

Case Report

Angioleiomyoma of the cheek: a case report

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(Received October 26, 2014; Accepted February 16, 2015)

Abstract: Angioleiomyoma is a form of subcutaneous vascular leiomyoma that usually occurs in the extremities. Leiomyoma of the oral cavity represents only 0.4% of soft tissue neoplasms and 0.06% of leiomyomas. Isolated cases of angioleiomyoma have been reported in the knee and lower thigh, gastrointestinal tract, genital and renal tract, and brain. Histopathologic examination by biopsy is necessary to establish a diagnosis, and immunohistochemical staining, along with conventional hematoxylin-eosin staining, is important. The differential diagnosis includes hemangioma and angiosarcoma. At present, surgical resection is the standard therapy for leiomyoma, and recurrence is extremely rare. We report a rare case of angioleiomyoma of the cheek in a 45-year-old man. The postoperative course was uneventful, without complications. (*J Oral Sci* 57, 63-66, 2015)

Keywords: angioleiomyoma; vascular leiomyoma; oral cavity.

Introduction

Angioleiomyoma is a benign tumor resulting from nodular proliferation of vascular smooth muscle cells. It presents most often in the extremities, especially the

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doi.org/10.2334/josnusd.57.63
DN/JST.JSTAGE/josnusd/57.63

knee and lower thigh, and is more frequently observed in middle-aged women. It occurs rarely in the oral cavity. Angioleiomyoma of the head and neck region accounts for less than 0.06% of angioleiomyomas (1). The most common site of leiomyoma in the head and neck region is the lips, followed by the tongue, cheek and palate, gingiva, and mandible. Oral leiomyoma usually presents as angioleiomyoma, which accounts for 64-66.2% of reported cases of oral leiomyoma (1). We report a case of angioleiomyoma of the cheek.

Case Report

A 45-year-old man visited our department for evaluation and treatment of a painless mass in the right buccal region. His medical and family histories were unremarkable. He became aware of swelling in his right cheek and sought treatment at a dental clinic 4 days later. A tumor was suspected, and he was referred to our hospital for detailed assessment and treatment.

Findings outside the oral cavity

The patient was of medium build and with good nutritional status. A walnut-sized swelling was noted in the right cheek; the regional lymph node appeared normal.

Findings in the oral cavity

An elastic soft mass (diameter, about 20 mm) was palpable in the area from the subcutaneous tissue to submucosal tissue of the right cheek. The boundary between the mass and normal tissue was relatively clear. There was no adhesion with surrounding tissue, and the mass was movable. The skin and covering mucosa were normal, and neither spontaneous pain nor tenderness was



Fig. 1 A photograph of the cheek region, showing a walnut-sized area of swelling.

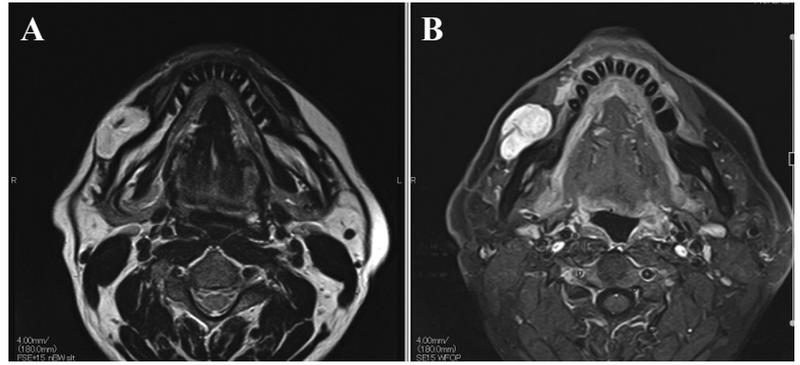


Fig. 2 **A.** Axial fast spin-echo (FSE) T2-weighted image showing a uniform signal and hyperintense lesion on the right side of the cheek. **B.** Enhanced axial (FSE) T1-weighted image showing the enhancement pattern and relation to the intraoral tissue of the cheek.

noted (Fig. 1).

MRI findings

The mass was located in the area from the subcutaneous to submucosal tissue of the right cheek region. A T2-weighted image showed a uniform signal and hyperintense lesion in intraoral tissue of the affected cheek. A contrast-enhanced T1-weighted image showed a homogeneously enhanced mass and a vessel leading to the tumor (Fig. 2). The findings were consistent with a clinical diagnosis of tumor of the cheek.

Treatment and clinical course

A diagnosis of angioleiomyoma/hemangioma was made based on the biopsy results. Extirpation was performed under general anesthesia. No adhesion with surrounding tissue was noted, and blunt removal of the tumor was straightforward. The feeding vessels were ligated and excised en bloc with the tumor. At this writing, 18 months after surgery, there is no evidence of recurrence.

The 22×15×11 mm tumor was elliptical and had a smooth dusky-red surface. Hematoxylin-eosin (H-E) staining showed a mass with a clear boundary containing various types of dilated arterioles and veins. The mass was located in the mucosal subepithelial connective tissue and was covered by a membrane. Proliferation of vessels with a smooth muscle wall and vessels surrounded by smooth muscle cells and with a slit-like vascular lumen were noted. No nuclear atypia or karyokinesis was observed (Fig. 3).

Immunohistochemical staining was positive for α -smooth muscle actin, partially positive for desmin, and slightly positive for vimentin. Only vascular endothelial cells were CD34-positive (Fig. 4).

These results confirmed a histopathologic diagnosis of angioleiomyoma.

Discussion

Angioleiomyoma is a benign tumor originating in the smooth muscle tissue in the tunica media. It usually presents in the subcutaneous region of the extremities and is rare in the oral cavity. Some reports indicate that angioleiomyoma represents 3.3-9% of tumors in the oral cavity. Ishikawa et al. found that the most frequently involved sites in the oral cavity were the lip (48.6%), palate (21.1%), buccal mucosa (9.2%), tongue (9.2%), mandible (8.3%), buccal sulcus (0.9%), labial sulcus (0.9%), mouth floor (0.9%), and gingiva (0.9%) (2). Age at onset ranges from 1 month to 84 years, although most patients are 30 years of age or older. Morimoto et al. reported that angioleiomyoma of the extremities is more prevalent in women (male:female ratio, 2:3) and that angioleiomyoma of the head and neck region is more prevalent in men (male:female ratio, 3:1) (3).

Angioleiomyoma development is believed to be triggered mainly by warming stimulus or mechanical trauma, venous stasis, and hormonal changes (4,5). We experienced one case of angioleiomyoma, in a 45-year-old man, the age and sex in which angioleiomyoma is more common. Although the tumor was positioned near teeth in the upper and lower jaws, there were no obvious problems in dentition, tooth crown morphology, or dental occlusion. Thus, it was unlikely that those teeth chronically stimulated the original tumor site. The patient had smoked 40 cigarettes per day for 25 years, and tobacco exposure was suspected to be the source of chronic stimulus at the site.

Tumors in the oral cavity are mostly painless and localized and present as a palpable elastic soft masses or elastic firm masses beneath the mucosa. The color of the surface mucosa varies widely. It may appear normal, pink, or dusky red. Lesions grow extremely slowly and

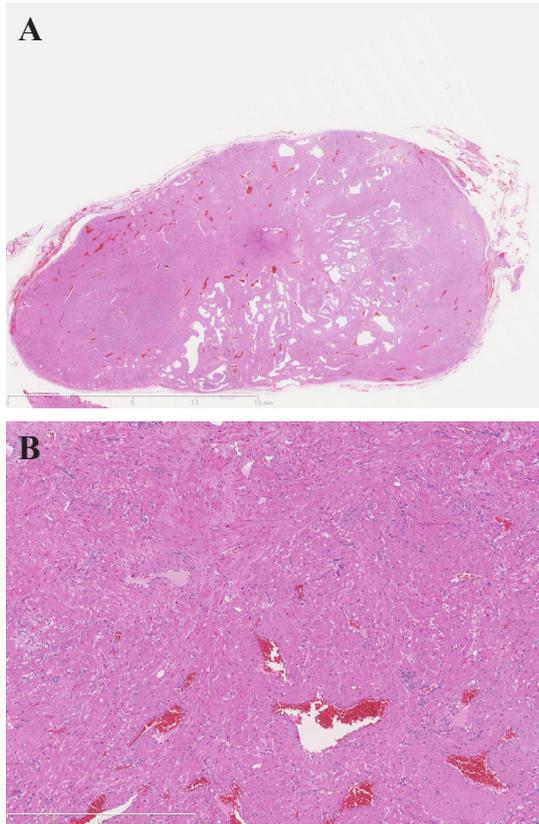


Fig. 3 Pathologic findings: The micrograph shows the tumor forming a definite border with several areas of vasodilation (A; H-E stain $\times 0.67$). The micrograph shows thick-walled blood vessels and bundles of smooth muscle cells (B; H-E stain $\times 5$).

form nodules with clear boundaries. The present tumor was also painless, localized, elastic, and soft, and the surface mucosa was normal.

The clinical differential diagnosis of angioleiomyoma includes pyogenic granuloma, lymphangioma, and malignancies such as angioleiomyosarcoma, which are oral tumors that can present as angioleiomyoma (6). However, they are impossible to differentiate clinically from angioleiomyoma.

The histologic features of angioleiomyosarcoma and angioleiomyoma are similar. Atypia, cellularity, pleomorphism, and necrosis suggest malignant disease, and mitotic activity is the most reliable criterion of malignant behavior (6). Tumors with 1-4 mitoses per 10 high-power fields (HPF) should be regarded as potentially malignant, and those with 5 or more mitoses per 10 HPF as malignant (7). The present tumor had a mitotic activity of 0/10 HPF.

To establish a diagnosis, histopathologic examination by biopsy is necessary, and use of immunohistochemical staining in addition to conventional HE staining is important. The immunophenotype of the neoplastic cells

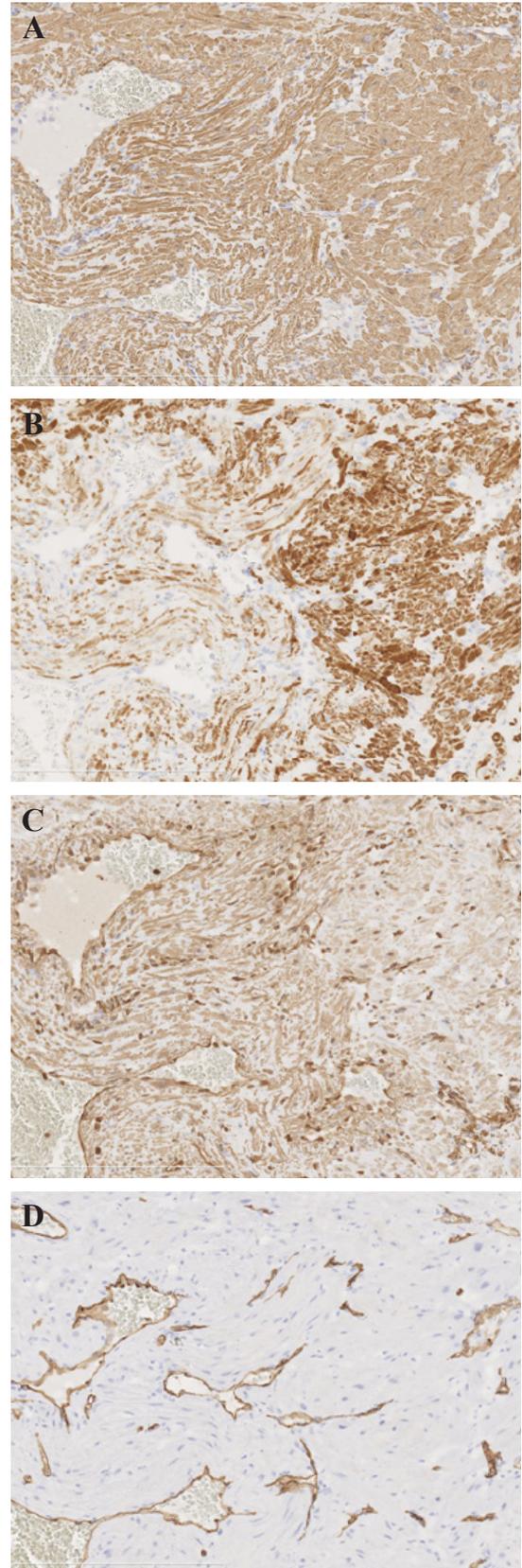


Fig. 4 Immunohistochemical staining with α -smooth muscle actin confirms the smooth muscle origin of tumor cells (A). Desmin was partial positive (B). Vimentin was weakly positive (C). CD34 was negative, although the endothelium of vascular spaces was positive (D). ($\times 20$)

is α -smooth muscle actin (+), vimentin (+), desmin (+), CD34 (-) (although the endothelium of vascular spaces is CD34+), and S-100 protein (-) (8).

In our patient, expression of α -smooth muscle actin was strongly positive, desmin was partially positive, vimentin was weakly positive, and CD34 was negative (although the endothelium of vascular spaces was CD34-positive). These results confirmed a diagnosis of angioleiomyoma.

Recurrence after extirpation is highly unlikely, and postoperative prognosis is favorable. However, recurrence due to inadequate extirpation has been reported (9), although such cases are rare. Angioleiomyomas are benign lesions. Although malignant transformation is a possibility, no case of malignant transformation has been reported. Histopathologic differentiation of leiomyoma from low-grade leiomyosarcoma can be difficult. Thus, careful long-term follow-up may be warranted for any smooth muscle neoplasm of the oral cavity (10).

At this writing, 18 months after surgery, our patient is making good progress, and there are no signs of recurrence. Long-term follow-up is continuing.

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