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An unusual lesion on the nose: microvenular hemangioma

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ABSTRACT Microvenular hemangioma (MVH) is an acquired, benign type of hemangioma that usually manifests itself as a solitary, slowly growing, red to violaceous, asymptomatic papule, plaque or nodule. It is typically located on the trunk or extremities of young adults. It can be difficult to differentiate MVH from other types of hemangioma and Kaposi sarcoma. Herein we report a case of MVH unusual for its location, age of onset, and morphologic features.

A 62-year-old man complained of an asymptomatic, bluish-red discoloration on the tip of his nose that had been present for two years. Dermatologic examination showed a violaceous patch 2 x 2 cm in diameter with indistinct borders. Incisional biopsy revealed irregularly branched small or medium-sized vascular spaces lined with benign endothelial cells, positive for CD34 and negative for HHV-8.

MVH is a rare lesion, and less than 70 cases have been published to date. A review of 40 reported cases revealed that 15% of MVH patients were over 40 years of age and only 3% of the cases showed macules or patches. A literature survey showed only two cases of MVH located on the facial region, one on the chin and the other on the cheek. Our case was unique for its location and interesting for other rarely encountered features. MVH should be considered in the differential diagnoses of vascular lesions on nasal skin.

Introduction

Microvenular hemangioma (MVH) is an acquired benign type of hemangioma with distinctive histological findings, first named in 1991 [1]. It is a rare lesion, and less than 70 cases of MVH have been published to date [2]. It usually presents with an asymptomatic, slowly growing reddish-blue papule, plaque, or nodule, less than 30 mm in diameter [1,2]. Herein, we describe an adult male with MVH, presenting unique and atypical clinical features.

Case Report

A 62-year-old man presented with a nontender, bluish-red area on his nose, present for two years. The lesion had gradually enlarged and deepened in color during the previous year.

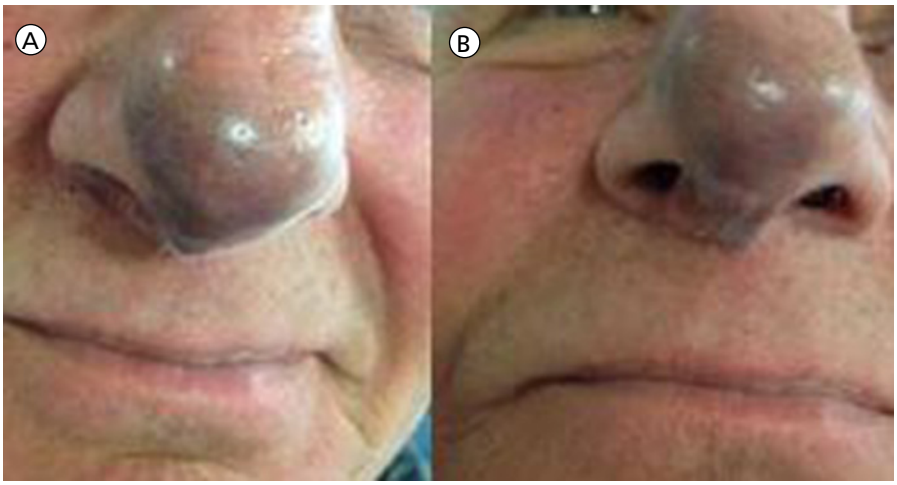

Figure 1. The lesion is a poorly circumscribed, bluish-red patch involving the tip of the nose (a), and columella (b). [Copyright: ©2018 Mansur et al.]

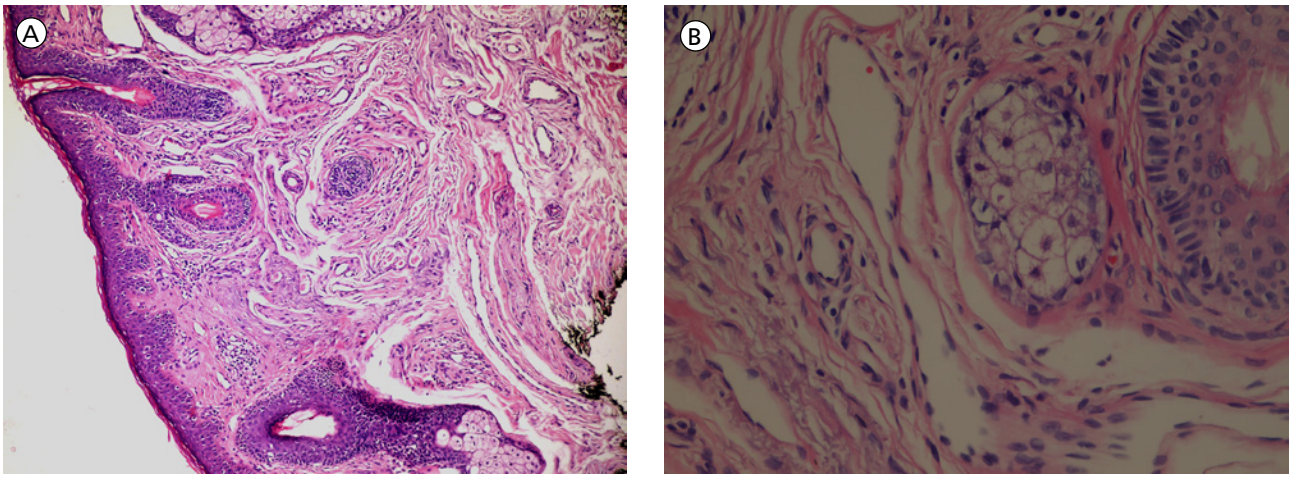

Figure 2. (a, b) Histopathologic features consisted of a proliferation of irregular vascular channels involving the dermis (hematoxylin-eosin stain; original magnification ×200). [Copyright: ©2018 Mansur et al.]

The patient had several comorbidities including hypertension, nodular goiter, erosive gastritis, renal calculi, cervical discopathy, gonarthrosis, and patellofemoral arthrosis. His medications included esomeprazole, isosorbide dinitrate, nifedipine, betahistine dihydrochloride, acemetacin, and chlorzoxazone. He had quit smoking two years prior and denied drinking alcohol. He had blue eyes, and skin type II. Cuperosis in the nasal lesion was considered by previous physicians, and sunscreen lotions were prescribed with no beneficial effect.

Physical examination revealed facial erythema and prominent telangiectasia on both cheeks. A bluish-red, partially blanchable patch with indistinct borders, measuring 2 cm in diameter, was seen on the tip of the nose and columella. Superficial and thin blood vessels were noticed throughout the patch (Figure 1a, b). Anterior rhinoscopy of the nose and otolaryngological examination was normal. An incisional biopsy was performed with a clinical prediagnosis of Kaposi sarcoma, hemangioma, pseudolymphoma, and angiolupoid type of cutaneous sarcoidosis.

Histologically, hyperkeratosis, irregular acanthosis, and minimal papillomatosis were observed in the epidermis. Small and medium-caliber, irregularly branched vessels, lined by a single layer of benign endothelial cells were distributed in the upper dermis. Between the blood vessels, increased fibrous tissue and sparse lymphoplasmacytic inflammatory infiltration were noticed (Figure 2 a, b). Immunohistochemically, the cells lining the lumina were positive for CD34 (Figure 3 a,b). HHV-8 immunostaining was negative for vascular lesions. Based on these findings, MVH was diagnosed.

Discussion

A review of 40 cases of MVH revealed that MVH mostly manifested itself with nodules, and less frequently with papules or plaques. Only a minority (3%) of the cases showed macules or patches [3].

MVH is typically located on the extremities and trunk and rarely the neck and face can be involved. The review men-

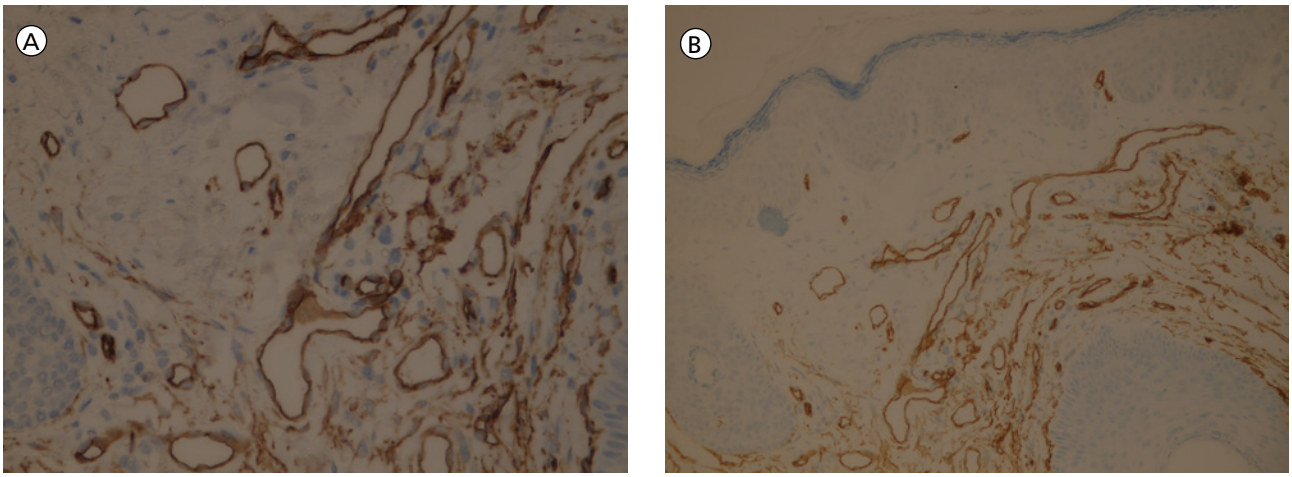

Figure 3. (a) Here, vascular spaces are clearly visible with endothelial cells positive for CD 34. (b) Vascular spaces are visible with endothelial cells positive for CD31. [Copyright: ©2018 Mansur et al.]

tioned above showed that 59% of the lesions were located on the trunk, 56% on the extremities, and only 10% on the head and neck region [3]. In cases where MVH was located on the head, lesions appeared on the chin and cheek [3-5]. To date, there is no report of MVH involving the nose.

In most cases, lesions occur in young to middle-aged adults, with equal incidence in males and females. Only 15% of MVH cases occur in patients older than 40 years. In the beginning, MVH shows a rapid growth in three months, then the lesion becomes stable or continues growing only slowly. The factors that predispose, trigger, or facilitate the development of MVH are not known yet [1-6].

Histologically the tumor is composed of irregularly branched, thin-walled small blood vessels of uniform diameter, infiltrating the superficial and deep dermis, surrounded by a collagenous or desmoplastic stroma. The endothelial cells are normal to plump and display no atypia, mitotic figures, or pleomorphism. The vascular lumina are narrow but recognizable with a few red blood cells inside, without any extravasation. There may be a mild lymphoplasmacytic infiltrate [5,6].

MVH expresses several vascular markers, such as CD34, CD31, WT1, VIII-related antigen, vWF, or UEA-1, confirming the endothelial origin of the tumor [7].

Clinically MVH can mimic other benign and malignant vascular lesions. Even some nonvascular tumors and inflammatory lesions, including leiomyoma, dermatofibroma, leukemia and lymphoma with cutaneous involvement, lymphomatoid papulosis, papulonecrotic tuberculoid, and bacillary angiomatosis may be listed in the differential diagnosis [2,6].

The correct diagnosis of MVH cannot be made based on clinical features in most patients, but it can be established with routine histology and immunohistochemistry. The most important vascular lesions that must be considered are Kaposi sarcoma (KS) and angiosarcoma, since some morphological similarities between these entities have been reported [2,6-8]. HHV8 is an important clue that helps to distinguish MVH from early stages of KS, since KS expresses the marker, while MHV does not [9]. KS and some other vascular lesions were taken into account for the differential diagnosis of MVH and are listed in Table 1.

Our patient's histologic and immunohistochemical findings were diagnostic of MVH, while his clinical manifestation was unique in its localization. In addition, our case was interesting in that the onset of MVH was in advanced age and it presented with a patch type lesion. Our report adds MVH to the broad list of the lesions that could be located on the nose. MVH should be considered in the differential diagnoses of vascular lesions in this area.

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TABLE 1. Clinical Differentiation of MVH from Cutaneous Vascular Tumors

	Clinical Features / Course / Prognosis	Histopathological Features	MVH Differential Points
Cutaneous Angiosarcoma	• Purplish-red, ill-defined bruise-like patch • Bluish-violaceous nodule or plaque that may bleed or ulcerate • Mostly on the head and neck in elderly people • Rapidly growing • High rate of recurrence and metastasis	• Involve dermis extensively, sometimes with subcutis and fascia • Irregular, dissecting, anastomosing vascular channels • Tumor cells pile up along vessel lumina	• Absence of a pericyte layer • a more disordered architecture • Prominent cytologic atypia with large cells, hyperchromatic and pleomorphic nuclei • High mitotic activity
"Kaposi Sarcoma	• Brown-red macules/papules bluish-purple nodules or plaques • Any location but typically on legs/feet, head and neck • Variable extention and course due to immune status • May involve oral mucosa, lymph nodes, viscerae	• Proliferation of spindle cells • Prominent slit-like vascular spaces extravasated red blood cells • Perivascular lymphocytes and plasma cells • Eosinophilic hyaline globules	• More architectural complexity • Absence of conspicuous pericyte layer • Anastomosing vascular spaces • Ectatic vascular channels surrounding the normal blood vessels (promontory sign) • IHC: HHV-8 (+)
Hobnail Hemangioma (targetoid hemosiderotic hemangioma)	• Violaceous central papule surrounded with palor and brown ring • Trunk and limbs • In young and middle aged people • Not a true neoplasm, instead reactive process	• Dilated superficial dermal vessels • Plump, "hobnail" endothelial cells that protrude to lumina	• Inflammation and fibrosis • Extravasated red blood cells Hemosiderin deposition Lymphangiectases • IHC: lymphatic markers (+); (D2-40)
Tufted Angioma	• Firm, dark-red, brownish or violet plaques/nodules • Mostly on trunk, neck and extremities • More frequent in infants and children • Benign, slow and progressive growth	• Tightly packed "tufted" capillaries in discrete lobules (cannonball appearance)	• Distinctive nodular growth pattern • Semilunar clefts at periphery of the lobules • IHC: lymphatic markers (+); (D2-40)
Pyogenic Granuloma	• Red-brown polypoid or pedunculated papule/nodule • Friable, prone to bleed • Gingiva, lips, finger, face • Usually in children and young adults • Evolves over weeks • Benign, may recur after treatment	• Multiple lobules of closely packed capillaries • Loose, edematous stroma • Mixed inflammatory infiltrate	• Well-developed collarette from elongated rete ridges • Fibrous connective tissue septae
Reactive Angioendotheliomatosis	• Red patches/plaques tumors/ ulcerated lesions • Any site on body • Coexistent systemic disease	• Intravascular proliferation of endothelial cells • Dilated vessels • Mild atypia • Minimal inflammation	• Fibrin thrombi • Reactive (fasciitis-like) dermal alterations

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