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Case Report

Atypical Lipomatous Tumor of the Tongue: Report of a Case

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The term atypical lipomatous tumor (ALT) is synonymous with well-differentiated liposarcoma (WDL). This tumor occurs very rarely in the tongue. Thus, it is difficult to predict its prognosis. Although recurrence of ALT/WDL is thought to be unlikely after complete excision, long-term follow-up is necessary when considering the pathologic conditions of this tumor at other sites. Here, we report a case of an ALT of the tongue, with a review of the literature. A 68-year-old man was referred to our hospital because of a tumor on the left side of his tongue. Upon palpation, the tumor was 12 mm in diameter, circumscribed, elastic and hard, well demarcated, movable, and painless. We diagnosed the lesion as a lipoma and extirpated the tumor under local anesthesia. Because the specimen was histopathologically diagnosed as an ALT, as a precaution, we excised an additional 5 mm from the area surrounding the original tumor under general anesthesia. Three years after the operation, the tongue demonstrated good healing without paresthesia or dysfunction, and to date there has been no evidence of recurrence.

Key words: atypical lipomatous tumor, well-differentiated liposarcoma, tongue

The term atypical lipomatous tumor (ALT) is often used interchangeably with atypical lipoma and well-differentiated liposarcoma (WDL) because these lesions are histologically indistinguishable. Use of the ALT designation in place of "sarcoma" has increased in recent years, because ALTs do not metastasize if dedifferentiation does not occur. This new classification term is somewhat controversial, but in 2002 the World Health Organization (WHO) adopted the terms atypical lipoma and atypical lipomatous tumor as valid terminology, and they are accepted and used by many investigators. ALTs of the tongue are very rare [1–3]. Only 3 cases of ALT/WDL of the tongue have been reported in Japan. This report details a case of ALT of the tongue and presents a review of the literature.

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Case Report

A 68-year-old man presented to the Department of Oral and Maxillofacial Surgery at Kagawa Rosai Hospital. Approximately one year prior, he noticed a mass on the left lateral border of the tongue. The mass was not painful, and its size did not change during the year between discovery and presentation at our facility. His past medical history included hypertension, gastric ulcer, and gout. His family history was non-contributory.

At the initial examination, the ovoid mass on the left lateral border of the tongue had a diameter of 12 mm. When it was palpated, it was elastic and hard, movable, well demarcated, and painless. The mucous membrane of that site was pale yellow, and there was neither spontaneous pain nor tenderness (Fig. 1). There was symmetry in the patient's facial appearance, and there was no swelling or tenderness of the regional lymph nodes.

The clinical diagnosis was lipoma of the left lateral border of the tongue. The mass was excised en bloc with surface mucosa under local anesthesia. There was no adhesion between the mass and the surrounding tissue. The mass was well demarcated and was easily separated from the surrounding tissue. The excised specimen was determined to be an ALT by histopathologic diagnosis. Thus, Ga-scintigraphy was performed to examine whether there were any lesions in other organs, but none were found. There was also no abnormality in the regional lymph nodes on MRI. Additional resection was performed under general anesthesia to remove a safety margin of approximately 5 mm from the site where the tumor was present. Histopathologic findings showed no neoplastic cells at the margin of the excised specimen. It has

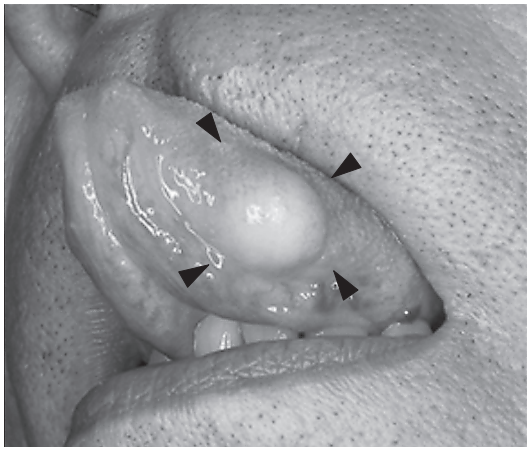

Fig. 1 Intraoral photograph at the initial examination. The 12 mm mass was observed on the left lateral border of the tongue (arrow). It was pale yellow, elastic hard, painless, and well demarcated.

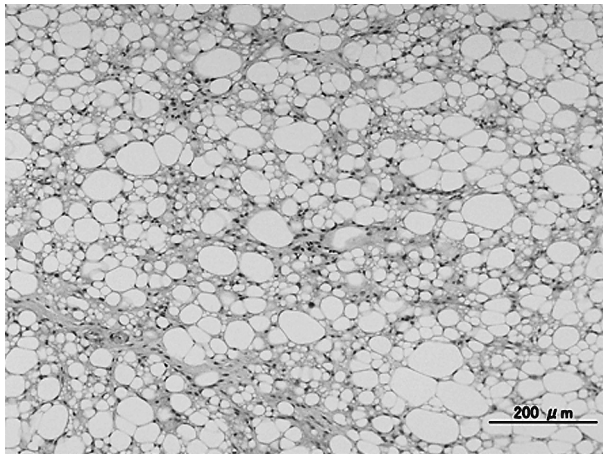

Fig. 3 Histopathologic findings (H-E staining, bar=200 μm). The tumor was composed of mature adipocytes and fibrous septa. The mature adipocytes varied in size.

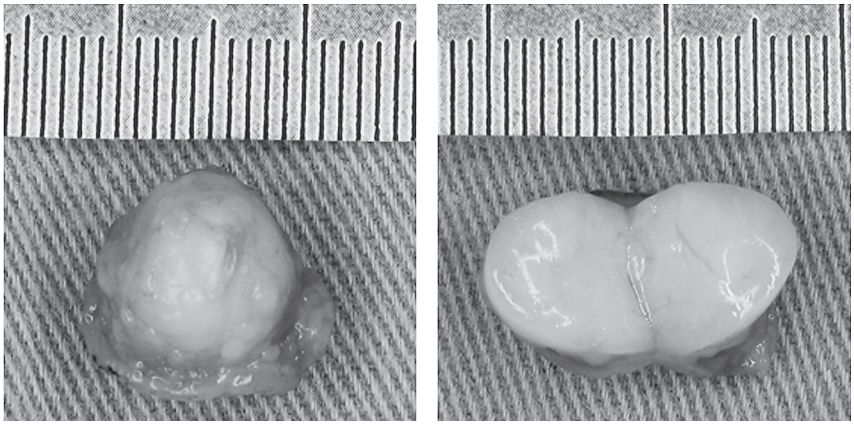

Fig. 2 Findings of the excised specimen. The 13×11×10 mm mass was covered with a fibrous capsule, and the cross section showed a pale yellow solid tumor.
Left, Excised specimen; Right, Cross section of the excised specimen.

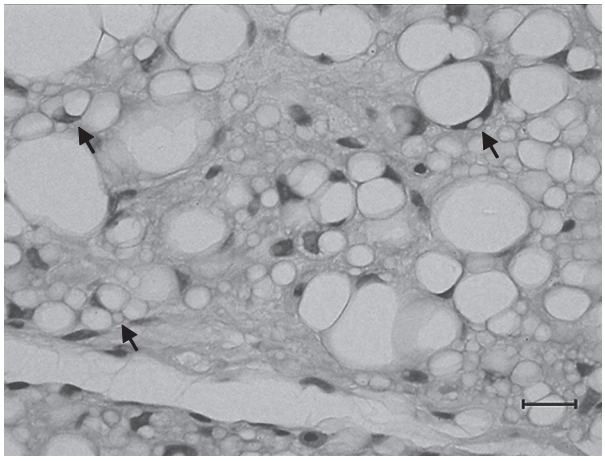

Fig. 4 Histopathologic findings (H-E staining, bar=40 μm). The presence of lipoblast-like cells with single or multiple vacuolated nuclei.

been 3 years since the surgery, and there has been no paresthesia or dysfunction of the tongue. Healing was uneventful, and recurrence has not been observed. The excised specimen was a 13×11×10 mm mass covered with a fibrous capsule. The cross section showed a pale yellow solid tumor (Fig. 2). In H-E staining of the resected soft tissue, the tumor had fibrous septa and proliferation of mature adipocytes of varying sizes (Fig. 3). It also contained stromal cells with hyperchromatic nuclei and lipoblast-like cells with single or multiple vacuolated nuclei (Fig. 4). The histopathologic diagnosis was ALT.

Discussion

The prognosis of WDL has historically been good if it is a subcutaneous WDL or an intramuscular WDL of the limbs located within superficial tissues. Thus, Evans *et al.* proposed the term atypical lipoma for these lesions in 1979 [4]. Furthermore, ALTs do not metastasize if dedifferentiation does not occur [5]. Therefore, Evans proposed the term ALT for these lesions, including WDL, in 1988 [5]. In 2002, ALT was established as being synonymous with WDL in the WHO classification of soft tissue tumors [6].

The classification of liposarcoma includes the category of ALTs/WDLs. Liposarcoma occurs at a relatively high incidence among malignant soft tissue tumors. The main predilection sites for liposarcoma are the thigh, gluteal region, and retroperitoneum [6]. The occurrence of liposarcoma in the head and neck region is very rare. According to the report of Tanaka *et al.* [7], liposarcoma in the head and neck region accounted for approximately 4% of all liposarcoma cases. DeWitt *et al.* [2] stated that in approximately 90 liposarcoma cases of the oral region, 38% involved the buccal mucosa, 33% involved the tongue, 7% involved the palate, and 7% involved the floor of the mouth. ALTs/WDLs account for 40%–45% of all liposarcoma cases [6]. ALTs/WDLs are seen mostly in middle-aged individuals, with a peak incidence in the 6th decade, and they are very rare in children. There is no predilection for one gender over the other [6]. ALTs/WDLs occur mostly in the deep tissues of the limbs, followed by the retroperitoneum, paratestis, and mediastinum. Although they can occur subcutaneously, these lesions occur very rarely on the skin [6].

According to our extensive search, 33 cases of ALT/WDL of the tongue were reported worldwide, including our case, between 1976 and 2008 [1–3, 8–24]. In Japan, there were 3 cases, including our case (Table 1) [1, 13]. The patient age ranged from 37 to 86 years, and the mean age was 63 years. There was clearly a higher number of males (23 males) than females (10 females). The tumors were 3.5 cm or less in diameter, with a mean diameter of approximately 1.5 cm. There were 9 cases of ALT and 24 cases of WDL according to the histological diagnosis. Although there was no clear histological difference between ALT and WDL, some clinicians believe that if the lesion is superficial, it should be classified as ALT, and if it is deep, it should be classified as WDL [2, 3, 19].

Clinical findings of the tongue indicate that lesions requiring differential diagnosis are slow-growing lesions, such as lipoma, lymphoepithelial cyst, and neurilemmoma. ALTs/WDLs have very similar findings with the aforementioned lesions, and thus, differentiation among them is difficult. When liposarcoma is compared with lipoma, liposarcoma tends to be harder, to be more elastic, and to adhere more to the surrounding tissue. However, differential diagnosis is difficult based only on clinical findings. Therefore, biopsy is necessary for differential diagnosis [1–3].

Histopathologically, ALTs/WDLs are formed entirely or in part from the proliferation of relatively

mature adipocytes. The size of the adipocytes varies greatly compared to those of a lipoma [6]. ALTs/WDLs consist of a mixture of mature adipocytes and fibrous connective tissues. In localized regions, nuclei of adipocytes are densely stained and atypical multinucleated stromal cells are frequently observed. There are also lipoblasts with vacuolar, multinucleation, or densely stained nuclei.

In the present case, the lesion was located in the superficial area of the tongue. Histopathologically, the whole tumor was composed of a proliferation of mature adipocytic cells with fibrous septa. A significant variation in cell size was easily recognized. A presence of stromal cells with hyperchromatic nuclei and lipoblast cells with vacuolated nuclei were observed, which are usually identified in a part of lipoma-type ALTs as a characteristic image. All these findings were considered to be consistent with ALT.

The treatment of this tumor involves surgical resection that includes a safety margin. If no neoplastic cells are in the margin of the resected lesion, then recurrence is considered unlikely [6]. In our literature search, we found 3 cases of recurrence among 32 cases of ALT/WDL of the tongue (Table 1). The recurrence rate was 9%, and recurrence was observed in 2 cases in which local excision and 1 case in which debulking was performed. The recurrence rates were approximately 40% and 90% in the limbs and retro-

Table 1 Reported cases of ALT/WDL of the tongue (1976–2008)

	Author (yr)	Age/ Gender	Clinical manifestation			Histopathologic Diagnosis	Treatment Modality	Follow-up
			Appearance	Size (cm)	Duration			
1	Larson et al. (1976)[8]	42/F	NM	1.5×1.5	NM	WDL	HG	N/A
2	Wescott and Correll (1984)[9]	61/M	NM	3.5×3×2	NM	WDL	LE	Recurrence
3	Saddik et al. (1996)[10]	76/M	Multi-nodular	2.5	Longstanding	WDL	LE	23 years 5 months Reccurence
4	Kacker and Taskin (1996)[11]	78/M	Nodular	4	6 years	ALT	LE	N/A
5	Nelson et al. (1998)[13]	37/M	NM	3×3×3	>10 years	WDL	WE	N/A
6	Noguchi et al. (1998)[14]	70/M	Nodular	2×1×0.8	2 years	WDL	WE	NED, 8 months
7	Kasper et al. (2000)[12]	71/M	NM	NM	NM	WDL	WE	NED, 2 years
8	Gagari et al. (2000)[15]	73/M	Nodular	2×1×1	NM	WDL	LE	N/A
9	Orita et al. (2000)[16]	70/M	Nodular	1×3.5	1 month	WDL	HG	NED, 6 months
10	Moore et al. (2001)[17]	43/M	NM	0.8	NM	ALT	WE	NED, 10 months
11	Miya et al. (2002)[1]	79/M	NM	3.5×2.5×2	6 months	WDL	WE	NED, 1 year
12	Nascimento et al. (2002)[18]	36/M	NM	0.6	1 year	WDL	LE	NED, 9 years
13	Nascimento et al. (2002)[18]	50/M	NM	1.2	NM	WDL	LE	NED, 4 years
14	Nascimento et al. (2002)[18]	76/M	NM	1	3 months	WDL	LE	NED, 1 year
15	Nascimento et al. (2002)[18]	47/F	NM	2.2	NM	WDL	LE	NED, 1.5 years
16	Nascimento et al. (2002)[18]	74/M	Multi-nodular, bilateral	3 (each side)	Months	WDL	D	1 year 3 months Recurrence
17	Nascimento et al. (2002)[18]	77/F	NM	0.6	NM	WDL	LE	NED, 2 years
18	Nascimento et al. (2002)[18]	43/M	NM	1	5 years	WDL	LE	NED, 1 year
19	Nascimento et al. (2002)[18]	72/M	NM	0.7	NM	WDL	LE	NED, 2 years
20	Nascimento et al. (2002)[18]	80/M	NM	1	1 year	WDL	LE	NED, 2 years
21	Nascimento et al. (2002)[18]	53/M	NM	0.8	months	WDL	LE	NED, 2 years
22	Fanburg-Smith et al. (2002)[19]	43/F	NM	NM	5 years	WDL	WE	NED, 2 years
23	Fanburg-Smith et al. (2002)[19]	64/F	NM	NM	3 years	WDL	WE	NED, 16 years
24	Nunes et al. (2002)[20]	65/M	NM	1×1×0.7	3 years	WDL	WE	NED, 1.5 years
25	Capodiferro (2004)[21]	58/F	Nodular	2.5	6 months	WDL	WE	NED, 2 years
26	Allon et al. (2005)[3]	68/F	Nodular	1	years	ALT	WE	NED, 1 year
27	Allon et al. (2005)[3]	72/M	Nodular	0.7	1 year	ALT	WE	NED, 1 year
28	Allon et al. (2005)[3]	57/M	Nodular	2	1 year	ALT	LE	NED, 1 year
29	Angeles et al. (2005)[22]	86/M	Nodular	0.5	NM	ALT	LE	N/A
30	DeWitt et al. (2008)[2]	62/F	Nodular	0.7	6 months	ALT	LE	NED, 1 year
31	DeWitt et al. (2008)[2]	58/F	Nodular	0.6	1 year	ALT	LE	NED, 1.5 years
32	Sanchez et al. (2008)[24]	36/F	Nodular	0.5	2 years	WDL	LE	NED, 1 year
33	Present study (2009)	68/M	Nodular	1.3×1.1×1	1 year	ALT	WE	NED, 3 year

WE, Wide excision; LE, Local excision; HG, Hemiglossectomy; D, Debulking; WDL, Well-differentiated liposarcoma; ALT, Atypical lipomatous tumor; N/A, no recorded follow-up; NED, No evidence of disease; NM, Not mentioned.

WDL and ALT of the tongue: demographic data, treatment modality, and follow-up information.

peritoneum, respectively [23]. It is rare for ALT/WDL to result in death if it occurs in the limbs or subcutaneously, where complete excision is possible. If a complete resection is difficult, as in the retroperitoneum, the mortality rate is over 80% at 10 to 20 years after onset. The median survival time ranges from 6 to 11 years after onset [6]. There are still few reported cases of ALT/WDL of the tongue. Thus, it is difficult to predict its prognosis. Although recurrence of ALT/WDL is thought to be unlikely after complete excision, long-term follow-up is necessary when considering the pathologic conditions of this tumor at other sites. We plan to follow up our patient carefully.

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