Angiomyxolipoma (Vascular Myxolipoma) of the Oral Cavity. Report of a Case and Review of the Literature

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Abstract Angiomyxolipoma is a rare tumor characterized by a proliferation of adipose tissue associated with a myxoid stroma and multiple vascular channels. To date, ten cases of angiomyxolipoma located in subcutaneous tissue, spermatic cord and subungual area have been reported in the literature. We describe the clinical and histopathologic findings of the first case of intraoral angiomyxolipoma in a 12 year-old male, as well as the utility of immunohistochemistry as a useful tool in differential diagnosis of this particular lesion with respect to other benign and malignant lesions derived from adipose tissue.

Keywords Lipoma · Angiomyxolipoma · Myxolipoma · Immunohistochemistry

Introduction

In the oral cavity, lipomas are the most common mesenchymal neoplasms and based on histology classified as classic, spindle cell/pleomorphic, fibrolipoma or chondroid lipomas [1, 2]. Angiomyxolipoma was first described by Mai et al. [3] as a rare variant of lipoma, characterized by a variable amount of mature adipocytes associated with myxoid areas, as well as numerous vascular channels. To our knowledge, this is the first case report of an angiomyxolipoma of the oral cavity, and the aims of this article are to present the salient clinicopathologic features, to discuss its differential diagnosis and to describe its management and follow-up.

Case Report

A 12-year-old male was originally seen in the Hospital Juárez of México City in 2004 due to an asymptomatic swelling located in the right buccal mucosa (Fig. 1). Clinically it presented as an irregular, firm and multilobulated submucosal mass which produced no changes in color or texture of the overlying mucosa and skin (Fig. 2a). Under local anesthesia, the patient underwent total excision (enucleation) of the encapsulated multilobulated lesion (Fig. 2b). The excised specimen measured 60 × 45 × 23 mm and on cut surface, had a yellow colored and appeared solid and well defined. Formalin-fixed, paraffin embedded representative sections were stained with H&E and immunohistochemical studies were performed using the avidin-streptavidin-peroxidase method with 3–3'diaminobenzidine as the chromogen; the antibodies applied in present case were Vimentin (clone Vim 3B4, dilution 1:400, Dako A/S Glostrup Denmark), Alpha Smooth

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Fig. 1 Extraoral view: note swelling of the *right side*

Muscle Actin-SMA (clone 1A4, dilution 1:400, Dako A/S Glostrup Denmark), Ki-67 (clone MiB-1, dilution 1:100, Dako A/S Glostrup Denmark), S-100 protein (dilution 1:10,000, Dako A/S Glostrup Denmark), CD-68 (clone MII5, dilution 1:400, Dako A/S Glostrup Denmark), Melanosome (clone HMB-45, dilution 1:200, Dako A/S Glostrup Denmark) and CD-34 (clone QBEnd10, dilution 1:50, Dako A/S Glostrup Denmark).

Histologically, the specimen consisted of a combination of mature adipocytes and bland spindle cells scattered within a myxoid stromal background containing abundant small to medium sized vascular channels (Fig. 3a, b). Some adipocytes presented with an eccentric nucleus and slightly vacuolated cytoplasm, which resembled lipoblasts. No

necrosis, chondroid metaplasia, mitoses or hemorrhage was observed. The proportion of adipocytes to fibrous and myxoid background varied through different areas of the tumor (Fig. 3c). Immunohistochemical analysis revealed positivity of cells for vimentin in different intensities in stromal cells, as well as a weak reaction for S-100 protein in adipocytes. CD-34 and SMA disclosed numerous vascular structures, but it was non-reactive in tumor cells (Fig. 3d). CD68, SMA, HMB-45 and Ki-67 reactions were negative. After 4 years of follow-up, there has been no recurrence (Fig. 4).

Discussion

Lipomas are the most common mesenchymal lesions of the oral region [1]. According to the series of 125 lipomas reported by Furlong et al. [2], the most frequent types found were the classic type and the spindle cell/pleomorphic lipoma (SCL) (49.6 and 47.2%, respectively), while the other variants comprised around 3.2%. In 1996, Mai et al. [3] reported a case of a rare variant of lipoma of the spermatic cord in a 34 year-old man, characterized by a proliferation of adipocytes associated with a myxoid background and multiple blood vessels and designated it as angiomyxolipoma. Since then, ten cases of this rare variant have been reported in the English literature, [3–12] which have been located mainly in subcutaneous tissues (eight cases), one case occurred as subungual angiomyxolipoma and the other developed in the spermatic cord. Our patient is the first case of angiomyxolipoma originated within the oral tissues (Table 1).

Histological differential diagnosis of angiomyxolipoma includes benign lesions, such as SCL and its variants (myxoid, vascular, pseudoangiomatous), dendritic fibromyxolipoma, myxolipoma, angiolipoma, superficial angiomyxoma, angiomyofibroma, cellular angiofibroma, lipoblastoma, benign mesenchymoma as well as aggressive

Fig. 2 **a** Clinical view of the multilobulated intraoral swelling within the right cheek mucosa. **b** Multilobulated aspect of the surgical specimen

Fig. 3 **a** Mature adipocytes and bland spindle cells scattered within a myxoid stromal background containing abundant small to medium sized vascular channels. **b** Fibrous areas surrounding scattered foci of

adipocytes and myxoid background. **c** Numerous thin-walled blood vessels scattered in a myxoid stroma. **d** SMA immunohistochemistry disclosed abundant vascular channels within the lesion

Table 1 Clinical features and immunoprofile of reported cases of angiomyxolipoma

Case	Ref	Sex/age	Location	Duration	Immunohistochemistry						
					CD34	Vimentin	S100	SMA	Desmin	HMB45	Ki67
1	Mai et al. [3]	M/32	Spermatic cord	3 mo	NP	+ (S,A)	+ (A)	+ (V)	–	–	NP
2	Zamecnik [4]	M/57	Scalp	NS	+S	NP	NP	+ (V)	NP	–	NP
3	Sciot et al. [5]	F/60	Thigh	4 mo	+ (V,S)	+ (S,A)	+ (A)	+ (V)	NP	–	NP
4	Tardío et al. [7]	M/66	Scalp	NS	+ (V,S)	+ (S,A)	+ (A)	+ (V)	–	–	NP
5	Lee et al. [8]	M/44	Arm	7 years	+ (V,S)	+ (S,A)	+ (A)	+ (V)	–	NP	–3%
6	Lee et al. [8]	M/57	Wrist	2 years	+ (V,S)	+ (S,A)	+ (A)	+ (V)	–	–	–3%
7	Sánchez et al. [9]	M/43	Subungual area	1 years	NP	NP	NP	NP	NP	NP	NP
8	Kang et al. [10]	M/38	Gluteal area	3 years	+ (V,S)	+ (S,A)	+ (A)	+ (V)	–	–	NP
9	Song et al. [11]	M/69	Hip	3 years	+ (V,S)	+ (S,A)	+ (A)	+ (V)	–	–	NP
10	Kim et al. [12]	M/9	Thigh	5 years	+ (V,S)	+ (S,A)	NP	NP	NP	NP	NP
11	Present case	M/12	Buccal mucosa	4 years	+ (V)	+ (S,A)	+ (A)	+ (V)	NP	–	–

SMA smooth muscle actin, M male, F female, NP Not performed, S spindle cells, A adipocytes, V vascular structures, NS not specified, Modified from Song et al

angiomyxoma; malignant lesions to be discarded includes myxoid liposarcoma and low-grade myxofibrosarcoma [8]. Histological features, such as presence/absence of vascular structures, spindle cells and pleomorphic cells (floret-like cells) are diagnostic criteria for differential diagnosis of angiomyxolipoma from lesions like SCL. According to

previous reports, angiomyolipoma is composed of a significant number of smooth muscle cells and lack the myxoid component present in angiomyxolipoma [13]. The absence of necrosis, cellular pleomorphism, lipoblasts and atypical mitoses observed within adipose tissue may aid in differentiating angiomyxolipoma from malignant lesions,

Fig. 4 Postoperative clinical view; no recurrence was observed after 4 years of follow-up

such as myxoid liposarcoma and low-grade myxofibrosarcoma [3, 8, 15].

The adipocytes demonstrated immunoreactivity for S-100 protein, however the stromal cells (fibroblastic component) were negative, a finding consistent with the cases previously reported by Suster and Fisher [14], Nascimento et al. [15] and Fregnani et al. [16]. Negativity immunoreactivity for CD-34 in spindle cells and absence of giant cells and mast cells in the present case discarded the possibility of a spindle cell lipoma, but it did highlight multiple vascular structures associated with the spindle cells and myxoid stroma [17, 18]. Low index of expression of Ki-67 (<1%) in our case was important in distinguishing angiomyxolipoma from myxoid liposarcoma and lipoblastoma; on the other hand, negative stain for HMB-45 and SMA discarded the possibility of an angiomylipoma [13]. After 4 years of follow-up, no recurrence has been observed in this case, confirming the benign nature of the lesion.

In summary, we present the first case of angiomyxolipoma reported in the oral region, a benign uncommon lesion characterized by a proliferation of vascular vessels, prominent amount of mature and immature adipocytes associated with an abundant myxoid component, and describe the importance of differentiating this entity from other benign and malignant lesions that contain adipose tissue, such as liposarcoma, lipoblastoma, benign mesenchymoma and pleomorphic cell lipoma.

Competing interests None.

Ethical Approval Not required.

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