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Case Report

Spindle cell lipoma of the tongue: Two case reports

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ABSTRACT

Spindle cell lipoma (SCL) is a rare tumor of soft tissues, and was first described in 1975 by Enzinger and Harvey SCL often occurs in elderly men as a solitary lesion in the posterior neck and back, and less commonly in the oral cavity. SCL of the tongue is relatively uncommon, and only 14 cases have been reported to date. Here, we describe two cases of SCL of the tongue. A 78-year-old male and a 77-year-old female were diagnosed with tongue tumors, and both underwent resection. Histopathological examination of the resected tumors revealed that they consisted of masses of mature fat cells with fibrous bundles accompanied by hyperplasia of spindle cells surrounding the adipose cells. Immunohistochemistry was performed for histological diagnosis. Using immunohistochemical staining, the spindle cells of each tumor were found to be positive for CD34 but negative for S-100. The nuclei of the spindle cells were negative for Rb and STAT6, and CDK4 and MDM2 immunostaining was negative in every tumor cell. In case 2, some spindle cells were positive for p16, but all were completely negative in case 1. Finally, these tumors were diagnosed as SCLs. To make a definitive pathological diagnosis for SCL, a panel of immunohistochemical markers incorporating CD34, S-100, STAT6, Rb, p16, MDM2 and CDK4 is required to avoid confusion with histologic mimics.

1. Introduction

Spindle cell lipoma (SCL) is a benign lipomatous tumor that predominantly occurs in the posterior neck and shoulder area. In the oral cavity it is a relatively uncommon neoplasm, particularly in the tongue, which is relatively devoid of fat cells. SCL is a soft tissue tumor that was first described in 1975 by Enzinger and Harvey [1]. SCL is a very rare disease, accounting for 1.5% of all fatty tumors, often occurring in elderly men with a median age of over 55 years as a solitary lesion in the posterior neck and back, but less commonly also involving the oral cavity. SCL has a superficial location and a well-defined mass [2]. Histologically, SCL consists of bland spindle cells in a background of thick collagen fibers and variable number of adipocytes. Although SCL has previously been distinguished from pleomorphic lipomas characterized by the appearance of floret-like multinucleated giant cells, given its clear-cut overlapping clinical, histological, immunohistochemical, and cytogenetic features, these lesions are best considered as one entity [3]. They are classified into the same adipose tumors categories according to the WHO classification of 2013, and are now considered to be the same tumors.

Cytogenetic analysis of SCL/pleomorphic lipoma cases has demonstrated consistent deletions of the long arm of chromosome 13, often in combination with loss of the long arm of chromosome 16. The tumor suppressor gene *RB1*, which encodes the retinoblastoma (Rb) protein, is located at 13q14 within a minimally deleted region in SCL. Recently, the majority of SCL and pleomorphic lipomas were demonstrated to show a loss of nuclear Rb expression by immunohistochemistry, while Rb expression is intact in histologic mimics, such as lipoma, solitary fibrous tumor, and myxoid liposarcoma. These suggest the diagnostic utility of immunohistochemistry for Rb in distinguishing these histologically related tumors [4].

Oral SCL on the floor of the mouth cavity was first reported in 1984 [5]. Although a review of the current literature reveals that more than 50 cases of intraoral SCLs have been reported, oral SCL is considered as rare a tumor as SCL occurring in other locations than the oral cavity [6]. In a most recent study, however, 34 SCL cases (27%) among 125 lipomatous tumors in the oral region were reported, indicating that oral SCL may be more common than

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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previously estimated [7]. In this report, six of the 34 oral SCL cases (18%) were located in the tongue [7].

In this study, we reported two cases of SCL developing in the tongue that displayed negative results for Rb immunohistochemistry, indicative of Rb deficiency in pathological examination of the resected specimens.

2. Case report

Case 1. A 78-year-old male was aware of a tumor of the tongue, and visited our hospital for medical examination and treatment. The 7 mm × 6 mm painless tumor was located on the right side of the tongue at the time of the initial visit (Fig. 1A). Perception disorder and motility disturbance of the tongue were not observed. The clinical diagnosis was a fibrolipoma of the tongue, and tumor excision was performed. In histopathological examination of the resected tumor, a well-defined tumor was seen beneath the squamous mucosa, and the tumor consisted of proliferating mature fat cells with fibrous bundles accompanied by hyperplasia of spindle cells between the adipose cells (Fig. 1B–D). Atypical features were not observed in the nuclei of the spindle and adipose cells, and multinucleated giant cells were not evident. Using immunohistochemical staining, the spindle cells were identified as positive for CD34 antibody (NU-4AI, 1:200 dilution; Dako, Kyoto, Japan) (Fig. 2A) and positive for BCL-2 antibody (1:10 dilution; Dako), but negative for S-100 antibody (Z311, 1:800 dilution; Dako) (Fig. 2B). The nuclei of the spindle cells were negative for STAT6 (SC-621, 1:20 dilution; Santa Cruz Biotechnology, Santa Cruz, CA) (Fig. 2C), Rb (G3-245, 1:50 dilution; Pharmingen, San Diego, CA) (Fig. 2D), and p16 antibodies (JC8, 1:100 dilution; Santa Cruz Biotechnology). Immunohistochemical staining for CDK4 (DCS-31, 1:100 dilution; Invitrogen, Paisley, UK) and MDM2 (IF2, 1:40 dilution; Oncogene, Darmstadt, Germany) were also negative in the whole tumor cells. Finally, the tumor was diagnosed as an SCL. The patient experienced no tumor recurrence or metastasis within 14 months after surgery.

Case 2. A 77-year-old female was aware of a painless tumor of the tongue. The tumor was 5 mm × 5 mm in size, and was located in the left dorsum of the tongue (Fig. 3A). The clinical diagnosis was a fibroma of the tongue, and the tumor was excised. Histologically, a well-defined tumor was observed beneath the squamous mucosa and the tumor consisted of a mass of mature fat cells with foci of myxoid change (Fig. 3B and C). In the myxomatous lesions of the tumor, hyperplasia of small blood vessels and a mixture of thick collagen fibers and proliferative spindle cells was observed (Fig. 3D). Using immunohistochemical staining, the spindle cells were positive for CD34 (Fig. 4A) and BCL-2 antibody, but negative for S-100 antibody. The nuclei of the spindle cells were negative for STAT6 and Rb staining (Fig. 4B and C). Unexpectedly, a number of the spindle cells were weakly positive for p16, but CDK4 and MDM2 immunostaining was negative in every tumor cell including the p16-positive-spindle cells (Fig. 4D–F). Finally, the tumor was diagnosed as an SCL. The patient experienced no tumor recurrence or metastasis within 22 months after surgery.

3. Discussion

In a recent study of 125 lipomatous tumors that included 34 SCL cases in the oral cavity, six cases were located in the tongue [7], and the most common location for lipomas in the oral region was the buccal mucosa, whereas the common location for SCL was the lip (35%), buccal mucosa (32%) and the tongue (18%) [7]. A review of the literature revealed more than 50 cases of intraoral SCL, but also that SCL of the tongue is relatively uncommon, and cases occurring bilaterally in the margin of the tongue are rarely reported [5,8–16]. Although 17 cases of lipomatous tumor in the maxillofacial region were recorded in the pathological case files at our hospital from 2001 to 2013, SCL was only present in two cases

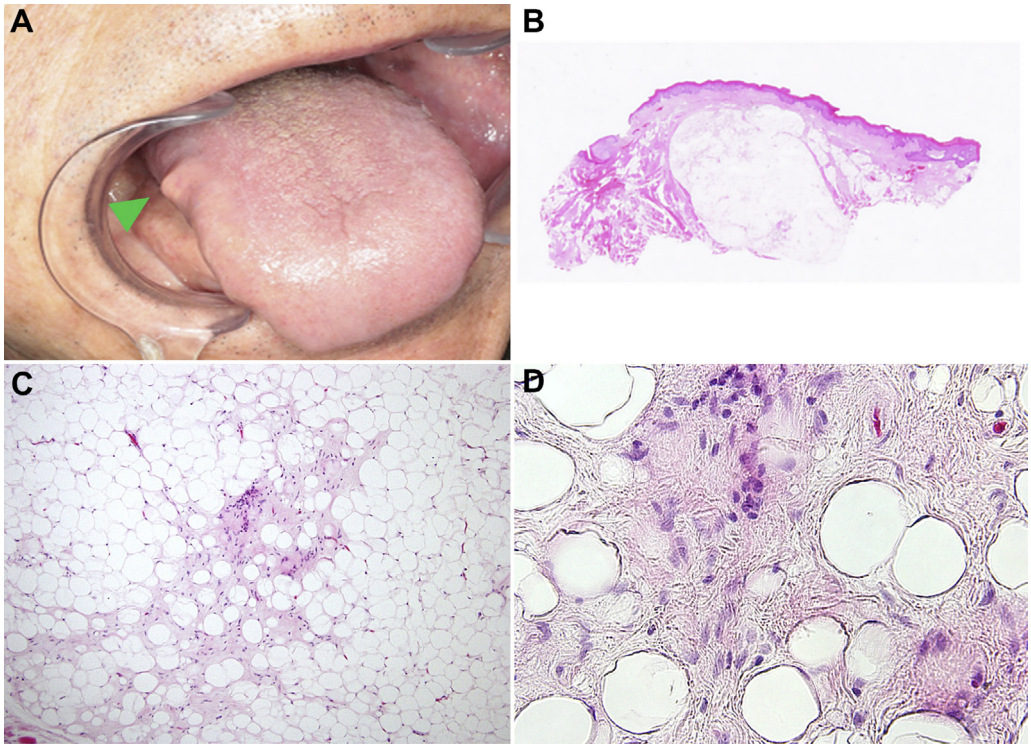

Fig. 1. Clinical appearance of case 1 patient. A submucosal lipomatous nodule (arrow head) was seen in the right side of the tongue (A). At low magnification, the resected tumor was nodular and the border between the tumor and adjacent tissues was well-defined (B). Higher magnification revealed a mass of mature fat cells with fibrous bundles accompanied by spindle cells (C and D).