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Case Report

Spindle cell lipoma of the floor of mouth ☆

Yuta Yanai, DDS, PhD^{a,*}, Yasutaka Kubota, DDS, PhD^b, Tomoko Kitsuki, DDS, PhD^a,
Risa Yanai, DDS, PhD^a, Tomohiro Yamada, DDS, PhD^a

^a Section of Oral and Maxillofacial Surgery, Division of Maxillofacial Diagnostic and Surgical Science, Faculty of Dental Sciences, Kyushu University, 3-1-1, Maidashi, Higashi-ku, Fukuoka, Japan

^b Department of Oral and Maxillofacial Surgery, Sasebo Kyosai Hospital, 10-17, Shimanji-cho, Sasebo, Nagasaki, Japan

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ABSTRACT

The patient was a man in his 60s with the gradually increasing mass in his mouth. A well-defined, elastic soft mass with a major diameter of 60 mm was found on the right floor of mouth. The magnetic resonance imaging findings showed a well-defined mass with high signal on both T1 and T2-weighted image in the right sublingual space. The mass was slightly heterogeneous inside and had a septum-like appearance. The tumor was resected with care not to damage the capsule. Histopathological findings showed mature adipocytes, spindle-shaped cells, and collagenous components. Spindle cells were CD34-positive. The tumor was diagnosed as spindle cell lipoma. The patient was followed up for 6 months with no recurrence. Spindle cell lipoma is a rare entity and this is the largest case in the oral cavity. Because there are wide variety of adipocytic tumors, the careful examination of their imaging and histopathological findings is essential.

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Introduction

Spindle cell lipoma (SCL) is a benign adipocytic tumor that consists of mature adipocytes, bland spindle cells, and “rope-like” collagen in various proportions [1]. SCL and pleomorphic lipoma (PL) are classified as a single neoplasm, SCL/PLs [1]. SCL/PLs account for approximately 1.5% of all adipocytic tumors, and predominantly occur subcutaneous from the posterior neck to the back and shoulders, and rarely in the oral cavity [1,2]. Because SCL/PLs exhibit varying imaging and

histopathological findings [1–4], it could be difficult to distinguish from other tumors. We report a rare case of SCL in the floor of mouth.

Case report

A man in his 60s presented to our department due to the gradually increasing mass in his mouth. A well-defined, elastic soft mass with a major diameter of approximately 60 mm was

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* Corresponding author.

E-mail address: yanai@dent.kyushu-u.ac.jp (Y. Yanai).

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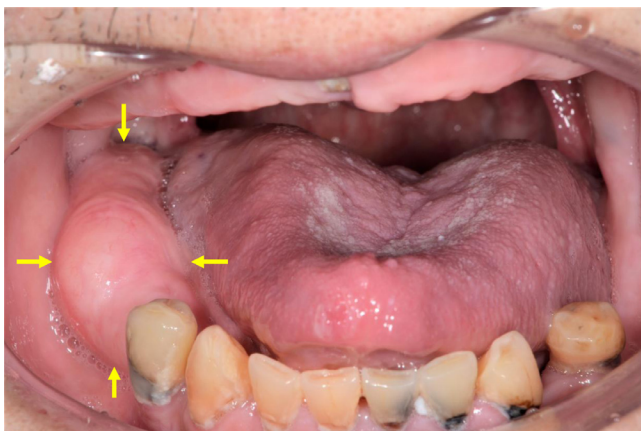


Fig. 1 – Intraoral findings. A well-defined mass (arrow) with a major axis of approximately 60 mm originating from the right floor of mouth.

found on the right floor of mouth (Fig. 1). The covering mucosa was normal in appearance. He was unable to use dentures and had difficulty in chewing due to the mass. He had a history of diabetes and Parkinson's disease.

Magnetic resonance imaging (MRI) findings showed a slightly heterogeneous mass with a high signal on both T1 and T2-weighted image in the right sublingual space (Figs. 2 A and B). The mass had a septum-like appearance. No infiltration into the surrounding tissues was observed.

The tumor was resected under general anesthesia. The mass had a thin capsule and did not adhere to the mucosa or muscles. The size of the excised tumor was 63 × 35 × 15 mm (Fig. 3). Histopathological findings showed proliferation of mature adipocytes, and spindle-shaped cells against the background of collagenous components. Spindle-shaped cells were shown to be CD34-positive by immunohistochemical staining. The Ki-67 Labeling index (MIB-1 index) was lower than 1%. Adipocytes were negative for MDM2 and CDK4. Based on these findings, the tumor was diagnosed as SCL (Figs. 4 A and B).

The patient was followed up for 6 months postoperatively with no recurrence.

Discussion

SCL/PLs represent the morphologic spectrum of a single neoplasm [1]. SCL/PLs predominantly occur in men aged 45–60 years [1]. Approximately 80% of cases occur in the posterior neck and back and shoulders, while the remaining 20% of cases occur subcutaneously in the face, head, limbs and trunk, and submucosally in the oral cavity [1–4]. According to Furlong et al. [5], the lips (29%) and buccal mucosa (27%) are the major sites of occurrence of oral SCL (n = 41), while only 1 case (2%) was on the floor of mouth. SCL/PLs are slowly growing, painless masses that do not adhere to the skin or mucous membrane in many cases [1]. Subcutaneous lesions can grow up to 50 mm or more, but those in the oral cavity are often smaller

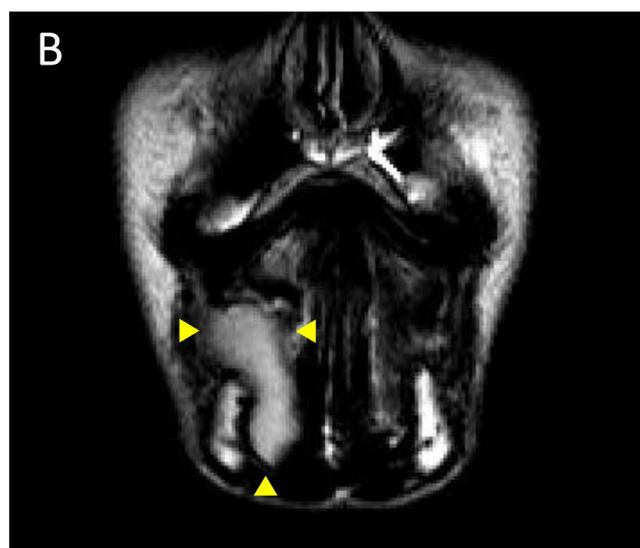
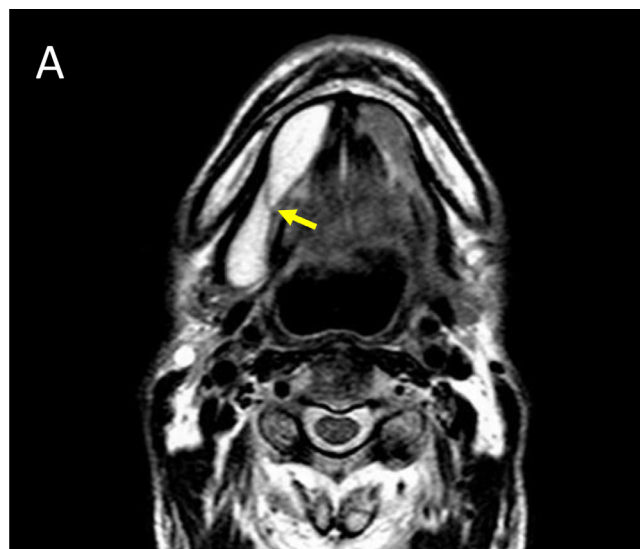


Fig. 2 – MRI findings. (A) T1-weighted image, horizontal plane. High signal area of the right sublingual space. The inside appears heterogeneous, and a septum-like appearance is observed (arrow). (B) T2-weighted image, frontal plane. The high signal area extends to the alveolar process of right mandible (arrow head). MRI, Magnetic resonance imaging.

[1,6]. Present case with a major diameter of 65 mm was the largest SCL in the oral region encountered by us [3,5,7].

Differential diagnosis by imaging findings between SCL/PLs and other adipocytic tumors may be challenging. Due to the various proportions of adipose and other tissues, MRI findings of SCL/PLs may exhibit heterogeneity inside the tumor [4,8]. Nonadipose tissues in SCL/PLs often appear similar signal intensity to muscle on T1-weighted images and may show high signals on T2-weighted images because they contain myxoid and vascular components [4,8]. Typically, conventional lipomas have clear boundary and exhibit a similar signal intensity to that of subcutaneous fat, and are homogeneous inside

Table 1 – Comparison of MRI findings in Lipoma, SCL/PLs, and liposarcoma.

	Internal uniformity	Fat content	Tumor boundary	Contrast of tumor boundary	Peripheral infiltration	Septum-like appearance	Nodular nonfat tissue
Lipoma	Homogeneous	High	Regular	No	No	No	No
SCL/PLs	Heterogeneous	Varies by case	Regular	No/Low	No/Yes	Yes	No
Liposarcoma	Heterogeneous	Varies by case	Irregular	High	Yes	Yes	Yes

MRI, Magnetic resonance imaging; PLs, pleomorphic lipomas; SCL, Spindle cell lipoma.

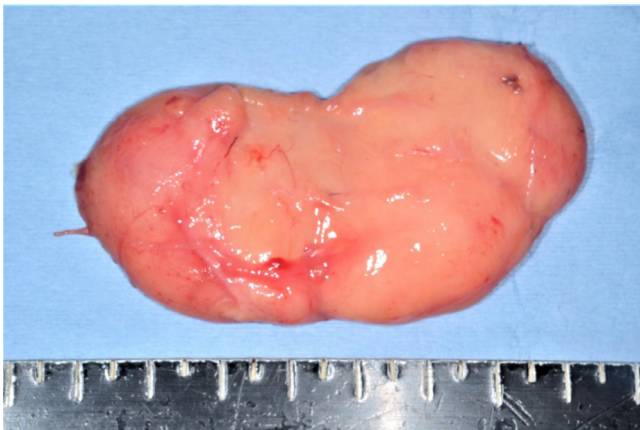


Fig. 3 – Findings of excised mass. Findings show a capsular mass measuring 63 x 35 x 15 mm.

[1,3,4]. Low-fat SCL/PLs may show similar findings to neurogenic or salivary gland tumors [8,9]. Additionally, it should be considered the possibility of liposarcoma or atypical lipomatous tumor (ALT), because some cases with invasion to surrounding tissues have been reported [4,6,10,11]. Liposarcoma has more irregular boundaries than SCL/PLs and is characterized by multiple thick septum, nodular nonfat tissue, and strong contrast-enhanced tumor boundaries and septum by enhanced MRI (Table 1) [4,8]. Our case showed a high signal on both T1 and T2-weighted image, and a slightly heterogeneous mass with a septum-like appearance, suggesting that there were some nonadipose tissues such as fibrous stroma and/or vascular components. Because no infiltration into the surrounding tissue was observed, clinical diagnosis was considered fibrolipoma or possibility of ALT. We performed the tumor resection with care not to damage the capsule.

Histopathologically, CD34-positive spindle cells are a distinguishing factor from other adipocytic tumors, and it is important to differentiate from liposarcoma or ALT [1–3,6,10–12]. The differences in size of adipocytes, nuclear atypia, and mitotic figures are findings suggestive of liposarcoma, and immunohistochemical staining is often positive for MDM2 and CDK4 [1–3,6,10,12]. In our case, the MIB-1 index was very low and no cell atypia or mitosis was observed. In addition, we confirmed that both MDM2 and CDK4 were negative, indicating that the tumor was not malignant. SCL/PLs are often consisted of both sections with dense adipocytes and sections with spindle cells and collagen fibers even in a single specimen. Therefore, differentiation from other adipocytic tumors

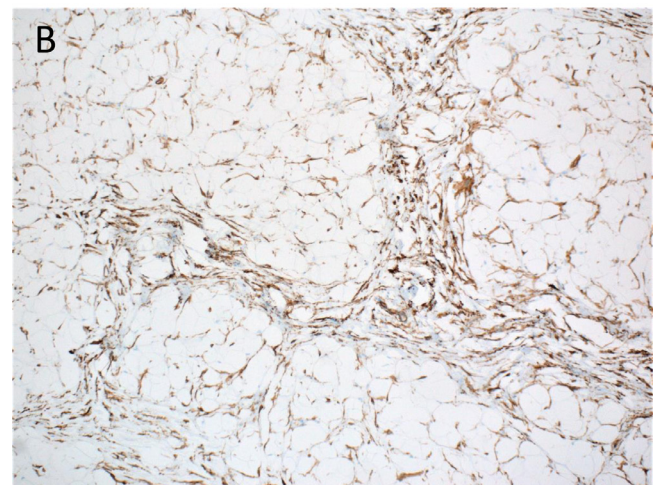
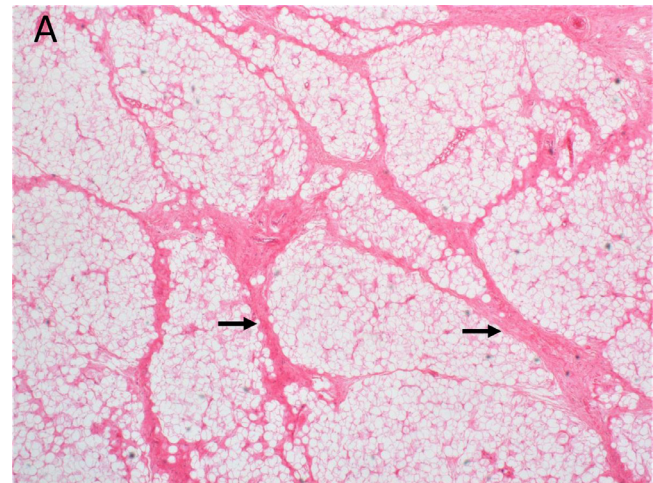


Fig. 4 – Histopathological findings. (A) H-E staining with weak enlargement (x40). Mature adipocytes proliferation and spindle-shaped cells against the background of “rope-like” collagen fibers (arrow) are observed. (B) Immunohistochemical staining of CD34 with strong enlargement (x100). Spindle-shaped cells are CD34 positive.

should be carefully evaluated using the entire excised specimen.

SCL/PLs are encapsulated and rarely infiltrate to surrounding tissue, and the prognosis is good with almost no recurrence [1–3,5,6,11]. However, in atypical SCL/PLs, recurrence is estimated to occur in 10%-15% of cases [1,12]. It is recom-

mended to extract a tumor without damaging the capsule because there may be cell atypia in any adipocytic tumors. Because there are wide variety of adipocytic tumors, the careful examination of their imaging and histopathological findings is essential to distinguish the type of tumor.

Conclusion

This is a rare case of largest SCL in the oral cavity. While differential diagnosis by imaging may be challenging, the morphology and intense enhancement of the tumor boundaries are the key to distinguish from liposarcoma. The careful analysis of imaging and histopathological findings is recommended for proper diagnosis, because there is wide variety of adipocytic tumors.

Patient consent

Informed consent for publication of this case was obtained from the patient.

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