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Case Report

Solitary fibrous tumor of the buccal mucosa: A case report

Hiroki Yamaguchi[1], Koki Takamatsu*[1,2], Yuya Kurasawa[3], Arisa Yasuda[1], Mai Kobashi[1], Takaaki Kamatani[1], Toshikazu Shimane[3] and Tatsuo Shirota[1]

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Abstract

Solitary fibrous tumors (SFTs) were reported in 1931 as tumors derived from mesenchymal tissue. They occur mainly in the pleura and peritoneum and rarely in the oral and maxillary regions. This report presents the case of a 48-year-old woman with a tumor in the left buccal mucosa. We suspected a salivary gland tumor or hemangioma. Excision was performed under general anesthesia. The lesion was removed as a single mass. Histopathological and immunohistochemical analyses led to the diagnosis of SFT. We report this case based on a literature review.

Key words: solitary fibrous tumor, mesenchymal neoplasm, immunohistochemistry

Introduction

Solitary fibrous tumors (SFTs) were first reported by Klemperer and Rabin in 1931 as mesenchymal neoplasms[1]. They are mesenchymal cell-derived tumors mainly occurring in the peritoneum and pleura[2]. There have been reports on salivary gland SFTs in the oral cavity, but the occurrence rate is low and rare[3]. Histopathological findings vary, making it difficult to make a definitive diagnosis. Thus, it is necessary to use immunohistochemical markers with the histopathological findings[4,5]. This report aims to present a case of SFT in the left buccal mucosa.

Case report

A 48-year-old female who noticed a painless swelling in the left buccal mucosa in June 2020 was referred to our department in November 2020 to investigate the swelling. Initial examination showed an 8×8 mm movable elastic mass in the left buccal mucosa (Fig. 1) without spontaneous pain, tenderness, or numbness. The salivary gland orifice was far away, and no ulceration or other findings were observed on the superficial layer of the buccal mucosa. Laboratory results were within normal limits. Contrast-enhanced computed tomography (CT) was performed the following month, which showed a mass lesion approximately 8×8×7 mm in size. The lesion boundaries were clear, and no invasion of the surrounding tissue was observed on imaging, suggesting a minor salivary gland tumor such as Warthin's tumor or hemangioma (Fig. 2). Based on the diagnosis of a minor salivary gland tumor or hemangioma, the tumor was excised under general anesthesia. The tumor was detached from the buccal submucosa and was excised as a single mass. No abnormal bleeding or other findings were observed at the removal site. Furthermore, the surrounding minor salivary gland tissue was excised. However, no evidence of infiltration into the surrounding tissues was observed (Fig. 3). Histological examination of the excised specimen showed a well-defined nodular lesion with a coating under the coated normal mucosal epithelium and no infiltration of the surrounding minor salivary gland tissue. The cells inside the lesions were spindle-shaped and proliferated in bundles, with a high cell density and round or oblong nuclei (Fig. 4a, b).

* Corresponding author
✉ Koki Takamatsu
k.1020@dent.showa-u.ac.jp

[1] Department of Oral and Maxillofacial Surgery, School of Dentistry, Showa University, 2-1-1 Kitasenzoku, Ota-ku, Tokyo 145-8515, Japan.
[2] Division of Oral and Maxillofacial Surgery, Department of Sensory and Motor Organs, School of Medicine, Faculty of Medicine, Tottori University.
[3] Department of Oral and Maxillofacial Surgery, Division of Oral Oncology, School of Dentistry, Showa University.

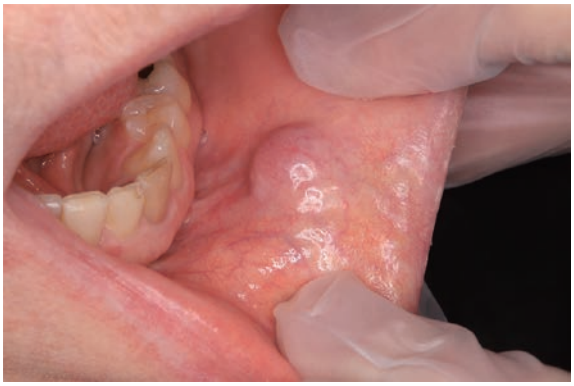

Fig. 1. An 8×8 mm soft mass with normal superficial mucosa is visualized on the buccal mucosa of the left mandibular first premolar

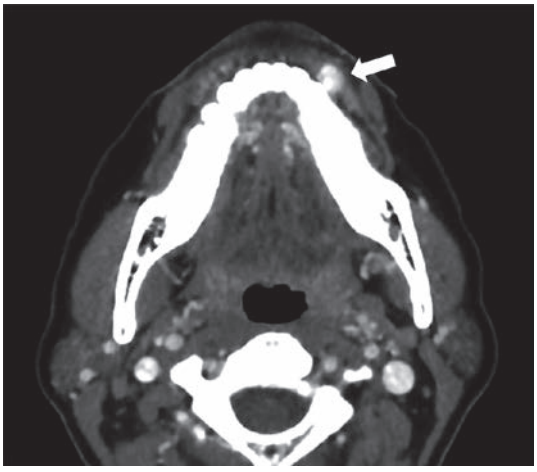

Fig. 2. A well-defined nodular lesion with heterogeneous enhancement is observed on contrast-enhanced CT images

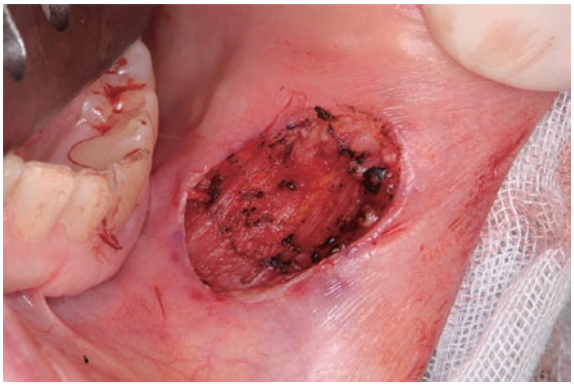

Fig. 3. The tumor, the buccal mucosa, and the surrounding minor salivary glands are removed as a single mass

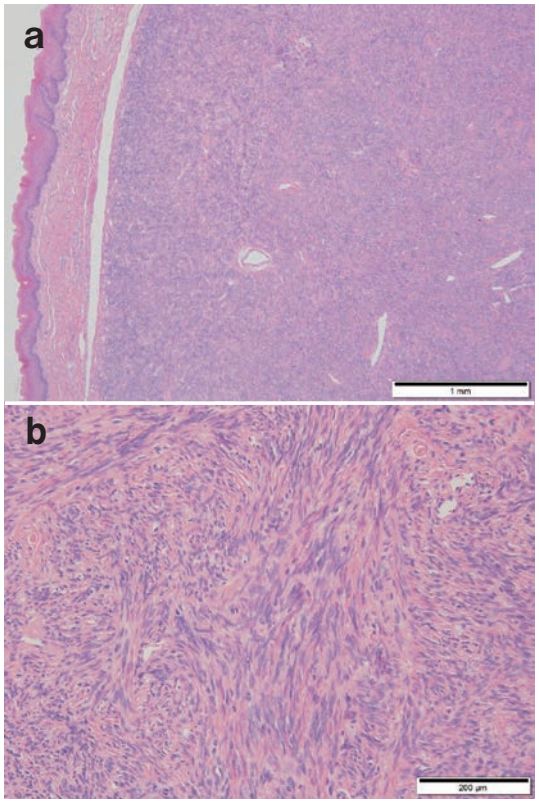

Fig. 4. (a, b) Hematoxylin and Eosin staining
a well-defined nodular lesion with a coating is observed under the epithelium of the coated mucosa, and a complex proliferation of spindle-shaped cells in bundles can be observed inside the lesion.

Nuclear atypia was scarce, and the mitotic count was approximately 1 mitosis per 10 high-power fields. We suspected neoplastic lesions of mesenchymal origin, such as fibromas or leiomyomas, and performed an immunohistochemical analysis. Immunohistochemical staining was performed using antibodies against tumor protein (p63), alpha-smooth muscle actin (α-SMA), S-100 protein, epithelial membrane antigen (EMA), CD34, B-cell lymphoma 2 (Bcl-2), CD99, and signal transducer and activator of transcription 6 (STAT6). The tumor cells were negative for p63, α-SMA, S-100, and EMA (Fig. 5) and did not differentiate into muscular, nervous, or epithelial lineages. The cells were positive for CD34, Bcl-2, and CD99, and some were positive for STAT6 (Fig. 6). The Ki-67 positivity rate was approximately 5% (Fig. 7). These results led to the diagnosis of an SFT.

Postoperative follow-up was conducted every few months. No recurrence or malignancy has been observed to date.

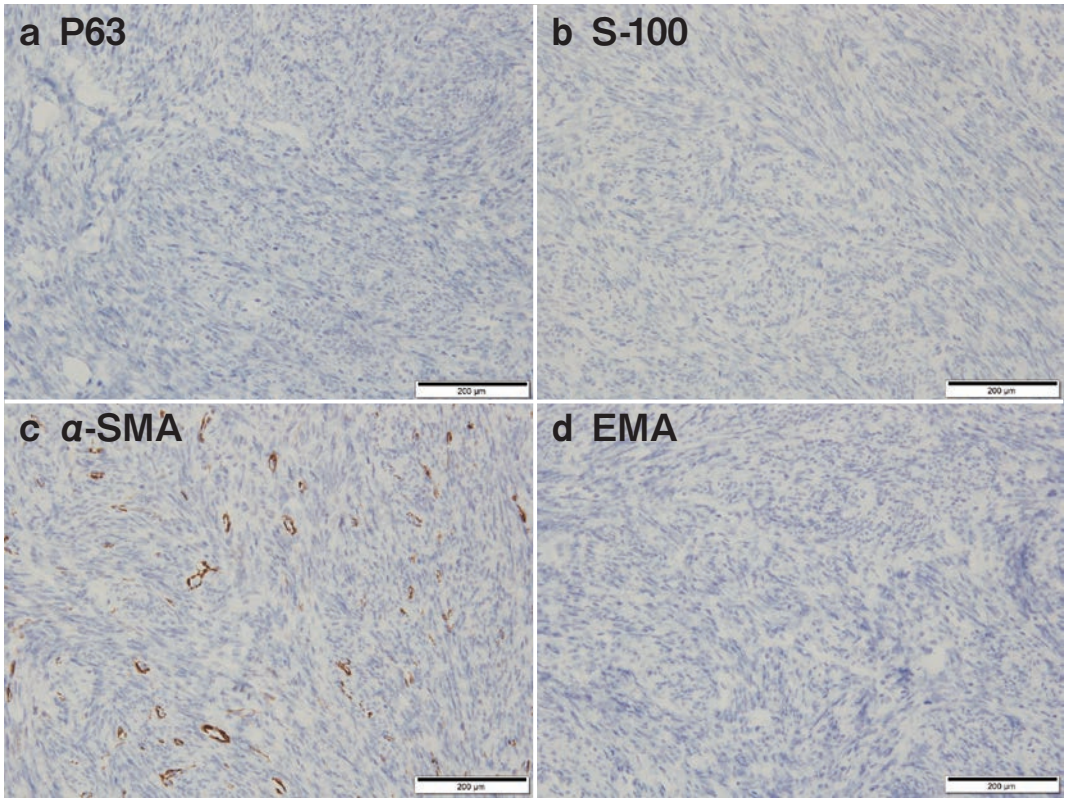

Fig. 5. Immunohistochemical staining
(a) tumor protein (p63), (b) S-100 protein, (c) alpha-smooth muscle actin (α-SMA), and (d) epithelial membrane antigen (EMA).

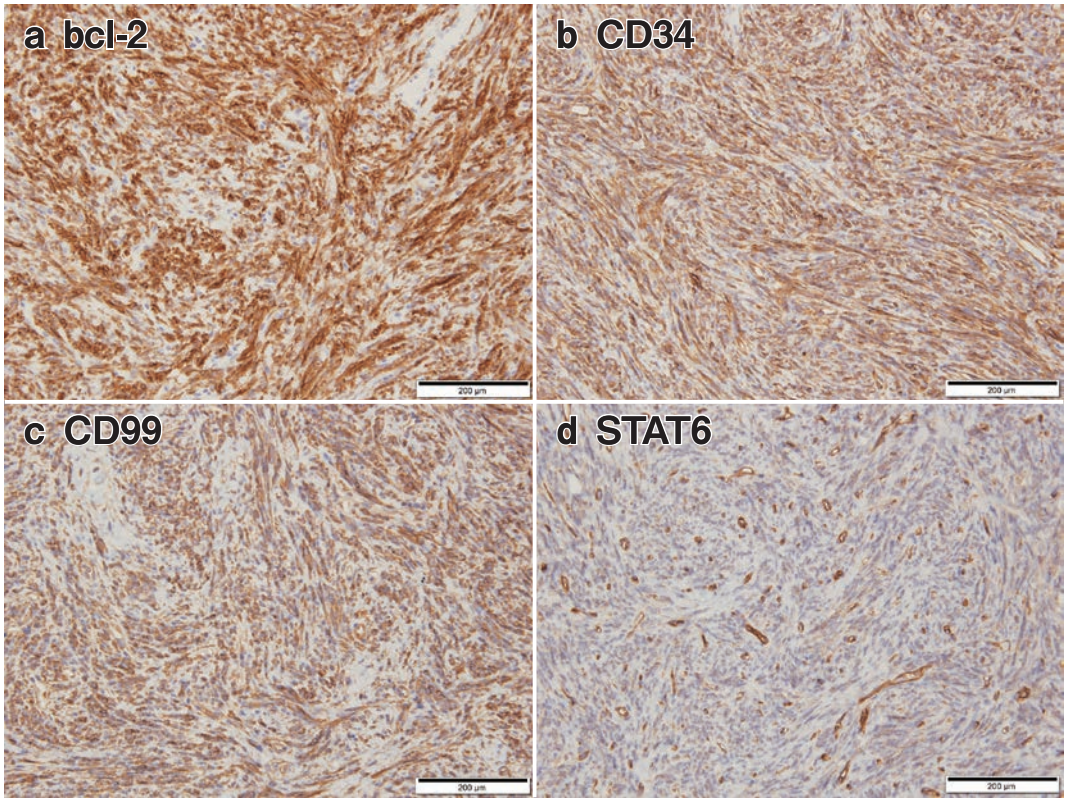

Fig. 6. Immunohistochemical staining
(a) B-cell lymphoma-2 (Bcl-2), (b) CD34, (c) CD99, and (d) signal transducer and activator of transcription 6 (STAT6).

Discussion

SFTs occur in the peritoneum, pleura, and oral and nasal cavities. However, its occurrence in the oral cavity is rare, and there have been reports on salivary gland SFTs[6,7]. Table 1 shows reports on SFTs in the buccal mucosa in Japan since 2001. These reports indicated that the mean age of the patients was approximately 59 years, with no sex difference. Regarding the difference between the left and right sides, the left side was more common in 10 of 13 cases. Out of the 13 cases, including this case, recurrence occurred in only one case. These results showed that SFTs occurring in the oral region have a low risk of recurrence. However, it has been reported that recurrence is more likely to occur when the Ki-67 positivity rate in SFT tumor cells exceeds 5%[8]. In our case, although no evidence of necrosis was observed in the tumor cells, the Ki-67 positivity rate was 5%, suggesting that careful follow-up was necessary.

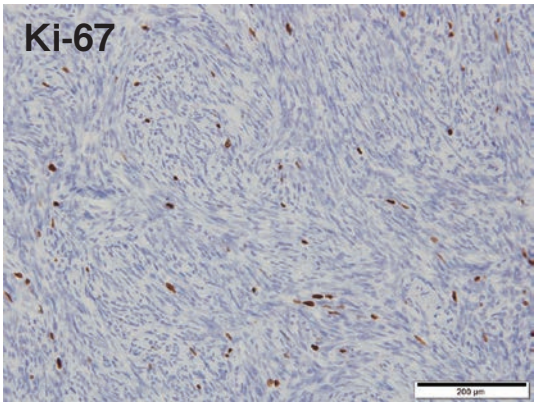

Fig. 7. Immunohistochemical staining: Ki-67

As the histological findings of SFTs are mainly fibroblast-like cell proliferation, histopathological findings are important in the diagnosis of SFT. Previous reports have shown that SFT cases do not present with any specific clinical symptoms. Diagnosis based on imaging findings alone is difficult, and histological findings are important. In our case, the patient visited our clinic complaining of painless swelling, and histological examination was necessary to make a definitive diagnosis. The differential diagnosis of SFT includes tumors of mesenchymal origin, such as cellular schwannoma, myofibroblastoma, and cellular angiofibroma[9], and immunohistochemistry should be performed. Moreover, CD34, a mesenchymal cell marker, has been reported to be effective. However, since it is not a specific marker, a combination of Bcl-2, α-SMA, and S-100 is recommended for diagnosis[4,5]. Bcl-2 has been reported to be an oncoprotein marker associated with the inhibition of apoptosis but not with SFT grading[7]. In our case, the tumor cells tested positive for Bcl-2,

Table 1. Reports on SFTs in the buccal mucosa in Japan since 2001

Author	Year	Age	Gender	Location	Size (mm)	Symptoms	Treatment	Recurrence (Interval, mos)
Yamaguchi *et al*	2021	48	F	Left Buccal mucosa	8×8×7	Painless tumor	Extraction	No (9)
Kin *et al*	2021	74	F	Right Buccal mucosa	40	Painless tumor	Resection	No (28)
Okabe *et al*	2021	27	F	Right Buccal mucosa	15×12×11	Painless tumor	Extraction	No (17)
Minamiyama *et al*	2019	50	F	Right Buccal mucosa	30×20	Painless tumor	Resection	No (60)
Ito *et al*	2018	78	M	Left Buccal mucosa	35×25	Painless tumor	Extraction	No (28)
Ito *et al*	2018	56	F	Left Buccal mucosa	12×10	Painless tumor	Extraction	Yes (2)
Oturu *et al*	2016	73	M	Left Buccal mucosa	20×15	Painless tumor	Resection	No (72)
Shibatuji *et al*	2010	72	M	Left Buccal mucosa	15×12	Painless tumor	Extraction	No (5)
Murase *et al*	2009	64	M	Left Buccal mucosa	30×20	Painless tumor	Extraction	No (9)
Fjii *et al*	2008	54	M	Left Buccal mucosa	40×30×20	Painless tumor	Extraction	No (14)
Tomihara *et al*	2008	52	F	Left Buccal mucosa	33×25×18	Painless tumor	Extraction	No (18)
Tanio *et al*	2005	54	M	Left Buccal mucosa	30×25×30	Painless tumor	Resection	No (14)
Kawahara *et al*	2003	52	F	Left Buccal mucosa	20×15×15	Painless tumor	Extraction	No (5)

which may be a useful diagnostic marker. Recently, STAT6 has been reported to be a useful marker for the diagnosis of SFT. STAT6 is an NAB2-STAT6 fusion gene reported in 2013 as a gene associated with cell proliferation. It was initially reported as a soft tissue marker in soft tissues such as the lung and liver[10, 11]. SFTs were defined as NAB2-STAT6 fusion gene-associated tumors of the fibroblastic phenotype with branchial vascularity in the 2017 WHO classification of the head and neck. In our case, STAT6 was also positive, suggesting that it is a useful diagnostic marker. Therefore, this is a valuable case in which the diagnosis of SFT was confirmed both histologically and immunohistochemically.

Surgery is the first choice for SFT treatment. SFTs are infrequent but tend to recur. Therefore, complete tumor excision is recommended. In our case, the tumor was excised as a single mass with the surrounding minor salivary gland tissues. Radiotherapy may be indicated if SFTs are suspected to be malignant or margin-positive. However, opinions regarding radiotherapy efficacy vary[12, 13].

Conclusion

We describe in this report our experience with an SFT of the left buccal mucosa based on a literature review. At the 9-month postoperative follow-up, the patient was well with no apparent signs of recurrence.

Acknowledgments

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Conflicts of interest

The authors declare that there are no conflicts of interest.

Consent for publication

Consent was obtained from all coauthors.

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