CASE REPORT

A case of spindle cell lipoma arising in the tongue

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Summary

Spindle cell lipoma (SCL) is a distinct histological variant of lipoma, characterized by replacement of mature fat cells by collagen-forming spindle cells. We report a case of SCL that arose in the tongue of a 75-year-old Japanese man. Immunohistochemical analysis revealed that the spindle cells expressed CD34, bcl-2, and vimentin. Interestingly, the tumor cells exhibited strongly positive immunoreactivity for cyclin-dependent kinase 4 (CDK4). Since it has already been reported that CDK4 is not expressed in ordinary lipomas but expressed in almost all cases of liposarcoma, the positive immunoreactivity for CDK4 in the present case suggests that SCL has a character that is distinct from ordinary lipoma.

KEYWORDS
Spindle cell lipoma; Tongue; Oral cavity; CDK4

Introduction

Spindle cell lipoma (SCL) is a benign, characteristically well circumscribed fatty tumor that rarely recurs. SCL arises most often in the shoulder and posterior neck region of adults, but has also been reported in many other locations, including the limbs, face, and anus. It is very rare in the oral cavity, especially the tongue.1–6

Immunostaining for CD34 and bcl-2 is useful for distinguishing SCL from liposarcoma and ordinary lipoma.1,7 Recently, CDK4 has been reported to be expressed in almost all cases of liposarcoma, but not in ordinary lipoma.8,9 On the other hand, it remains to be determined whether CDK4 is expressed in SCL. Here, we report a case of CDK4-expressing SCL that arose in the tongue.

Case report

A 78-year-old Japanese male patient noticed a painless mass at the left edge of the tongue 6 months before visiting our hospital. Intraoral examination revealed a mobile mass 1.0 × 0.8 cm
in size on the left edge of the tongue. The lesion was surgically excised under local anesthesia.

After fixation with 10% neutral buffered formalin, the specimen was embedded in paraffin according to the routine procedure. Subsequently, 5-μm-thick sections were cut and stained with hematoxylin and eosin (HE) and alcian blue. Immunohistochemistry was also performed by the ABC method using antibodies against S-100 protein, α-smooth muscle actin (α-SMA), vimentin, bcl-2, CD34, Ki-67, PCNA and CDK4.

Macroscopically, the resected tumor was found to be located in the lingual muscle, surrounded by multiple tiny islands of fat tissue. The resected mass was elastic-hard, and the cut surface was grayish and lobulated.

Microscopic examination showed that the tumor was composed of a mixture of mature adipocytes and uniform spindle cells in a mucoid matrix with collagen bundles (Fig. 1A). There were no lipoblasts or atypical cells, and mitotic figures were absent (Fig. 1B). The tumor cells were mainly located beneath the mucosa of the tongue. They were diffusely arranged, and some cells were found to have infiltrated into the muscle bundles (Fig. 1C). The mucoid matrix was positively stained with alcian blue (Fig. 1D). Immunohistochemical analysis showed that bcl-2 and vimentin were expressed in both the mature adipocytes and the spindle cells (Fig. 2D,E), whereas α-SMA was not expressed in either cell type (Fig. 2B). CD34 was expressed in the spindle cells but not in the mature adipocytes (Fig. 2C). In contrast, S-100 was positively expressed in the mature adipocytes but not in the spindle cells (Fig. 2A). Furthermore, although we were unable to find any Ki-67-positive cells in the tumor, a small number of PCNA-positive spindle cells were noticed (data not shown). Interestingly, we found that cyclin-dependent kinase 4 (CDK4) was strongly expressed in the nuclei of the spindle cells, whereas it is conclusively negative in the mature adipocytes (Fig. 2F). The immunohistological features of this case are shown in Table 1.

### Discussion

SCL usually arises in the posterior neck, upper back, and shoulder, but rarely in the tongue. Previous reports have indicated that SCL accounts for approximately 1.5% of all lipomatous tumors, and only 11 cases of SCL arising in the tongue have been reported. A histological analysis of 164 cases of lipoma located in the oral cavity showed that

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**Figure 1** Light microscopic observations of SCL (A; HE ×40). Mature adipocytes and spindle cells are admixed with a mucoid matrix and collagen bundles (B; HE ×400). No mitoses are observed (B; HE ×400). Some tumor cells have infiltrated into the muscle bundles (C; HE ×40). The mucoid matrix is positively stained with alcian blue (D; alcian blue ×400).
Figure 2  Immunohistochemical observations of SCL. Mature adipocytes are positively stained with anti-S-100 antibody (A ×400). Both mature adipocytes and spindle cells are negative for SMA (B ×400). Both mature adipocytes and spindle cells are positively stained with anti-bcl-2 antibody (C ×400) and anti-vimentin antibody (E ×400). Only spindle cells, but not mature adipocytes, are positively stained with anti-CD34 antibody (D ×400) and anti-CDK4 antibody (F ×400).

Table 1  Immunohistochemical analysis of lipoma, SCL and liposarcomas

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<th>Ordinary lipoma</th>
<th>SCL</th>
<th>Liposarcoma</th>
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<td></td>
<td>Mature adipocyte</td>
<td>Spindle cell</td>
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<td>S-100</td>
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<td>CDK4</td>
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\(^a\) In some cases of liposarcomas have been reported to express bcl-2 and CD34 with sparsely distribution.
only two cases were SCL, and another study revealed that only 6 out of 76 cases of oral lipoma were SCL.\textsuperscript{3,6} Since lipomas in general are infrequent in the oral region, it is evident that SCL is extremely rare in the tongue.

SCL is a distinct histological variant of lipoma composed of mature lipocytes and uniform spindle cells set in a mucinous and fibrous stroma. Macroscopically, SCL resembles liposarcoma, particularly the myxoid variant of liposarcoma, and atypical lipomatous tumors which are also referred to as well differentiated liposarcomas (ALT/WDLPS).\textsuperscript{1,5,9} In contrast to liposarcomas, however, it has been reported that lipoblasts are usually absent in SCL, and that the tumor cells rarely show mitotic figures. Nevertheless, in a very small number of cases, a few lipoblasts or lipoblast-like cells have been observed in SCL.\textsuperscript{10,11} Intramuscular and poorly circumscribed lipomas should also be differentiated from SCL, although uniform spindle cells are absent in such lipomas.\textsuperscript{7} Immunohistochemically, spindle cells express CD34, bcl-2, and vimentin, but not S-100 (Table 1).\textsuperscript{1,7} Both the spindle cells and mature adipocytes of SCL are reported not to express SMA (Table 1). With respect to proliferative activity of the tumor cells, it has been reported that a proportion of the spindle cells are positive for PCNA but negative for Ki-67, although mature adipocytes are negative for both.\textsuperscript{4} Thus, our present immunohistochemical data are compatible with those of previous reports.\textsuperscript{1,3–5,7,12}

Recently, CDK4, a well-known key regulator of cell cycle progression, was found to be expressed at a high level in liposarcomas.\textsuperscript{8,9} Because it is never expressed in lipomas and normal mature adipocytes (Table 1), the expression level of CDK4 protein appears to be aberrantly up-regulated in liposarcomas. It has also been reported that CDK4 expression is overexpressed in lung cancer and malignant melanoma.\textsuperscript{13,14} Interestingly, the clinical prognoses of cases of lung cancer showing CDK4 overexpression are reportedly poorer than those of CDK4-negative cases, suggesting that aberrant overexpression of CDK4 is associated with the degree of malignancy. In the present case, we found that CDK4 was also overexpressed in the SCL cells. To our knowledge, this is the first report describing the expression of CDK4 in SCL. It remains to be determined whether CDK4 overexpression is, in fact, associated with differences in pathologic nature among SCL, liposarcoma and lipoma. Further analysis of a large number of cases will be required to confirm this possibility.

References


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