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Myxoid Tumor of the Oral Cavity With Features of Superficial Angiomyxoma: Report of a Case

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Myxomas of oral and paraoral soft tissue have been well documented by Elzay et al.[1] However, oral angiomyxoma has not been reported in the English language literature. Recently, we encountered a myxoid tumor in the buccal mucosa with a prominent vascular component histologically and immunohistochemically that was compatible with a diagnosis of angiomyxoma.

Report of Case

A 19-year-old Chinese man complained of a nonpainful, nontender mass in the right buccal mucosa present for about 2 years. His medical history was noncontributory. Examination showed that the lesion was soft, freely movable, and covered with normal overlying mucosa. The clinical diagnosis of a benign mesenchymal soft tissue tumor was made. An excisional biopsy was performed that showed a well-defined polypoid, nonencapsulated mass. The patient had an uneventful postoperative course, and the lesion has not recurred 22 months after surgical excision.

On gross examination, the tumor was lobulated and grayish-white, measuring 5 × 3.5 × 3 cm (Fig 1). The cut surface had a yellow-white, gelatinous, glistening appearance. Histologically, the tumor was composed of loosely textured connective tissue with a prominent vascular component (Fig 2). The tumor cells were spindle-shaped with poorly defined, scanty cytoplasm and an absence of mitoses (Fig 3). A myxoid change was prominent in the stroma, with dispersed delicate wavy collagen fibers. The matrix was strongly positive on staining with colloidal iron and weakly positive with Alcian blue (pH 2.5). A prominent vascular component, ranging from small- to medium-caliber, thin-walled vessels without medial hypertrophy, was distributed haphazardly throughout the tumor tissue. Lipoblasts or other identifiable neoplastic components were not observed. Neither smooth muscle nor small nerves were seen.

An immunohistochemical study was performed using the avidin-biotin-peroxidase complex (ABC) technique. Primary antibodies against vimentin (1:5), actin (1:200), desmin (1:100), factor VIII-related antigen (1:400), S-100 protein (1:200), neurone-specific enolase (1:50), and α-1-antichymotrypsin (1:100) (Dakopatts, Copenhagen, Denmark) were used. Many tumor cells stained strongly for vimentin (Fig 4), weakly for actin, and negatively for desmin. Smooth muscle cells of the vessels stained positively for actin and desmin. No immunostaining was observed for S-100 protein, neuron-specific enolase, or α-1-antichymotrypsin. The endothelial cells of the blood vessels displayed immunoreactivity for factor VIII-related antigen (Fig 5). The diagnosis was soft tissue myxoma with features of angiomyxoma.

Discussion

The tumor was composed of a mixture of myxoid and vascular components, highly resembling an angiomyxoma, a rare mesenchymal myxoid tumor typically including aggressive[2] and superficial[3] types. Aggressive angiomyxoma, most frequently found in the female vulva, pelvic floor, and perineum,[5] shows local infiltrative growth and has a propensity to recur.[2] Superficial angiomyxoma involves merely the cutaneous and subcutaneous tissues without infiltrating deeper structures;[4] it has been reported in the skin[4] and umbilical cord.[4] Review of the English literature showed no previously reported case of angiomyxoma in the oral cavity; however, the histologic and immuno-

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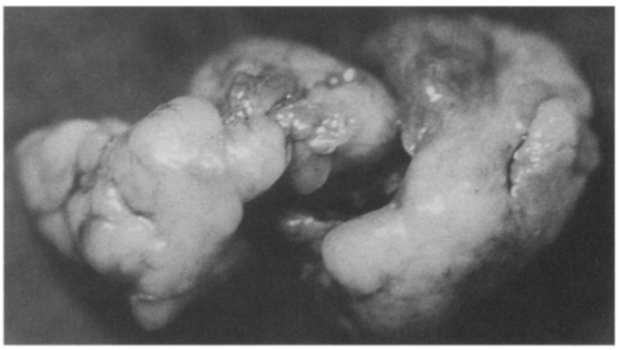

FIGURE 1. Lobulated appearance of the surgical specimen.

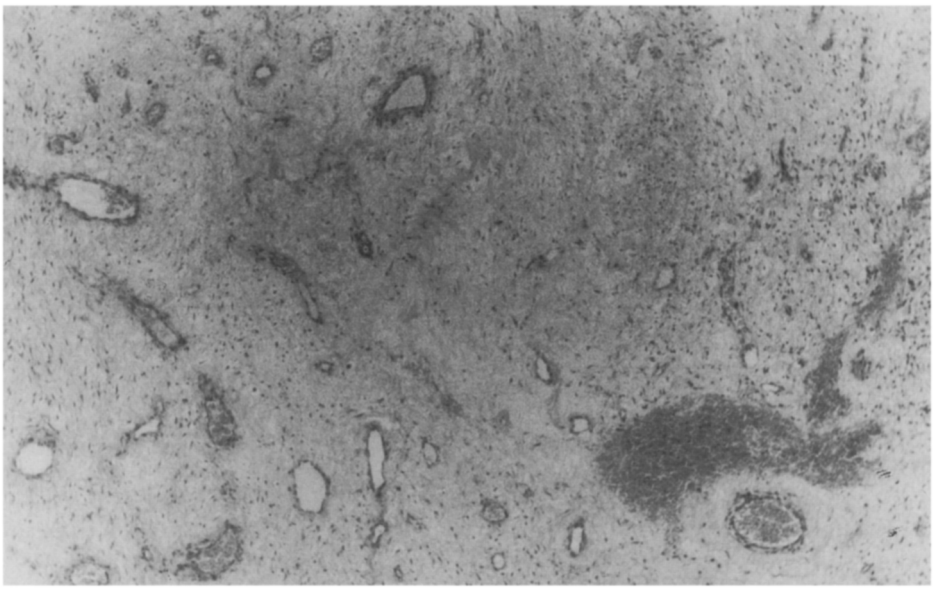

FIGURE 2. Photomicrograph showing loose collagenous stroma permeated by spindle-shaped cells and nonarborizing blood vessels. Focal areas of interstitial hemorrhage and vascular congestion are also seen (hematoxylin & eosin stain, original magnification ×40).

histochemical findings of the current lesion strongly resemble those associated with angiomyxoma.

The mesenchymal spindle and stellate cells of the current tumor were strongly positive on staining for vimentin and, to a lesser extent, for actin. Begin et al[5] claimed that vimentin the intermediate filaments of the tumor cells in angiomyxomas stain for, and have a fibroblastic origin. Steeper and Rosai [2] showed ultrastructurally that the spindle-shaped stromal cells showed features consistent with myofibroblastic differentiation. Because of these ultrastructural and immunohistochemical features, the histogenesis of angiomyxoma is regarded as being of myofibroblastic[2] or fibroblastic [5] origin.

Although distant metastasis has not occurred with aggressive angiomyxoma, local recurrences were common.[2] Conversely, local recurrence has also been reported in superficial angiomyxoma because of incomplete excision.[3] This suggests that angiomyxoma is a true neoplasm rather than a hamartoma or a reactive hyperplastic process. The differential diagnosis includes angiomyolipoma and various myxoid tumors such as myxoid lipoma and liposarcoma, nerve sheath myxoma, myxoid neurofibroma, myxoid type of embroyonal rhabdomyosarcoma, myxoid variant of malignant fibrous histiocytoma, and fibromatosis with focal myxoid areas.

Angiomyolipoma is composed of a mixture of

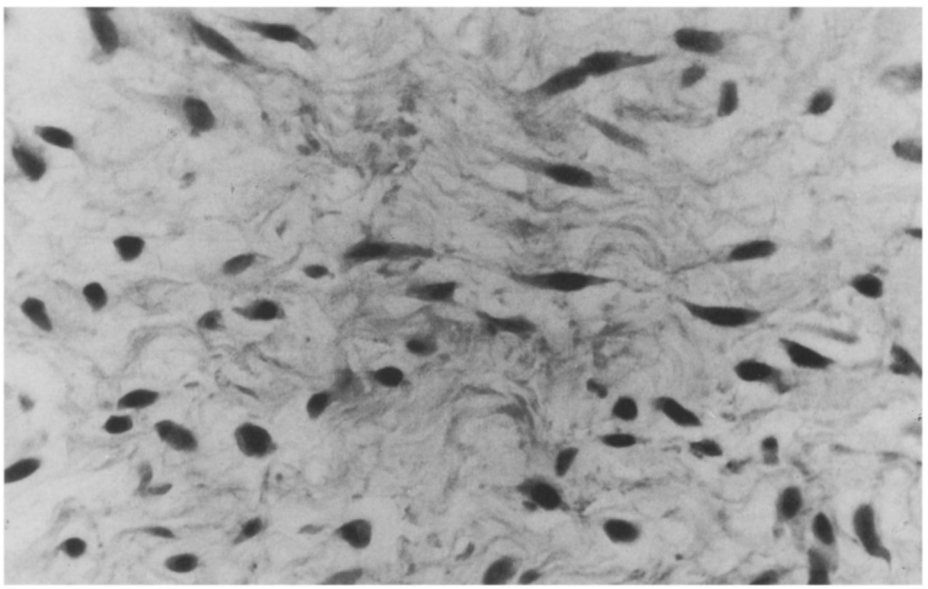

FIGURE 3. High-power view shows spindle and stellate cells with poorly defined, scanty, cytoplasm, and the absence of mitosis (hematoxylin & eosin stain, original magnification ×400).

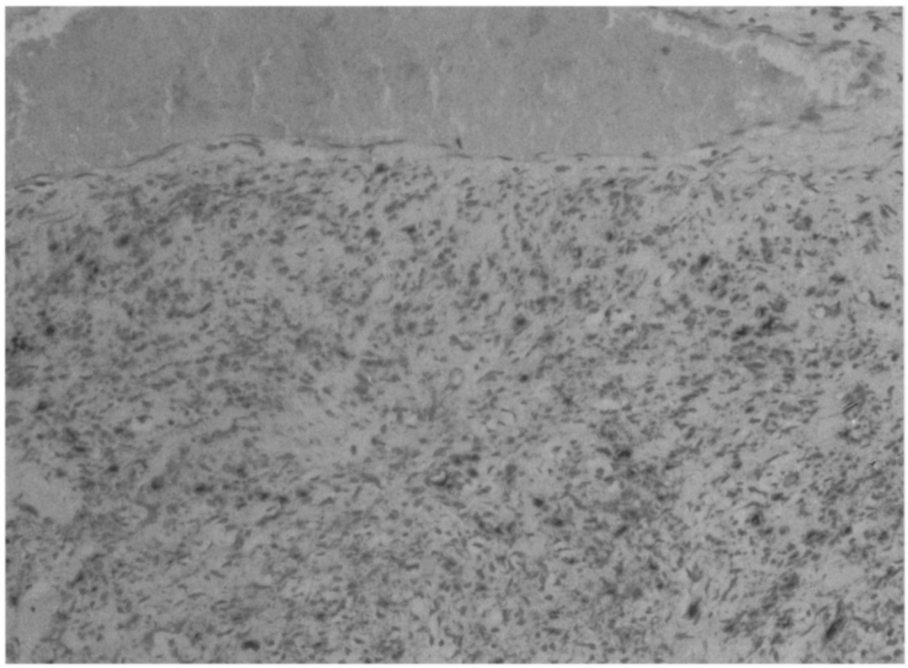

FIGURE 4. *A,* Many tumor cells stained strongly for vimentin (ABC stain, original magnification ×100).

thick-walled blood vessels, smooth muscle, and adipose tissue. It is found mostly in the kidney. Although a few cases of extrarenal, perinephric, angiomyolipoma have been described,[6] neither smooth muscle nor adipose tissue were seen in the tumor tissue of the current case. The distinctive histologic features (prominent vascular component, extensive myxoid areas, absence of mitosis) and immunohistochemical finding (strongly positive for vimentin) of this buccal lesion distinguished it from other myxoid tumors. Additionally, the microscopic distinction of angiomyxoma from other myxoid tumors has been summarized by Steeper and Rosai[2] and Allen et al.[3]

In summary, the clinical, histologic, and immunohistochemical findings of this lesion were consistent with those found in the superficial angiomyxoma. Because this lesion stays superficial, without affecting deeper structures, the prognosis for the patient would

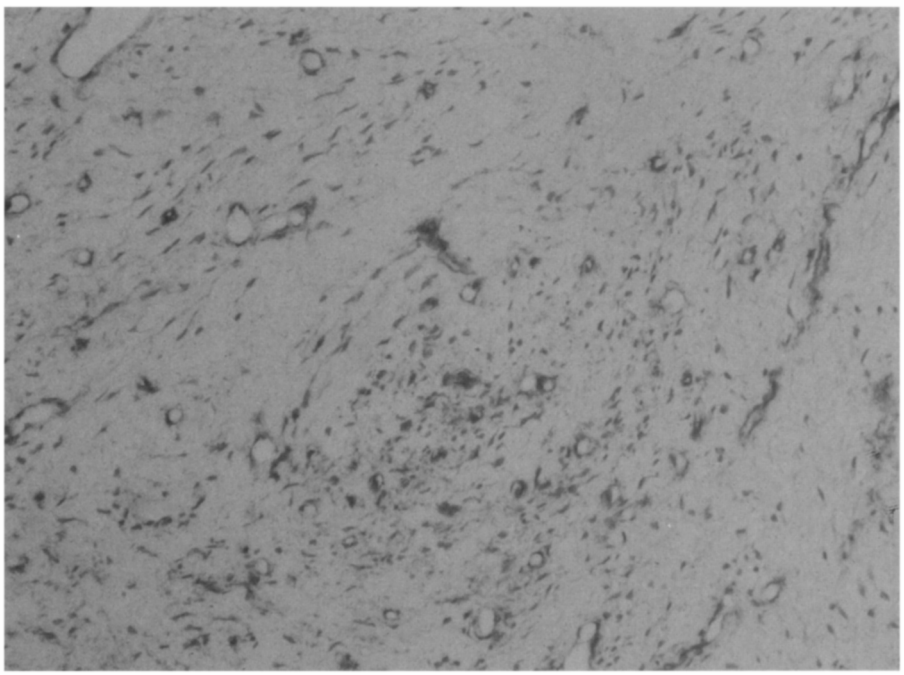

FIGURE 5. The endothelial cells of the blood vessels show immunoreactivity for factor VIII–related antigen (ABC stain, original magnification ×100).

seem to be good because a wide excision was done. However, there needs to be long-term follow-up.

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