Time to abandon the term angiomyolipoma for non-PEComatous angiomyomatous (or angiomatous) oral tumors with adipocytes

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Angiomyolipoma (AML) is the most common benign mesenchymal neoplasm of the kidneys with well-established clinical and morphological features. The oral and maxillofacial pathology literature contains several examples that identify angiomyomatous proliferations of the oral mucosa that contain an adipocytic component as analogous to classic renal AMLs although they differ significantly in their immunohistochemical phenotype. Herein, through review of the pertinent oral pathology literature and the detailed description of 2 lesions, one an oral angiomyoma with an adipocytic component and the other an apparently hamartomatous angioleiomyomatous proliferation with adipocytes, we provide, in our opinion, a solid argument against the use of the term AML for non-PEComatous oral tumors. **(Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;110:492-497)**

Angiomyolipoma (AML), originally believed to be a hamartoma, is a benign mesenchymal clonal neoplasm occurring most frequently in the kidneys as a sporadic tumor or as part of the tuberous sclerosis complex.[1,2] It is composed of a variable proportion of abnormal thick-walled blood vessels, spindle and epithelioid smooth muscle cells, and adipocytes.[3] Loss of heterozygosity of tuberous sclerosis gene TSC2 (tuberin) has been reported in syndromic and few sporadic AMLs.[4]

AML belongs to the spectrum of lesions referred to as perivascular epithelioid cell tumors (PEComas) that are defined by the presence of so-called perivascular epithelioid cells (PECs) featuring myomelanocytic immunohistochemical and ultrastructural properties.[3] Histologically, PECs do not have a known normal cellular counterpart, have epithelioid or spindle cell morphology, clear to granular eosinophilic cytoplasm, and a distinct perivascular distribution. Leiomyomatous origin has been hypothesized; however, there is no evidence up to now of a mechanism that causes smooth muscle cells to feature melanin-formative capabilities.

PECs generally stain for myocytic markers, such as smooth muscle actin, pan-muscle actin, muscle myosin, and calponin, as well as melanocytic markers, including HMB-45, MelanA/Mart1, tyrosinase, and microphthalmia transcription factor (Mitf).[1,2,5-8] Interestingly, PECs are most frequently negative for S-100, and HMB-45 positive musclelike and adipocyticlike cells in renal AML are thought to represent phenotypical derivates of PECs.[1,3,9] The PEComa group of tumors also includes clear cell "sugar" tumor of the lung, lymphangioleiomyomatosis, clear cell myomelanocytic tumor of the falciform ligament/ligamentum teres, and unusual clear cell tumors of other visceral and somatic tissues.[1,5,10]

AML may develop in extrarenal sites, in particular the liver, and rare examples have been reported in other anatomical sites.[2] A review of the English-language literature disclosed some tumors in the head and neck with microscopic features similar to those of renal AML: 10 in the oral cavity (summarized in Table I),[11-19] 8 in the regional skin,[20] 6 in the nasal cavity,[21-25] and 1 each in the parotid gland,[26] the larynx,[27] and the nasopharynx.[28] All tumors, however, were not justified immunohistochemically as analogous to renal AML. In contrast,

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Table I. Main clinical features of 12 cases of oral "angiomyolipoma"

Reference	*Age*	*Sex*	*Site*	*Duration, y*	*Maximum dimension, cm*	*Follow-up, mo*
Gutmann et al. 1975[11]	39	M	Hard palate	2	1	84 FOD
Yamamoto et al. 1995[12]	62	F	Hard palate	n/a	1	12 FOD
	69	F	Lower lip	10	1	12 FOD
Piattelli et al. 2001[13]	43	M	Palate	<1	n/a	84 FOD
Redman et al. 2001[14]	71	M	Lower lip	4	2	18 FOD
Lopez Lopez et al. 2004[15]	55	F	Lower lip	n/a	n/a	n/a
Farah and Zaini 2006[16]	54	F	Hard palate	20	1	n/a
da Silva et al. 2007[17]	43	F	Upper lip	6	2	48 FOD
Alvarez Alvarez et al. 2007[18]	52	M	Hard palate	n/a	n/a	a/a
Koizumi et al. 2008[19]	23	M	Tongue	2	0.8	48 FOD

n/a, not available; *M,* male; *F,* female; *FOD,* free of disease.

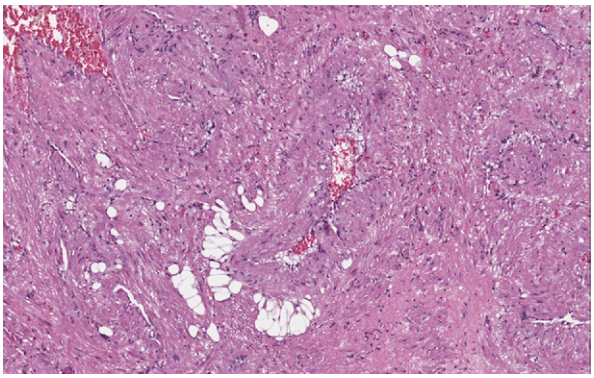

Fig. 1. Tumor composed of thick-walled blood vessels with abnormal shape and bundles of intensely eosinophilic smooth muscle cells (hematoxylin and eosin [H&E], original magnification ×100).

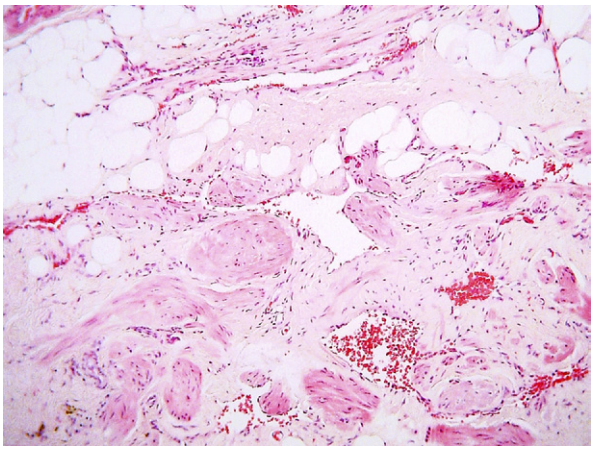

Fig. 2. Tumor composed of blood vessels, bundles of smooth muscle cells, and groups of fat cells (H&E, ×200).

there are only 2 well-documented cases of oral PEComas, one reported by Koutlas et al.[29] on the palate of a 46-year-old woman, the other by Accurso et al.[30] on the lower lip of a 58-year-old male.

We describe 2 oral tumors presenting a mixture of blood vessels, smooth muscle cells, and mature fat cells, compare them with previously reported oral AMLs, and argue against the use of the term AML for non-PEComatous oral lesions.

CASE 1

A 78-year-old otherwise healthy male presented with a painless lump of the upper lip of 1-year duration. His medical history was noncontributory. Clinical examination showed a 0.7-cm round submucosal mass just left of the upper lip midline. On palpation it gave the impression of a fluid-filled cavity and was covered by normal mucosa. It was surgically excised with a provisional diagnosis of mucocele. The postoperative course was uneventful and 6 months after excision no recurrence has been reported.

Histologic sections showed a tumor composed of thick-walled blood vessels, bundles of intensely eosinophilic smooth muscle cells radiating from the vessels' walls, and small group of mature fat cells (Fig. 1). The final diagnosis was angio(leio)myoma with an adipocytic component.

CASE 2

A 19-year-old otherwise healthy female presented with an enlargement of the right cheek of unknown duration. Clinical examination revealed a freely movable submucosal mass of elastic consistency that was covered by normal mucosa. Ultrasonographically, it was consistent with lipoma. The tumor was surgically excised under local anesthesia. The postoperative course was uneventful and 4 years after surgery no recurrence has been reported.

On microscopic examination, an ill-defined and unencapsulated tumor was appreciated. It was composed of blood vessels, bundles of smooth muscle cells, and groups of fat cells (Fig. 2). Blood vessels were mainly thin walled and capillary size, whereas some medium-

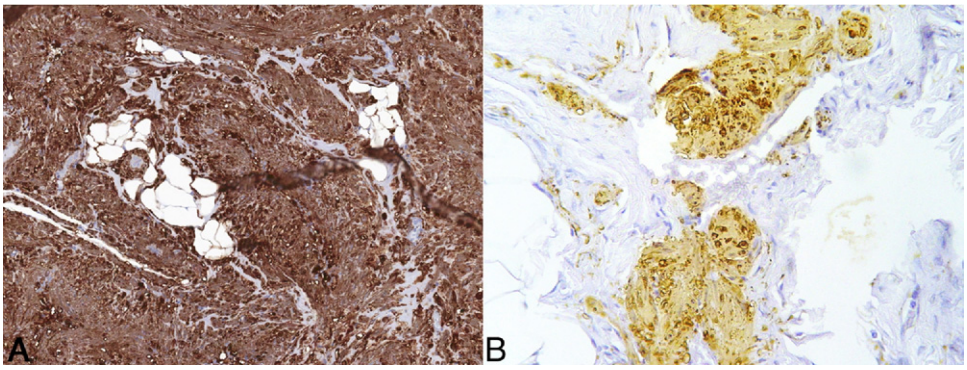

Fig. 3. **A** (Case 1), Smooth muscle bundles positive for actin (ABC-hematoxylin, original magnification ×200). **B** (Case 2), Smooth muscle bundles positive for actin (ABC-hematoxylin, original magnification ×400).

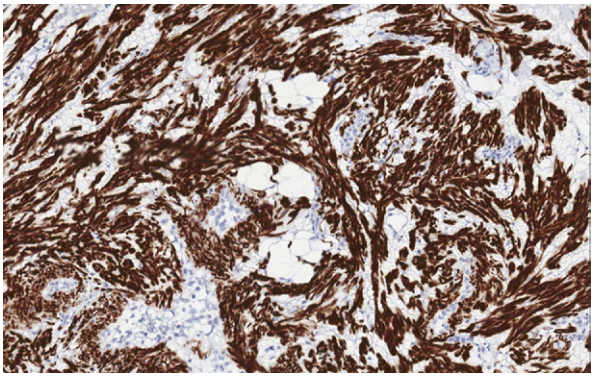

Fig. 4. Smooth muscle bundles positive for desmin (ABC-hematoxylin, original magnification ×200).

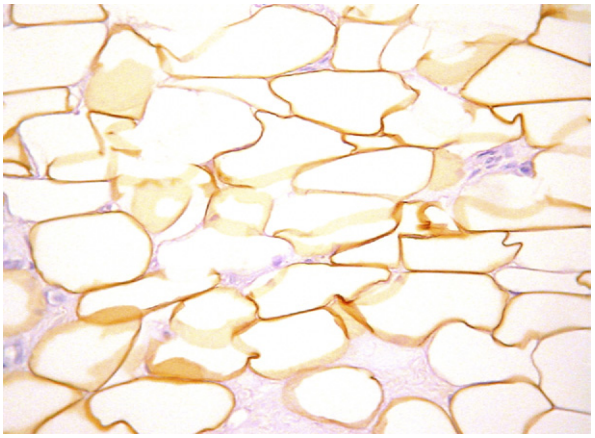

Fig. 5. (Case 2) Adipocytes exhibiting S-100 reactivity (ABC-hematoxylin, original magnification ×200).

sized vessels with muscular walls were seen in the periphery of the lesion. Discrete, small bundles of smooth muscle were close to but not associated with the vascular walls and groups of mature fat cells. Smooth muscles were enhanced by Masson trichrome stain. No epithelioid cells were present. The final diagnosis was angioleiomyolipomatous hamartoma.

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Both cases were studied immunohistochemically using the streptavidin-biotin-peroxidase processing of the Ventana BenchMark XT fully automated slide preparation system (Ventana Medical Systems Inc., Tucson, AZ) and using the iView DAB detection kit (Ventana). Appropriate positive and negative controls were used according to the instructions of the antibodies' manufacturers. In both cases the smooth muscle component was intensely positive for smooth muscle actin (asm1, 1:100; Novocastra, Newcastle upon Tyne, UK) (Fig. 3, *A* and *B*). Case 1 was positive for desmin (DER-11, 1:100; Novocastra) (Fig. 4) and case 2 negative. Adipocytes stained for S-100 protein (polyclonal, 1:100; Novocastra) (Fig. 5), whereas Factor VIII-related antigen (BGX016A, 1:50; Biogenex, San Ramon, CA) outlined the vessels. No reactivity was seen for HMB-45 (HMB45-CE, 1:25; Novocastra) and Melan-A (A103, 1:25; Dako, Carpinteria, CA).

DISCUSSION

Eble[9,p21] has stated that "the name angiomyolipoma is one of the most accurately descriptive (terms) in pathology." Thus, it is reasonable that it is applied to tumors showing a variable proportion of abnormal thick-walled blood vessels, spindle and epithelioid smooth muscle cells, and fat cells. Bonetti et al.[10] in 1994 established renal AML as a member of the PEComa group of tumors that are characterized by their immunoreactivity for both melanocytic and smooth muscle markers, in particular HMB-45 and muscle-specific actin.[1,3,5] Consecutive studies showed that HMB-45 is not diagnostic of renal and extrarenal AMLs, as HMB-45–negative AMLs were found in the

kidneys[9,31-33] and other visceral organs.[34-38] However, other melanocytic markers, i.e., Melan-A/Mart-1, tyrosinase, microophthalmia transcription factor (Mitf), in combination with myocytic markers, should be expressed.[33]

Both our cases featured "angiomyolipomatous" features, but immunohistochemically they were negative for HMB-45 and Melan-A. HMB-45 negativity has been described in all head and neck so-called "AMLs" tested, including oral, as well as cutaneous tumors. As HMB-45 staining intensity, distribution, and pattern of immunostaining may vary depending on the cellular composition of the tumor,[29] some authors[22,26] associate HMB-45 negativity to the scarcity of epithelioid cells in oral AMLs or to site-specific differences.[13,17] The former explanation is, in our opinion, not valid, as spindle cells of lymphangioleiomyomatosis are HMB-45 positive, whereas the latter represents an aphorism lacking scientific merit. It is true that in one example of oral PEComa HMB-45 staining varied, being more prominent in epithelioid cells.[29] However, elongated spindle cells revealed also immunoreactivity with this antibody. The other reported oral example was reportedly HMB-45 negative but featured positivity for Melan-A and CD10.[30]

In case 1, tumor circumscription and growth of the smooth muscle cell component circumferentially to thick-walled blood vessels is consistent with angiomyoma (vascular leiomyoma) with mature fat cells. Fat cells of previously reported "oral AMLs" were also mature. They were found singly[12,18] or in small groups,[11,14,16,18] and anisocytosis was described in one case.[13] Groups of mature adipocytes are not uncommon in angiomyomas. Sixteen of 48 (33.3%) angiomyomas of the head in the study of Hachisuga et al.[39] featured an adipocytic component and although there is only one such description[40] among the more than 100 cases of oral angiomyomas reported so far, it is possible that small groups of adipose tissue have been overlooked, as they should not be of diagnostic importance.

Authors have previously suggested that "oral AMLs"[12,19,29,41] and skin AMLs[42] are, in fact, vascular leiomyomas with a fat component. Absence of fat from the surrounding tissues[14] and "lipoblastlike cells"[12] have been stated as evidence against fat entrapment in vascular leiomyomas. However, adipose tissue is unanimously present in the oral mucosa and submucosa, whereas lipoblastlike cells are not a feature of renal AML. Immunohistochemically, fat cells were S-100 negative in one case of "oral AML" not tested for HMB-45,[16] whereas in 4 other cases, they were S-100 positive.[12,13] In renal AMLs, adipocyticlike cells may infrequently be S-100 positive and it is proposed that S-100(–), HMB-45(+) clear cells may represent PECs undergoing lipoid degeneration.[1,6]

In case 2, the tumor was ill defined and vascular, similar to the angiomyolipomatous hamartoma of the tongue reported by Ide et al.[41] In contrast to the latter, however, the smooth muscle bands were close to, but not associated with, thin-wall capillary-size blood vessels, and medium-sized vessels with thick walls were seen only in the periphery of the tumor. Some authors[11,19] characterize their "oral AMLs" as hamartomas, too, but it should be noted that those publications appeared in an age that renal AML was believed to be a hamartoma. Furthermore, in one of those cases[11] certain microscopic features are more reminiscent of venous or arteriovenous hemangioma with adipocytes and we feel that additional purported examples of "oral AMLs" may represent that entity.

Desmin negativity in some "oral AMLs" was considered indicative of perivascular origin of the smooth muscle component.[12] There are, however, examples of desmin-positive oral "AMLs."[13,17,18] Heterogeneity for desmin is common in smooth muscle cells of blood vessels and is contributed to the antibodies applied,[43] or the size and the anatomic location of the vessels.[44]

Although the diagnoses in our cases are straightforward, for the sake of differential diagnosis the possibilities of angiolipoma and myolipoma could have been considered. In angiolipoma, mature adipose tissue intermingles with thin-walled capillary-size vessels, as seen in case 2.[45] A few cases have been reported in the oral cavity, but in none of them was there a smooth muscle component. Also, fibrin clots, considered diagnostic in angiolipoma, were not found in our case.[45] As far as myolipoma is concerned, a rare oral example has been reported.[46] Such tumors are composed of mature smooth muscle and adipose tissues. However, they lack the medium-sized thick-walled blood vessels seen in case 1, whereas in contrast to case 2 the smooth muscle component is dominant and arranged in short fascicles of slender smooth muscle cells forming a sievelike pattern with blending groups of mature adipocytes.[47]

Apart from the stated differences, and with the exception of a poorly documented alleged example arising in the lip of a 55-year-old woman,[15] no reported cases of so-called oral AML have been associated with tuberous sclerosis complex. All tumors have behaved in a benign fashion, without recurrence after conservative excision. The vast majority of renal AMLs are benign neoplasms. However, they can predispose to life-threatening complications, the most significant being hemorrhage. Extrarenal AMLs reported in a variety of tissues and organs, including retroperitoneum, mediastinum, liver, adrenals, ovary, and lungs, are uncommon and can cause complications including hemorrhage and hy-

povolemic shock, and interference with major vessels as well as problems in surgical management.

In conclusion, it is our opinion that the term AML has been used deliberately in the oral and maxillofacial pathology literature to describe lesions characterized by smooth muscle or angiomatous proliferations with an adipocytic component. Such cases are either vascular leiomyomas, venous/arteriovenous hemangiomas, or hamartomas. More importantly, many cases of oral "AML"[11-14,17,18] have been discussed as counterparts of renal AML, a form of PEComa, although a myomelanocytic immunophenotype was not confirmed. Thus, we fully agree with Hashimoto and Quade[48] who discourage the use of the term AML for angiomyomatous lesions with adipocytes, as there is no evidence of any relationship of such lesions with renal or retroperitoneal AML, or with tuberous sclerosis. The same argument applies against the use of terms such as mucocutaneous angiomyolipoma and angiolipoleiomyoma.[20] More importantly, because a diagnostic term well known to clinicians and pathologists confers information relevant to the character, prognosis, and treatment of a lesion, we opine that the term AML be abandoned for non-PEComatous oral lesions because it is misleading.

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