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Case Report

Primary leiomyosarcoma in the floor of mouth: a case report

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Abstract: Leiomyosarcoma (LMS) is a malignant soft tissue tumor that exhibits smooth muscle differentiation, and its occurrence in the oral cavity is exceedingly rare, especially in the floor of mouth. A 54-year-old male was admitted for evaluation of a painless nodular mass in the left floor of mouth with a 3-month history. With the help of computed tomography (CT) and magnetic resonance imaging (MRI) examination, the patient went on a surgery excision, and by pathological and immunohistochemical findings, a diagnoses of LMS was confirmed. Then the patient went on chemotherapy, and six-month follow up showed no recurrence of cancer. The diagnosis of LMS is difficult to be made, and it is usually confirmed by immunohistochemical stains, such as positive for smooth muscle actin (SMA), vimentin, and Ki-67.

Keywords: Leimyosarcoma, the floor of mouth, immunohistochemistry

Introduction

LMS is a malignant smooth muscle tumor, which is usually seen in the uterus, gastrointestinal tract and retroperitoneal region, reflecting the preponderance of smooth muscle in these locations. In the head and neck region, its occurrence is exceedingly rare [1], because of the paucity of smooth muscle, and in those region this tumor is believed to arise from the tunica media of the blood vessels [2, 3]. Clinically, it is very aggressive, and the prognosis is poor [4]. It is rarely seen in the floor of mouth [5]. We reported thus a case of leiomyosarcoma in the floor of mouth, with the immunohistochemical staining of tumor cells, which is useful for diagnosis.

Case report

A 54-year-old male was admitted for evaluation of a painless nodular mass in the left floor of mouth with a 3-month history. On clinical examination a pedunculated growth in the region of the left floor of mouth was observed (**Figure 1A**). The lesion was soft on the surface and non-tender inside, measuring about 3 cm × 3 cm in dimension, with large basement, and the overlying mucosa was normal without ulceration. Lymph nodes were not palpable in the cervical region. Computed tomography (CT) sections presented abnormal heterogeneous intensity with higher density behind the alveolar bone in the region of the left floor of mouth (**Figure 1B**). On magnetic resonance imaging (MRI) examination, a lesion in the region of right frenal linguae which displayed slightly longer intensity on T1 and T2-weighted image (WI), and slightly hyper intensity on fat suppression T2-weighted image (FS-T2WI) was seen, with an unclear margin, measuring about 1.3 cm × 1.1 cm in diameter, and a diagnosis of hemangioma was suggested (**Figure 1C**, **1D**). There was no regional lymphadenopathy, electrocardiogram and chest X-ray was normal.

Complete excision of the mass and neck dissection were performed. Microscopic examination revealed a malignant mesenchymal tumor that was composed of spindle cell proliferation forming rough bundles and fascicles with interlacing pattern. Scattered abnormal mitotic figures were also present. No tumoral necrosis was observed (**Figure 2A**, **2B**). Immunohistochemical staining showed, the cyto-

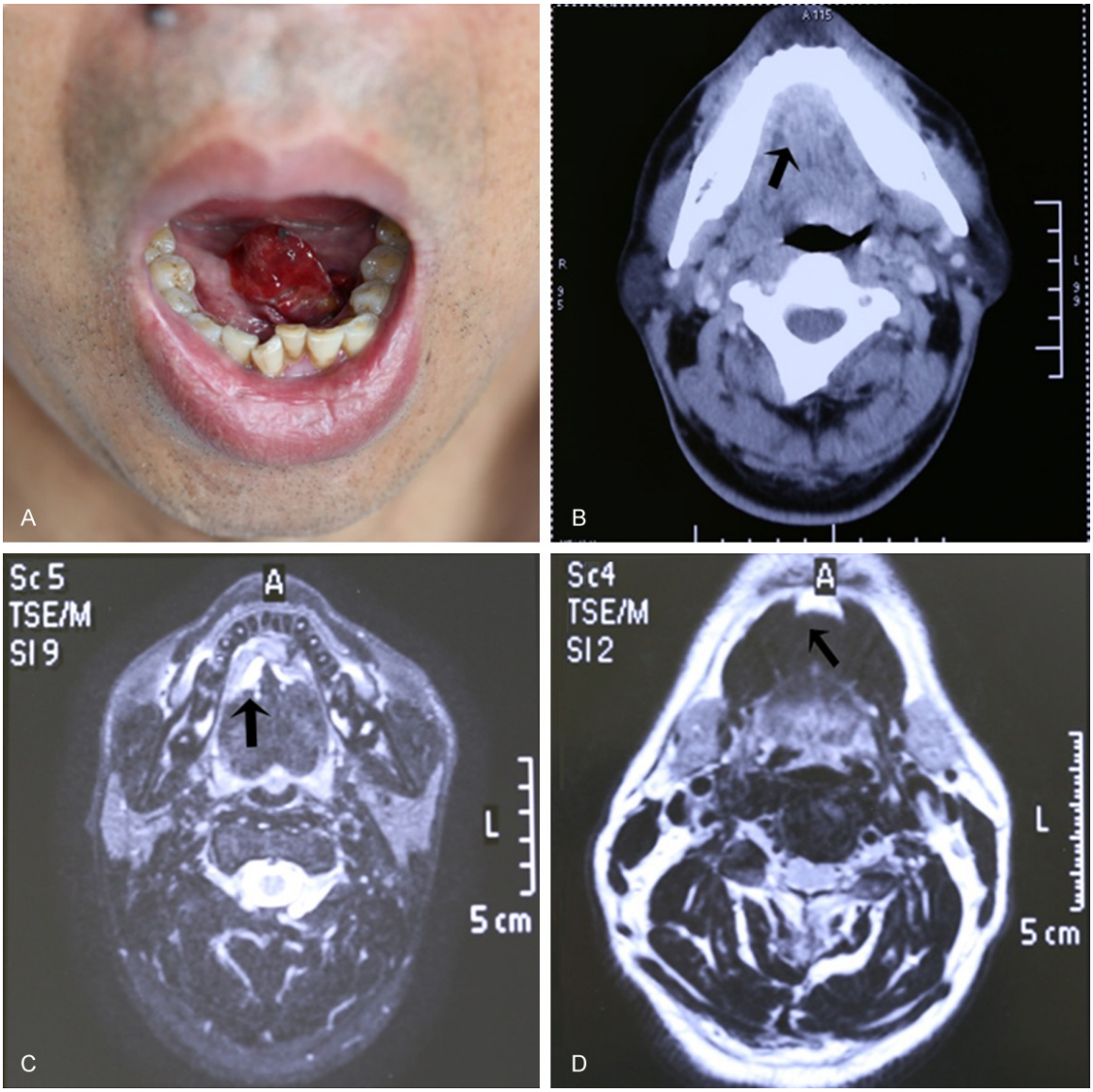

Figure 1. Oral imagination, CT and MRI findings. A. A pedunculated growth in the region of the left floor of mouth was observed. B. Higher density behind the alveolar bone in the region of the left floor of mouth was shown on CT. C, D. On MRI examination, longer intensity on T1 and T2-weighted image was seen.

plasm of tumor cells was positive for smooth muscle actin (SMA) (**Figure 2D**), and high proliferative index displayed by Ki-67 (**Figure 2C**), which was satisfied with the diagnosis of LMS.

Then the patient went on chemotherapy in the local hospital, and six-month follow-up showed no recurrence of tumor.

Discussion

Intraoral LMS account for only 3%-10% of all LMS among the whole body, and the most frequent region in the intraoral locations of these tumors are as follows: buccal mucosa, mandible, gingiva, maxilla, floor of the mouth, tongue, soft palate, hard palate and maxillary sinus [6]. Smooth muscles are paucity in the head and neck region and are mainly found in the walls of blood vessels. So it is believed that in the head and neck region, LMS is most probably arisen from tunica media of the blood vessels [7]. Among all ages, there is no age predilection, and male and female have the same opportunity to be affected [8].

In histological examination, LMS is typically composed of spindle cells with abundant cyto-

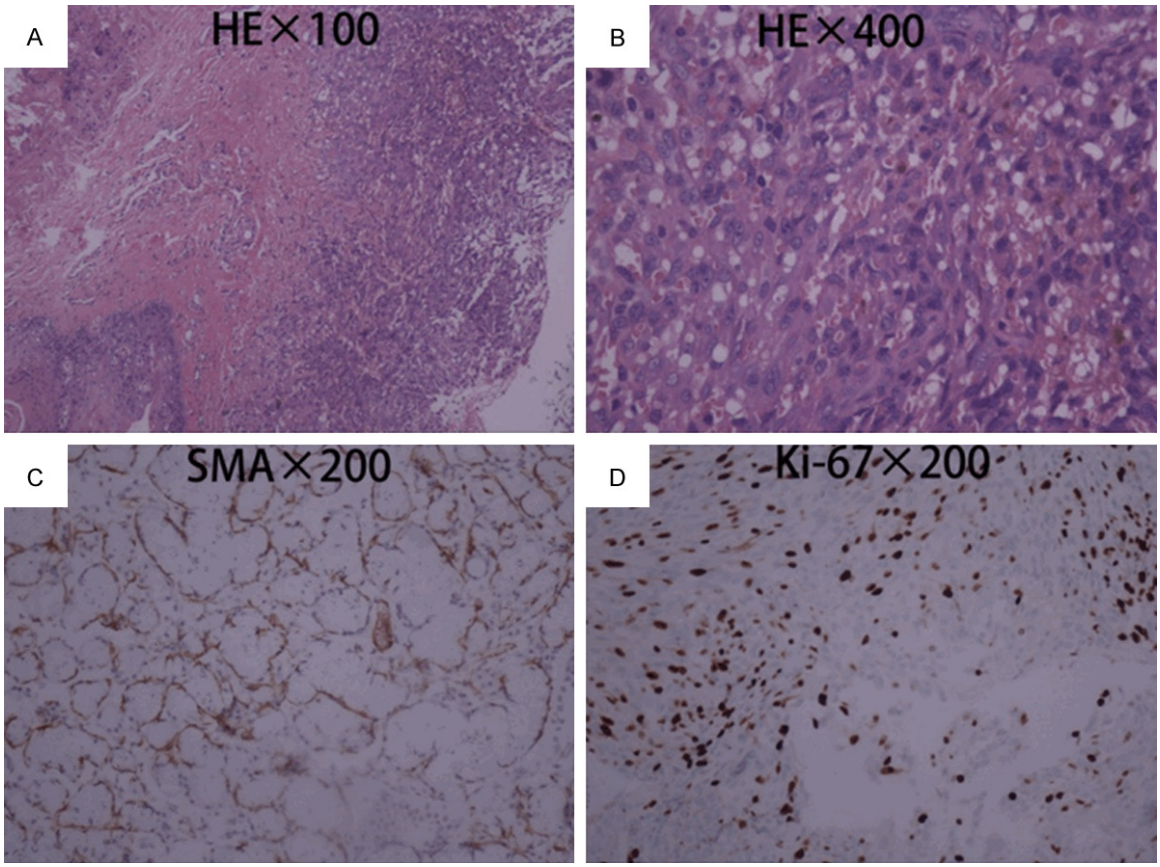

Figure 2. Pathology and immunohistochemistry of the LMS. (A, B) Interlacing fascicles and bundles of spindle cells. (C) Immunohistochemistry showed positive for SMA, and (D) high proliferative index displayed by Ki-67. HE, hematoxylin-eosin stain.

plasm and a centrally placed nucleus. Multinucleated giant cells are common. Among immunohistochemistry, LMS is immune-positive for vimentin, SMA, Ki-67 and muscle specific actin (MSA) and immune-negative for S-100 and cytokeratin [7, 9].

There is a possibility of LMS from other primary region metastasize to the oral cavity, and oral LMS tends to metastasize to cervical nodes, lung and liver, therefore once it is identified as LMS, it is necessary to determine whether the lesion is primary or a metastasis [10]. As a general rule, the likelihood of distant metastasis is related to histological grade and tumor size; the larger and higher grade lesions, the higher risk of metastasis [11].

Nowadays, the most popular treatment on this tumor is extensive surgical resection, with neck dissection for lymph node metastasis. Adjuvant radiotherapy and chemotherapy have also been beneficial for treatment. Overall the prognosis of LMS is usually poor and hence early diagnosis is the key to the whole treatment.

On clinical situation, when met a patient shows as a nodular painless, well circumscribed mass and adherent firmly to the surrounding tissues, LMS should be considered. Wide surgical excision with neck dissection for lymph node metastases, and long term regular follow up is the mainstay of treatment.

Disclosure of conflict of interest

None.

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