

CASE REPORT

Angiolipoma of the buccal mucosa: a possible role of mast cell-derived VEGF in its enhanced vascularity

Hiroko Ida-Yonemochi¹, Wael Swelam¹, Chikara Saito², Takashi Saku¹

Divisions of ¹Oral Pathology and ²Reconstructive Surgery for Oral and Maxillofacial Region, Department of Tissue Regeneration and Reconstruction, Niigata University Graduate School of Medical and Dental Sciences, Niigata, Japan

A case of angiolipoma occurring in the buccal mucosa of a 69-year-old male is described. The patient had noticed a painless mass in his buccal mucosa for 2 years. The surgically removed tumor, measuring 9 mm in diameter, was mainly located in the submucosal layer with focal expansion into the muscle layer. Histologically, the tumor was well-demarcated and composed of proliferations of mature fat cells and fibrous connective tissue containing many small blood vessels, which were evenly distributed. There was diffuse infiltration of a large number of mast cells, which were immunopositive for vascular endothelial growth factor (VEGF) especially around blood vessels, suggesting that VEGF produced by mast cells in angiolipomas plays an important role in the vascular proliferation in this particular tumor.

J Oral Pathol Med (2005) 34: 59–61

Keywords: angiogenesis; angiolipoma; mast cell; tryptase; vascular endothelial growth factor

The patient was a 69-year-old Japanese male. He had noticed a painless nodule over the course of 2 years, after he happened to bite his cheek. Since the mass increased in size gradually, he was referred to our hospital by his physician. Intraoral examination revealed a polypoid mass, measuring 9 mm in diameter, in the right buccal mucosa. Under a clinical diagnosis of lipoma, the lesion was surgically removed under local anesthesia.

Histopathologically, the lesion was a well-demarcated tumor located in the submucosal to muscle layers (Fig. 1). The tumor was composed of proliferation of mature fat cells and fibrous connective tissues, which

were mixed diffusely with each other, and lacked fibrous septa forming lobular structures. The connective tissue contained many small blood vessels (Fig. 2a). The fat cells had no conspicuous cellular atypia. The blood vessels, which were demonstrated with CD34 immunohistochemistry, were evenly scattered within the lesion (Fig. 2b). There was a diffuse and plentiful infiltration of mast cells, which showed immunopositivities for tryptase (Fig. 3a), while no other inflammatory cells were seen in the lesion. Mast cells were sometimes condensed around capillaries and those in the perivascular space were immunopositive for vascular endothelial growth factor (VEGF) (Fig. 3b). When double immunofluorescence was performed for mast cell tryptase and VEGF, positive fluorescence signals for these two antigens were merged on most of the mast cells (Fig. 4).

The patient's recovery was uneventful with no recurrence for 2 years after surgery.

Comments

Angiolipoma, a vascular variant of lipoma, appears usually as a subcutaneous nodule in the trunk and extremities. Including the present case, only 15 cases of

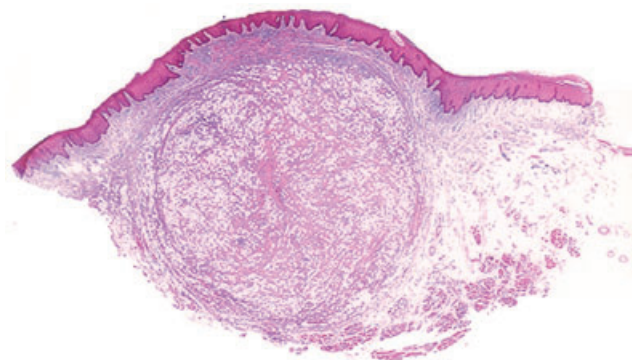


Figure 1 Angiolipoma of the buccal mucosa (H&E, ×10). A well-demarcated round-shaped tumor was located in the submucosal to muscle layers. There was no definite capsular structure.

Correspondence: Dr Takashi Saku, Division of Oral Pathology, Department of Tissue Regeneration and Reconstruction, Niigata University, Graduate School of Medical and Dental Science, 2-5274 Gakkocho-dori, Niigata 951-8126, Japan. Tel: (+81) 25 227 2832, Fax: (+81) 25 227 0805. E-mail: tsaku@dent.niigata-u.ac.jp
Accepted for publication January 26, 2004

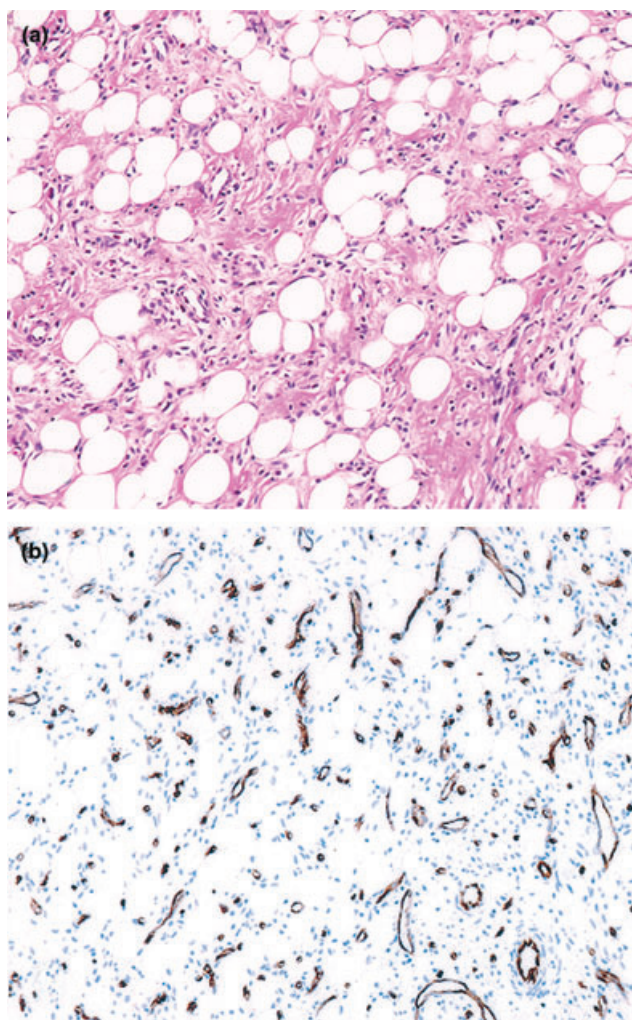


Figure 2 Angiolipoma of the buccal mucosa in high power view. (a) H&E, (b) immunoperoxidase stain for CD34 with hematoxylin counterstain ($\times 160$). The tumor is composed of a diffuse proliferation of mature fat cells intermixed with thick fibrous connective tissues containing many small blood vessels (a). By CD34 immunohistochemistry, blood vessels were demonstrated to be evenly scattered within the lesion (b).

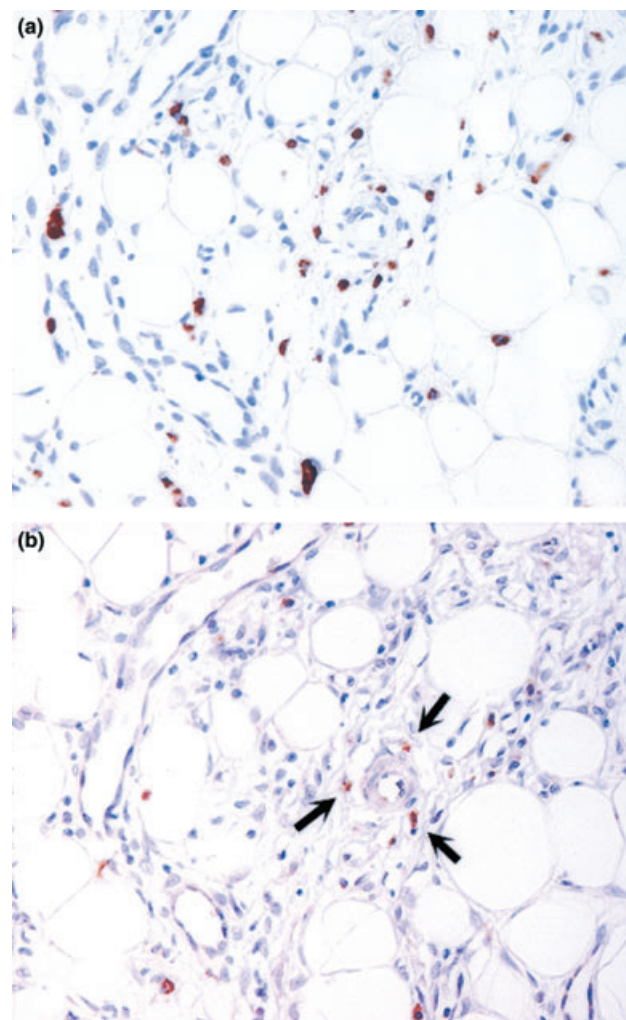


Figure 3 Mast cell infiltration in angiolipoma of the buccal mucosa. (a) Immunoperoxidase stain for tryptase. (b) Immunoperoxidase stain for vascular endothelial growth factor (VEGF). Counterstain with hematoxylin, $\times 400$. Tryptase immunopositive mast cells were scattered diffusely within the tumor tissue with a denser distribution around capillaries (a). Perivascular mast cells (arrows) were simultaneously immunopositive for VEGF (b).

angiolipoma arising in the oral cavity have been reported in the English literature (1, 2). Among these 15 cases, 9 were male and 6 were female, a ratio of 1.5:1. The ages of the patients ranged from 4 to 81 years (mean of 37 years). Among them, there were 7 tumors in the cheek (47%), 3 in the mandible (20%), 2 each in the tongue and palate (13%), and 1 in the lip (6%). No traumatic history was documented in most of these cases, while our patient had a history of trauma due to cheek biting. The histological characteristics of angiolipoma are a mixture of mature adipocytes and interspersed connective tissues with vascular vessels containing fibrin thrombi and mast cell infiltration, which are distinct from usual lipomas (3). Mast cells have been thought to mediate angiogenesis in various pathophysiological conditions (4). Also, in

angiolipomas, it has been speculated that mast cells might play some roles in their increased vascularity, based on the data that angiolipomas had 10 times the number of mast cells than classic lipomas (5). However, there has been no direct demonstration of molecular mechanisms for angiogenesis participation by mast cells in angiolipoma. In the present case, we could show that mast cells around blood vessels strongly expressed VEGF, which is known to be an essential growth factor for endothelial cells in vasculogenesis. Although we did not perform *in situ* hybridization for it, VEGF production by mast cells should be highly probable, because there were no other inflammatory cells within the tumor tissue. The result indicates that mast cell-derived VEGF might be responsible for the enhanced vascularity in this tumor.

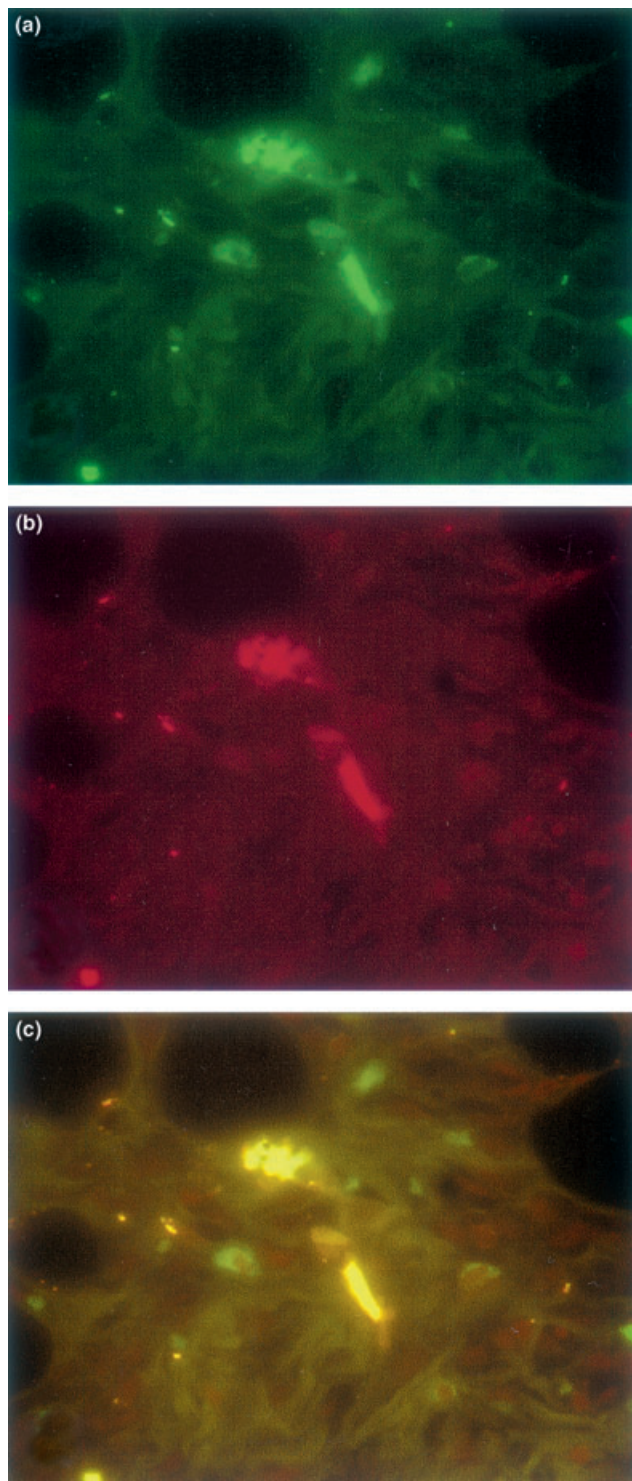


Figure 4 Mast cell infiltration in angiolipoma of the buccal mucosa. Double immunofluorescence stain for tryptase (fluorescein isothiocyanate (FITC)) (a) and vascular endothelial growth factor (VEGF) (rhodamine) (b), $\times 400$. (c) Merged image of (a) and (b). Simultaneous immunofluorescence for tryptase (green, a) and VEGF (red, b) was confirmed in mast cells (yellow, c).

References

1. Hamakawa H, Hino H, Sumida T, Tanioka H. Infiltration angiolipoma of the cheek: a case report and a review of the literature. *J Oral Maxillofac Surg* 2000; **58**: 674–7.
2. Manganaro AM, Hammond HL, Williams TP. Intraosseous angiolipoma of the mandible: a case report and review of the literature. *J Oral Maxillofac Surg* 1994; **52**: 767–9.
3. Enzinger FM, Weiss SW. *Soft tissue tumors*, 4th edn. St Louis: CV Mosby, 2001; 582.
4. Levi-Schaffer F, Pe'er J. Mast cells and angiogenesis. *Clin Exp Allergy* 2001; **31**: 521–4.
5. Shea CR, Prieto VG. Mast cells in angiolipomas and hemangiomas of human skin: are they important for angiogenesis? *J Cutan Pathol* 1994; **21**: 247–51.

This document is a scanned copy of a printed document. No warranty is given about the accuracy of the copy. Users should refer to the original published version of the material.