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Oval-Shaped Solitary Fibrous Tumor of the Tongue

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Abstract

Solitary fibrous tumor (SFT) is a rare neoplasm that mostly occurs as a pleural or serosal tumor. The present study reports a 28-year-old man who presented to the hospital with a 10-year old history of a painless mass in the tongue that he noticed after a tongue bite. He was suspected with a benign tumor, and an excisional biopsy was performed under general anesthesia. The histopathological examination of the specimen confirmed SFT. The patient's condition was well and free of disease at 12 months after the surgery. In this present case study, we concluded that SFT may be considered as a differential diagnosis of soft tissue tumors in the oral cavity.

Introduction

Solitary fibrous tumor (SFT) was firstly described by Klemperer and Robin in 1931 as a mesenchymal tumor in the pleura [1]. These SFTs are observed in tumors arising in various areas of the body including the oral cavity, but their occurrences in the tongue are very rare [2-8]. SFT is composed of spindle-shaped fibroblastic cells and exhibits immunoreactivity for CD34 in many cases [2-8]. According to 2002 WHO classification of Pathology and Genetics of Tumors of Soft Tissue and Bone, extrapleural SFT is defined as an intermediate malignant tumor having a high tendency of local recurrence and a low propensity of metastasis. The present study reports a case of SFT of the tongue through 10 years starting from the awareness of the non-malignant mass.

Case Report

A 28-year-old man presented to Kimitsu Chuo hospital with a 10-year old history of a painless mass in the tongue, which he noticed after a tongue bite. The mass slowly increased in size and became asymptomatic. Intraoral examination revealed a well-circumscribed elastic, hard, and movable mass measuring 40 mm × 30 mm × 20 mm on the tip of the tongue (Figure 1). However, the patient was healthy and in good condition.

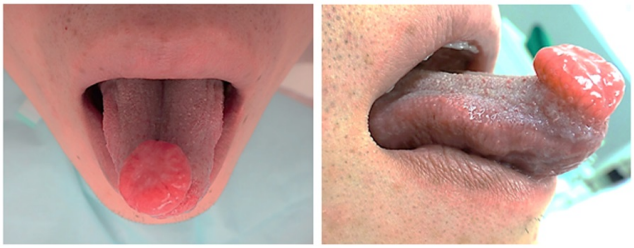

Figure 1: Intraoral preoperative view of lesion.

On further diagnosis, the magnetic resonance imaging (MRI) showed a well-defined oval lesion with moderate signal intensity on the T1-weighted image (Figure 2a) and high signal intensity on the T2-weighted image (Figure 2b) inside the mass.

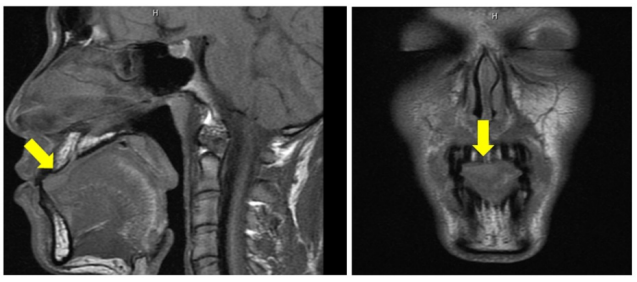

Figure 2a: MRI sequence (a) T1 weighted.

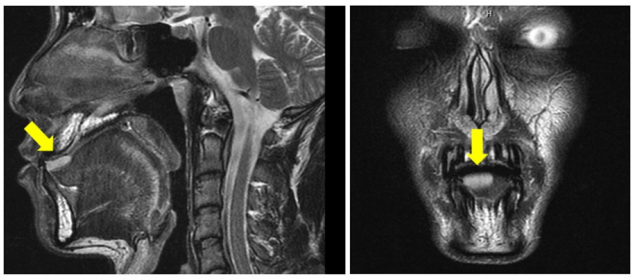

Figure 2b: MRI sequence (b) T2 weighted.

The patient was suspected with a benign tumor, and an excisional biopsy was performed under general anesthesia for further confirmation. The patient's tongue was operated, and the tumor was removed, which was relatively easy due to the lack of adhesions between the tumor and muscle layer. During the post-operative analysis, the oval shaped surgical specimen was encapsulated by relatively dense fibrous tissue (Figure 3).

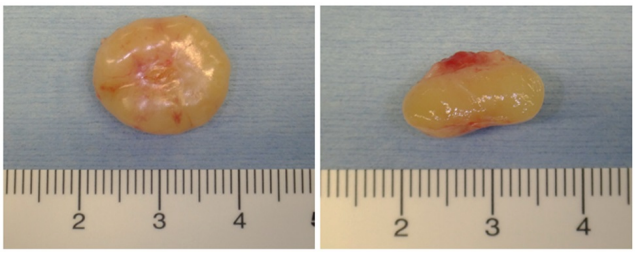

Figure 3: Surgical specimen.

The histopathological examination of the section stained with hematoxylin and eosin showed the spindle-shaped tumor cells arranged around dilated vascular spaces in a storiform or fascicular pattern (Figure 4). No mitotic activity, necrosis, or nuclear pleomorphism was noticed in the tumor. Immunohistochemical (IHC) studies were found to be positive for CD 34, Bcl-2, CD 99 (Figures 5a, 5b and 5c). On the basis of these features, the final diagnosis was confirmed as SFT. The patient's condition was well and free of disease at 12 months after the surgery.

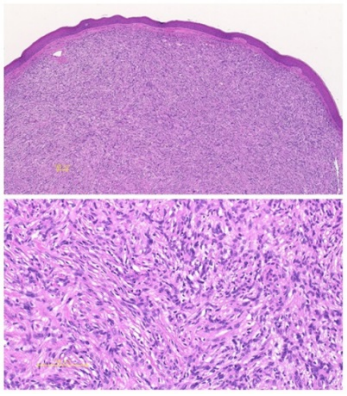

Figure 4: Histopathologic section (hematoxylin and eosin stain).

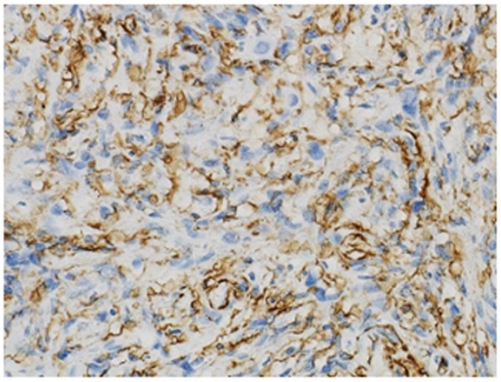

Figure 5a: Immunohistochemical staining with (a) CD34.

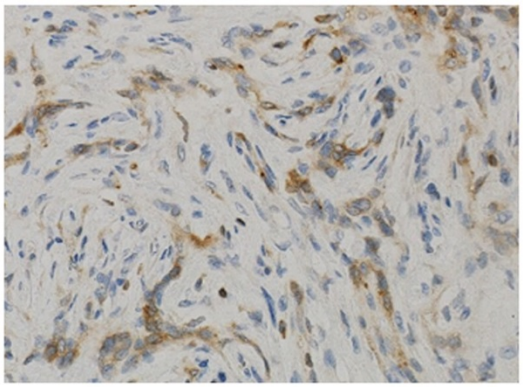

Figure 5b: Immunohistochemical staining with (b) CD99.

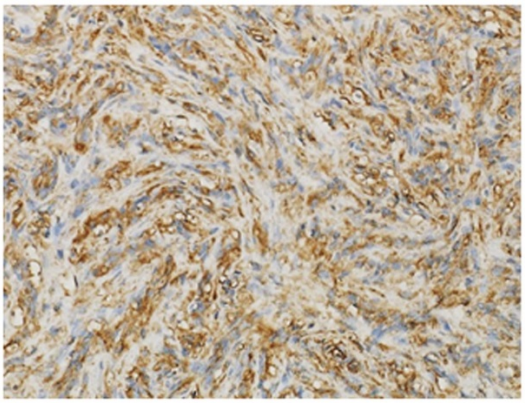

Figure 5c: Immunohistochemical staining with (c) Bcl-2.

Discussion

SFT is a rare neoplasm that mostly occurs as a pleural or serosal tumor. Classically, SFT was described to originate from the spindle-shaped cells of the pleura, but recently, it has been reported in a wide variety of extrapleural sites such as liver, adrenal glands, and skin, and less commonly in the head and neck region [2-8].

The definite diagnosis of SFT depends solely on IHC. Histopathologically, SFT is composed of spindle-shaped cells arranged in a randomly and nonspecific pattern with varying vascularity, which is described as "patternless pattern" [4-8]. This pattern is in accordance with the present case study, where we reported the presence of the spindle-shaped tumor cells arranged around dilated vascular spaces in a storiform or fascicular pattern (Figure 4).

The most consistent and reliable immunohistochemical marker for SFT is CD34, and both pleural and extrapleural SFT typically express CD34 and vimentin [2-8]. However, vimentin is considered to be a non-specific marker because it is expressed by many mesothelial and epithelial neoplasms [9]. Furthermore, the positivity of CD34 is observed in a variety of soft tissue tumors, such as neurofibromatosis, neurilemmoma, angioleiomyoma, and hemangiopericytoma (HPC) [10]. Moreover, SFT often manifests CD99 and Bcl-2 positivity, and the immunoreactivity for these markers is helpful in confirming the

diagnosis [4-8]. In the present study, the mitotic activity, necrosis, or nuclear pleomorphism was not noticed in the tumor, and immunohistochemical studies were found to be positive for CD 34, Bcl-2, CD 99 (Figures 5a, 5b and 5c). This analysis further narrates the findings of the previous research works.

SFT is usually regarded as a benign tumor, and its prognosis is accurate with complete resection. But, it may become rarely malignant and metastasize [4-7,11]. Therefore, long-term follow-up of the patients with SFT is highly recommended. However, this is not applicable to this present case study (Figure 3).

There are some histological features that are considered to be typical for malignant SFTs. Vallat-Decouvelaere et al. mentioned in his research that clinicopathologic features like nuclear atypia increased cellularity, necrosis, and mitotic activity of more than 4 mitoses/10 HPF are associated with aggressive clinical behavior [11]. Although the pathological malignancy was not observed and the postoperative course was uneventful in our case, a periodic follow-up is needed.

Conclusion

In conclusion, we reported an SFT of tongue tip in a 28-year-old male patient. We also concluded that immunoreactivity for CD34, bcl-2, and CD-99 is helpful in confirming the diagnosis. Though regarded as benign, SFTs may rarely become malignant and metastasize. Therefore, a long-term follow-up of the patients with SFT is necessary after complete excision regardless of the histological grade. Moreover, we suggest that SFT may be considered as a differential diagnosis of soft tissue tumors in the oral cavity.

Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

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