Angioleiomyoma of the upper lip: report of a case

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Abstract. A rare case of vascular leiomyoma of the upper lip in a 51-year-old man is presented. The differential diagnosis, frequency, treatment, and prognosis are discussed.

Leiomyomas are benign mesenchymal tumors derived from smooth muscle. They are subdivided into three groups: vascular (angiomyomas), which comprise approximately 74%; solid/leiomyomas, which comprise approximately 25%; and epithelial leiomyomas. Vascular leiomyomas are uncommon, slow-growing, subepithelial, and subdermal or subcutaneous tumors. They are most commonly found in the anterior aspect of the lower extremities (approximately 70%), and affect predominantly middle-aged women. The face is rarely affected. 5-10% of all angioleiomyomas are as follows: lip = 26%, tongue = 21%, cheek = 20%, palate = 19%, and nose = 11%.

A 51-year-old Caucasian man was referred to the oral and maxillofacial department of the University Hospital of Nijmegen. The lesion was discovered 4 years ago, and has increased in size over the last year. Histopathology revealed an angioleiomyoma of the upper lip. Surgical excision was performed, and the patient is well 6 months later.

Case report

18% and others 14% (gingival floor of the mouth, etc.). There are currently only 84 reported cases of oral angioleiomyomas of which 15 occurred on the upper lip.

Fig. 1. Photograph of a 51-year-old patient with bi-elastic swelling measuring 1.5 × 1 cm of the left upper lip causing aesthetic inconvenience and annoyance.

Fig. 2. Histopathological appearance of angioleiomyoma. Large, smooth muscle tumor cells (c) with rounded ends (HE × 50, inset: thickened vessel walls). Some lipocytes present (HE × 300).

Key words: angioleiomyoma, vascular leiomyoma, upper lip.
A n a s t a s s o v a n d v a n D a m m e

The lesion was completely removed with a small margin of clinically healthy tissue. The specimen felt solid and did not appear to be a nodule, cyst, or hemangioma; it was submitted for histopathologic examination. The postoperative course was uneventful, and there has been no evidence of recurrence 14 months postoperatively.

### Histopathology

The tumor was embedded in paraffin and stained with HE. The microscopic examination revealed a largely encapsulated lesion composed of irregularly arranged smooth-muscle cells with some adipose tissue and abundance of arterial-type blood vessels of varying calibers. The tumor cells were large and had the elongated nuclei with rounded ends which are characteristic of smooth-muscle tumor cells (Fig. 2). The immunohistochemical analysis confirmed the presence of proliferation of smooth-muscle cells after positive staining for alpha-SM1. Special pericyte stainings as well as endothelial stainings (factor VIII-related antigen) were negative for the tumor cells. The final histopathologic diagnosis was angioleiomyoma.

### Discussion

Vascular leiomyomas of the upper lip are rare. The exact origin of leiomyomas is still unknown, but most authors agree that the tumor arises from the smooth muscle of vessel walls, aberrant adnexial smooth muscle, arteriovenous anastomoses, and ectopic thyroglossal ducts, as well as hamartomas. However, leiomyomas and angioleiomyomas in particular are histologically similar and are composed of vascular spaces of different caliber

The smooth-muscle cells are interconnected between and with the surrounding smooth-muscle cells from the adjacent vessels. It is likely, therefore, that the histologic origin of these benign tumors is related to the smooth muscle of the vascular wall.

Histologically, the lesion somewhat resembles hemangiopericytoma, but does not exhibit the distinguishing characteristics of pericytomas, which are composed of pericytes with contractile properties but lacking myofibrils. Due to the abundance of small arterial blood vessels, a diagnosis of hemangiopericytoma could be considered. However, the presence of numerous smooth-muscle cells with rounded nuclei and the positive immunohistochemical stain for alpha-SM1 confirm the histopathologic diagnosis of angioleiomyoma. A test highly specific for vascular lesions, the factor VIII-related antigen immunoassay, was negative.

There is consensus regarding the treatment of this lesion, i.e., surgical excision. The postoperative prognosis is generally good. The recurrence rate is very low, recurrence being thought to be due to inadequate excision of the initial lesion.

### Acknowledgments

We would like to thank Dr P. C. M. De Wilde, DMD, PhD, of the Department of Pathology, St Radboud University Hospital, Nijmegen, The Netherlands, for the histopathologic analysis, and Armando Gama for linguistic advice.

### References


