

## CASE REPORT

## Non-infiltrating angiolipoma of the upper lip: A rare entity

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### ABSTRACT

Angiolipoma is a variant of lipoma with a prominent vascular component. It is the most common tumor of the forearm followed by trunk and extremities of young individuals. Its occurrence in head and neck region is rare. In this report, we present clinical and histopathologic features of non-infiltrating angiolipoma occurring on the upper lip in a 9-year-old female. Toluidine blue stain is used to demonstrate increased mast cell density around blood vessels, speculating its role in vasculogenesis.

**Key words:** Angiolipoma, lip, mast cell, swelling, toluidine blue

### INTRODUCTION

Angiolipoma (AL) is a relatively rare tumor of the head and neck region, although it occurs more commonly in the forearm and the trunk regions. This tumor has been rarely reported in the oral cavity, and to best of our knowledge, it has only been reported twice in the lip.<sup>[1]</sup>

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.<sup>[2]</sup> An association between mast cells roles and increased vascularity has been speculated.<sup>[3]</sup> We present, a rare case of non-infiltrating angiolipoma occurring on the upper lip in a 9-year-old female. Toluidine blue staining was done to demonstrate mast cell in tumor tissue.

### CASE REPORT

A 9-year-old female presented with an asymptomatic swelling of upper lip since birth. She complained of a slight increase in size of swelling for the past 1 month. There was no history of trauma. Family, medical and dental history was unremarkable.

Clinical examination revealed a swelling of 2 × 2 cm in diameter at the midline of the upper lip [Figure 1]. Swelling was well circumscribed, soft in consistency, movable, non-tender, non-fluctuant, and no bruit was present. Overlying skin was

normal. No other abnormality was detected extraorally or intraorally. The mass was dissected free of the adjacent tissue and the lesion was surgically removed under local anesthesia. On gross examination, the mass was multilobulated, grayish white in color, soft in consistency and measuring about 2 × 2 cm in diameter [Figure 2].

Histopathologically, H and E stained section under low magnification revealed an encapsulated tumor mass consisting of adipose tissue separated by branched vascular network [Figure 3]. Thin walled small capillary like vessels were present at the periphery of the tumor mass [Figure 4]. Under higher magnification numerous mature adipocytes with variable sized endothelial lined blood vessels were seen [Figure 5]. Few areas show presence of fibrinous microthrombi [Figure 6]. Toluidine blue staining showed an increased mast cells density in and around blood vessels [Figure 7] with degranulation [Figure 8]. A diagnosis of non-infiltrating angiolipoma was made. Postoperative course of the patient was favorable. There were no sign or symptoms of recurrence 6 months after surgery.

### DISCUSSION

It is widely assumed that benign lipomatous tumors represent a common group of neoplasm's that cause few complaints or complications and present little diagnostic difficulty.<sup>[4]</sup> Lipomas are the most common neoplasm arising from fat tissue. Thirteen percent of all lipomas occur in head and neck, including cheek, tongue, palate, parotid gland, neck and larynx.<sup>[1]</sup> However, only 1%–4% of cases involve the oral cavity. Oral lipoma represents 0.5%–5% of all benign neoplasms of oral cavity. They are usually asymptomatic, slow-growing, soft and well-circumscribed submucosal or superficial lesion, mainly located on the buccal mucosa.<sup>[5]</sup>

The bulk of lipomatous tumors may be grouped into four categories: Superficial lipoma, a tumor composed of mature fat and arising in the superficial (subcutaneous) soft tissues,

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represents by far the most common mesenchymal neoplasm. Deep lipomas arise from or are intimately associated with tissues deep to the subcutis or with specific anatomic sites. The main subdivisions of this group are angiomyolipoma,

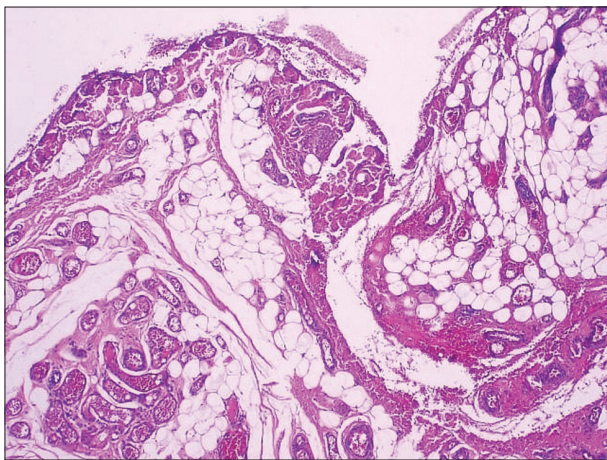
intramuscular and intermuscular lipoma, lipoma of the tendon sheath, neural fibrolipoma with or without macrodactyly and lumbosacral lipoma. Infiltrating or diffuse neoplastic or non-neoplastic proliferations of mature fat may cause



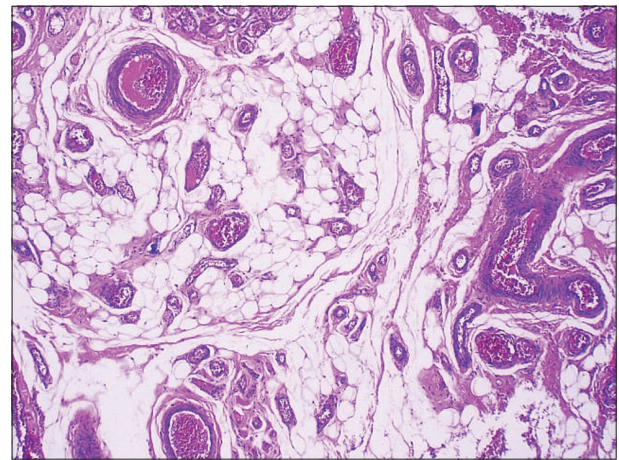
**Figure 1:** Clinical photograph of patient



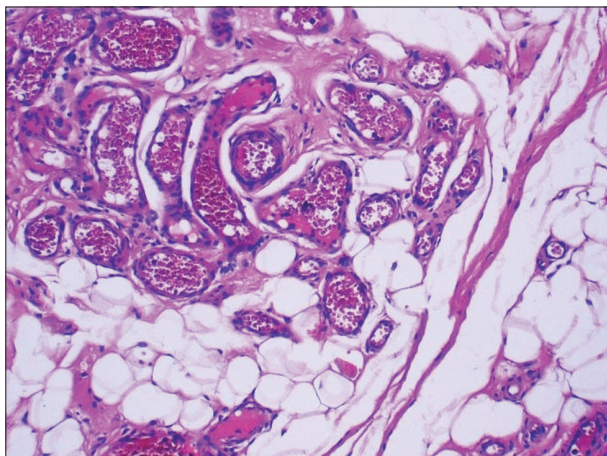
**Figure 2:** Gross specimen showing multilobulated mass



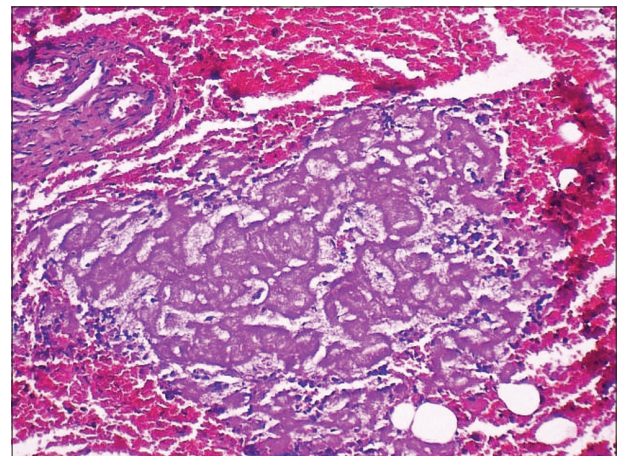
**Figure 3:** Encapsulated tumor mass consisting of adipose tissue separated by branched vascular network (H and E stain, original magnification,  $\times 10$ )



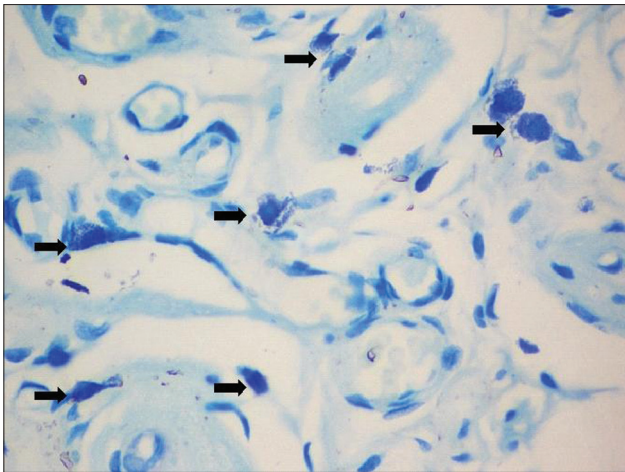
**Figure 4:** Numerous mature adipocytes with variable sized endothelial lined blood vessels (H and E stain, original magnification,  $\times 10$ )



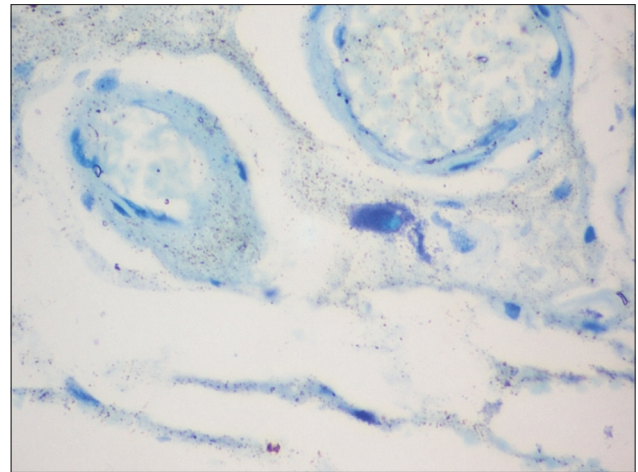
**Figure 5:** Thin walled capillary like vessels present at the periphery of the tumor mass (H and E stain, original magnification,  $\times 40$ )



**Figure 6:** Presence of fibrinous microthrombi (H and E stain, original magnification,  $\times 40$ )



**Figure 7:** Tumor tissue showing increased mast cells density in and around blood vessels (toluidine blue stain, original magnification ×40)



**Figure 8:** Tumor tissue showing degranulating mast cell in and around blood vessels (toluidine blue stain, original magnification ×40)

compression of vital structures or may be confused with atypical lipomatous neoplasm/well-differentiated liposarcoma. This group is composed of six entities: Diffuse lipomatosis, pelvic lipomatosis, symmetric lipomatosis, adiposis dolorosa, steroid lipomatosis, and nevus lipomatosus. Variants of lipoma are much less common and differ from ordinary lipoma by characteristic microscopic picture and specific clinical setting. These include angioliipoma (AL), myoliipoma, angiomyoliipoma, myeloliipoma, chondroid lipoma, spindle cell/pleomorphic lipoma, hibernoma and lipoblastoma/lipoblastomatosis.<sup>[4]</sup>

AL is a benign mesenchymal tumor made up of mature lipocytes and proliferating blood vessels. These tumors were originally described by Howard and Helwig in 1960.<sup>[6]</sup> They are benign subcutaneous lesions most common in young male patients in their second or third decades of life.<sup>[7]</sup> They can be multiple in nature, and are most commonly seen on forearm (two-third of cases) followed by the trunk and upper arm. Only 38 cases of head and neck ALs have been reported.<sup>[7]</sup> To the best of our knowledge there are only two cases of AL of the lip reported in the English literature. When seen in the oral cavity, it has been noted to be on the lip, tongue, palatal tissue, cheek being the most common site.<sup>[1,2]</sup> In a review of AL of head and neck, Alvi *et al.*<sup>[8]</sup> mentioned certain distinct characteristics which are present between head and neck AL and non-head and neck AL [Table 1].

Differential diagnosis of a soft mobile mass in the head and neck, and especially in the upper lip, should alert the clinician to consider other entities such as canalicular adenoma, basal cell adenoma, pleomorphic adenoma, angioleiomyoma, schwannoma, neurofibroma, other benign mesenchymal tumors and xanthogranuloma.<sup>[2]</sup>

Pathogenesis of this lesion remains unclear. Many suggest trauma as a possible etiologic factor. Increased familial incidence has also been reported. But the origin of angioliipoma

**Table 1: Characteristics of hand neck versus non-head and neck AL**

Features	Head and neck AL	Non-head and neck AL
Pain	(-)	(+)
Pubertal age	(-)	(+)
Sex predilection	(-)	(+/-)
Family history	(-)	(+/-)
Multiple tumors	(-)	(+)
Trauma	(-)	(+)

AL - Angioliipoma

is still controversial. Different theories suggest that a lipoma differentiates because of some unknown stimulus; the tumor is of neurogenic origin; and it is congenital. Possible causes include fatty metamorphosis of a central hemangioma, hyperplasia of fat with an associated increase in vascular channels, or a true neoplasm. There is support for the theory that an AL originates as a congenital lipoma, which later undergoes vascular proliferation. Howard and Helwig think that embryonic sequestration of multipotential cells become activated at puberty by hormones and differentiate into a simple lipoma. Further stimuli such as trauma can cause vascular infiltration of the lesion, but trauma is not present in many cases of AL.<sup>[6,7]</sup>

Mast cell has been thought to mediate angiogenesis in various pathophysiological conditions. It has been speculated that mast cells might play a role in their increased vascularity. Based on the data of Ida-Yonemochi *et al.*<sup>[3]</sup> AL have 10 times the number of mast cells than classic lipomas. Mast cells around blood vessels strongly express vascular endothelial growth factor (VEGF) in angioliipoma, which is known to be an essential growth factor for endothelial cells in angiogenesis.<sup>[3]</sup> In the present case, toluidine blue staining showed an increased mast cells density around blood vessels, with degranulation, speculating its role in vasculogenesis. This was consistent with the findings of Ida-Yonemochi *et al.* However, there has been no direct demonstration of molecular mechanisms for angiogenesis participation by mast cells in angioliipoma.<sup>[3]</sup>

Based on studies by Gonzales-Crussi *et al.*<sup>[9]</sup> AL has two histologic types: Infiltrating and non-infiltrating. The non-infiltrating type is the most common. It presents as painless or tender subcutaneous nodules, generally in pubescent patients and is rare before puberty. Non-infiltrating AL occurs in multiple sites in 79% of cases. Histologically it is encapsulated, and is a mixture of mature adipocytes and a proliferation of thin-walled vascular channels. Infiltrating ALs are usually diagnosed in older patients. They extend into the surrounding tissue and are characterized by a non-encapsulated tumor mass. They have two anatomical forms: Intermuscular and intramuscular. Microscopic examination is necessary for conclusive diagnosis.<sup>[9]</sup> The present case was that of non-infiltrating AL of 2 × 2 cm in diameter located in the upper lip of young female. Following are the current histologic guidelines for diagnosis of AL:<sup>[7,9]</sup>

1. Well encapsulated (non-infiltrating ALs) or poorly encapsulated (infiltrating ALs).
2. Evidence of 50% mature adipocytes in the tumor.
3. Interspersed angiomatous proliferation in the tumor.
4. Fibrinous microthrombi.
5. Absence of other mesenchymal elements (smooth muscle) or pleomorphism.

Histopathological differential diagnosis includes hemangioma, lipoma, angiomyolipoma, infiltrating lipoma, angiofibrolipoma, angiomyxolipoma, and liposarcoma. Hemangioma, there is no lipomatous component. Angiomyolipoma is composed of varying amounts of blood vessels, smooth muscles and fat cells. Infiltrating lipoma consists of lesional fat tissue infiltrating in the deeper tissue, in the form of long thin streaks radiating from the intratumoral mass. Angiofibrolipoma is composed of varying amounts of blood vessels, fibrous tissue and fat cells. Angiomyxolipoma is composed of adipose tissue component with myxoid changes and blood vessel proliferation. It is the most common benign tumor of the kidney and is strongly associated with tuberous sclerosis.<sup>[10]</sup> The hypovascular lesions may be difficult to distinguish from ordinary lipomas, although the identification of microthrombi allows this distinction.<sup>[7]</sup> Liposarcoma can be confused with AL but can usually be differentiated by the presence of embryonal adipose tissue, pleomorphism, increased number of mitosis, and metastasis.<sup>[8]</sup>

The AL has not been shown to spontaneously regress and has been shown to continue to enlarge as opposed to other entities such as a hemangioma. The treatment of choice for a non-infiltrating AL is surgical excision, whereas a wide local excision with free margins has been advocated for the infiltrating type. In the case of inadequate resection, radiation therapy may be necessary.<sup>[11]</sup> The recurrence of AL has been reported to be dependent on whether the mass is infiltrating. Infiltrating ALs have been reported to be more common in an older age group. Their recurrence has been reported to be as high as 62.5% in areas outside the head and neck. Noninfiltrating types are seen more commonly in a younger age group and have no tendency to recur.<sup>[1]</sup> A successful

treatment of a giant infiltrating angioliipoma with interferon alfa also has been reported.<sup>[10]</sup> ALs reported in the oral cavity have not been reported to recur.<sup>[2]</sup>

## CONCLUSION

In conclusion, lipomas represent about 1%–4% of all neoplasm's of the oral cavity. They are usually asymptomatic, slow-growing, soft and well-circumscribed submucosal or superficial lesion. ALs are rarely seen in head and neck region. It consists of mature adipocytes and interspersed connective tissues with vascular vessels containing fibrin thrombi and mast cell infiltration. Histopathological evaluation is mandatory to confirm the diagnosis. An association between mast cell and vasculogenesis has been speculated. Wide surgical excision with free margins is the treatment of choice, offering a good prognosis and an almost negligible relapse rate.

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