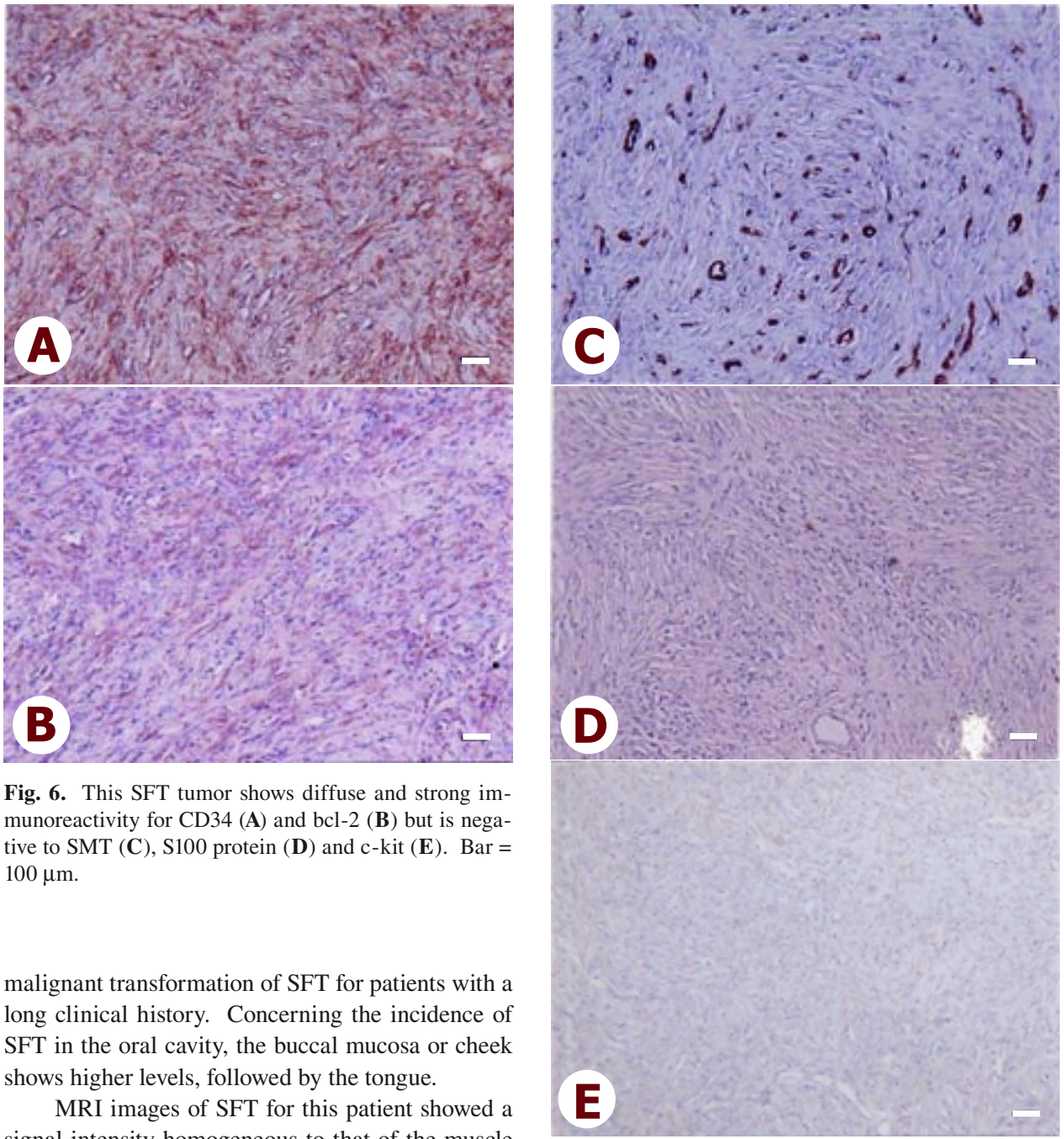

Fig. 6. This SFT tumor shows diffuse and strong immunoreactivity for CD34 (**A**) and bcl-2 (**B**) but is negative to SMT (**C**), S100 protein (**D**) and c-kit (**E**). Bar = 100 μm.

malignant transformation of SFT for patients with a long clinical history. Concerning the incidence of SFT in the oral cavity, the buccal mucosa or cheek shows higher levels, followed by the tongue.

MRI images of SFT for this patient showed a signal intensity homogeneous to that of the muscle on both T1- and T2-weighted images. Likewise, Sato et al. (1998) reported that low signal levels on both T1- and T2-weighted images would be characteristic of SFT. However, even though some reports showed low levels on T1-weighted images, high levels on T2-weighted images were also seen (Yanamoto et al., 2003). We think that the histological diversity of SFT leads to the difference in MRI findings.

Generally, diagnosing SFT is difficult because of its broad range of morphologic characteristics, including storiform, hemangiopericytic, herringbone and palisading areas (Chan, 1997). In the present case, we observed a storiform pattern along with a stag-horn appearance occasionally seen in hemangiopericytoma. Therefore, the current diagnosis of SFT has been based on histologi-

Table 1. Clinical and pathologic features of 39 patients with intraoral SFT

Number	Report		Age (yr)	Sex	Location	Size (mm)	Clinical history	CD34	Vimentin	bcl-2	SMA	Desmin	S-100	Cytokeratin	F-8	ckit
1	Gunhan et al.	1994	55	M	Sublingual gland	30	12 yr	NA	+	NA	NA	–	–	NA	NA	NA
2	Suster et al.	1995	50	F	Soft palate	40	NA	+	+	NA	NA	–	–	NA	–	NA
3	Suster et al.	1995	83	M	Cheek	15	NA	+	+	NA	NA	–	–	NA	–	NA
4	Piatteli et al.	1998	66	F	Tongue	10	1 yr	+	+	+	NA	NA	–	–	–	NA
5	Iwai et al.	1999	27	M	Buccal mucosa	20	5 yr	+	+	NA	NA	NA	–	NA	NA	NA
6	Kurihara et al.	1999	34	F	Buccal mucosa	15	3 yr	+	+	NA	–	–	–	–	–	NA
7	Perez-Ordonez et al.	1999	39	F	Mandible	10	1 mo	+	+	+	NA	–	–	NA	NA	NA
8	Perez-Ordonez et al.	1999	70	M	Buccal mucosa	40	NA	+	+	–	NA	–	–	NA	NA	NA
9	Brunnemann et al.	1999	46	F	Buccal mucosa	29	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
10	Brunnemann et al.	1999	53	M	Oral mucosa	22	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
11	Lukinmaa et al.	2000	56	M	Cheek	20	10 yr	+	+	+	–	NA	–	–	NA	NA
12	Lukinmaa et al.	2000	45	F	Cheek	15	6 mo	+	+	+	–	NA	–	–	NA	NA
13	Lukinmaa et al.	2000	70	M	Cheek	10	6 mo	+	+	+	–	NA	–	–	NA	NA
14	Hirano et al.	2001	64	F	Mental	32	9 yr	+	+	NA	+	–	–	NA	NA	–
15	Kuo et al.	2001	31	M	Infraorbital	40	4 yr	+	+	+	–	NA	–	–	NA	NA
16	Alawi et al.	2001	44	F	Tongue	11	NA	+				NA			NA	NA
17	Alawi et al.	2001	40	M	Buccal mucosa	12	1 yr	+				NA			NA	NA
18	Alawi et al.	2001	55	M	Buccal mucosa	33	3 yr	+				NA			NA	NA
19	Alawi et al.	2001	51	F	Buccal mucosa	40	3 yr	+				NA			NA	NA
20	Alawi et al.	2001	38	F	Buccal mucosa	14	NA	+				NA			NA	NA
21	Alawi et al.	2001	31	F	Buccal mucosa	30	2 yr	+				NA			NA	NA
22	Alawi et al.	2001	61	M	Buccal mucosa	33	2.5 yr	+	+, 13/13†		+, 2/13†	NA		+, 0/5†	NA	NA
23	Alawi et al.	2001	63	M	Buccal mucosa	9	NA	+				NA			NA	NA
24	Alawi et al.	2001	60	F	Buccal mucosa	8	15 yr	+		+, 12/13†		NA	+, 0/10†		NA	NA
25	Alawi et al.	2001	58	F	Buccal mucosa	8	NA	+				NA			NA	NA
26	Alawi et al.	2001	64	F	Lower Lip	13	11 mo	+				NA			NA	NA
27	Alawi et al.	2001	67	F	Buccal mucosa	12	NA	+				NA			NA	NA
28	Alawi et al.	2001	60	M	Buccal mucosa	17	NA	+				NA			NA	NA
29	Alawi et al.	2001	73	F	Buccal mucosa	13	50 yr	+				NA			NA	NA
30	Alawi et al.	2001	48	F	Tongue	8	8 mo	+				NA			NA	NA
31	Alawi et al.	2001	36	M	Lower Lip	8	4 mo	+				NA			NA	NA
32	Shin et al.	2001	46	M	Buccal mucosa	27	1 mo	+	+	NA	NA	–	–	–	NA	NA
33	Harada et al.	2002	32	M	Gingival	10	5 yr	+	+	NA	–	–	–	NA	NA	NA
34	Hardisson et al.	2002	56	F	Cheek	15	2 yr	+	+	+	NA	–	–	NA	–	NA
35	Vargas et al.	2002	20	F	Cheek	30	6 mo	+	+	+	–	–	–	–	NA	NA
36	Vargas et al.	2002	65	F	Tongue	48	15 yr	+	+	+	–	–	–	–	NA	NA
37*	Shnayder et al.	2003	57	F	Tongue	25	25 yr	+	+	+	–	–	–	–	NA	–
38	Yamamoto et al.	2003	32	M	Buccal mucosa	35	1 yr	+	+	+	–	–	–	–	–	NA
39	Tanio et al.	Present	56	M	Buccal mucosa	30	24 mo	+	NA	+	–	NA	–	NA	NA	–

* Case 37 was the only malignant tumor in this series.

† Positive case(s)/examined cases reported in Alawi et al. (2001).

F, female; M, male; mo, month(s); NA, not available; SFT, solitary fibrous tumor; SMA, smooth muscle actin; yr, year(s).

cal and immunohistochemical findings. In 1997, Chan proposed an immunohistochemical profile in which SFT is immunopositive to vimentin, CD34 and bcl-2, but is negative to cytokeratin, von Willebrand factor, SMA and S-100 protein. In 56 pleural and extrapleural SFTs, Suster et al. (1998) showed a close correlation between the expressions of CD34 and bcl-2. The present case and other cases shown in Table 1 were positive to CD34 and bcl-2. Although many reports have emphasized the importance of CD34 in diagnosis of SFT, CD34 immunopositivity is not specific to SFT. Tumors including hemangiopericytoma, neurofibroma, neurilemmoma, angioleiomyoma, gastrointestinal stromal tumor (GIST) and dermatofibrosarcoma protuberans are positive to CD34. Neurofibroma and neurilemmoma are usually positive to S-100 protein, whereas SFT is negative. Dermatofibrosarcoma protuberans is rarely involved in oral soft tissues, but it can be distinguished from SFT by its malignant cytologic features. Also, angioleiomyoma is positive to SMA, where SFT is usually negative. GIST is positive to c-kit. Hemangiopericytoma is probably the most difficult to be distinguished from SFT, because SFT often presents hemangiopericytoma-like areas. Hemangiopericytoma also has immunoactivity for CD34 like SFT. However, hemangio-pericytoma lacks the other morphologic features associated with SFT, such as the storiform pattern, herringbone pattern, neural palisades and thick collagenous stroma. These facts convinced us that we should differentiate this case as SFT.

Diagnosis of SFT is difficult and, although uncommon, it should be contained in the differential diagnosis of oral soft tissue tumors. We reported a rare case of SFT and reviewed 38 cases of intraoral SFT.

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Correspondence: Kazuhiko Tanio, DDS, PhD