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Editorial
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Angiolymphoid Hyperplasia with Eosinophilia with Uncommon Location Successfully Treated with Cryotherapy

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Editorial

Angiolymphoid Hyperplasia with Eosinophilia (ALHE), also known as epithelioid hemangioma, is an uncommon benign vascular proliferation usually seen in young to middle-aged women characterized by a prominent eosinophil-rich mixed inflammatory infiltrate [1]. Most often, it occurs in the skin of head and neck and especially in the retro auricular region. We present a case of ALHE in a young man with atypical location.

A 27-year-old male with no medical history with an itchy and nodular ulcerated lesion on the right ankle evolving for 3 months. Physical examination revealed a 10 cm nodular tumor with infiltrated raised borders and central crater covered by crusts (Figure 1). These were no regional lymph nodes. The patient was otherwise healthy. The proposed diagnosis was cutaneous leishmaniasis. It was eliminated by negative parasitological examination and PCR. A biopsy was performed. It showed a vascular proliferation of the dermis composed of small turgescent vessels accompanied by a polymorphic inflammatory infiltrate rich in eosinophils, lymphocytes and macrophages arranged in lymphoid follicles (Figure 2a). Laboratory investigation including complete blood count and kidney function tests were within normal ranges. The diagnosis of AHLE was made. The patient was treated by cryotherapy once a week for one month. A complete regression of skin lesions, without any recurrence during four years of follow-up was achieved (Figure 2b).

ALHE is a rare benign tumor. It occurs in adult women aged between 20 and 50 years [2]. It seems to have a higher incidence in women victims of traumatism or during pregnancy. It is manifested clinically as a reddish bleeding papulonodular infiltration measuring about 2 cm to 3 cm. The lesions occur preferentially on the face, scalp, auricular region, and neck. An underlying arteriovenous shunt might be found. Our case is atypical with the large size and the site of the disease. On reviewing other sites reported in literature, we found one case of ALHE on the thigh and four cases of AHLE on the penis [3]. Our case is the first case of ALHE on the foot.

Histopathology reveals a diffuse inflammatory infiltrate consisting primarily of eosinophils and lymphocytes with nodular architecture together with turgescent vessels protruding into the lumen with histiocytoid endothelial cells [4]. Its main differential diagnosis is Kimura's disease,

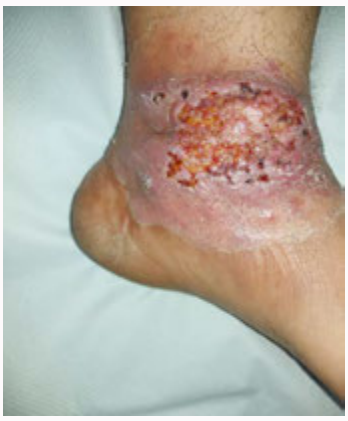

Figure 1: Clinical features of the case, a 27-year-old Tunisian male that presented with an itchy and nodular ulcerated lesion on the right ankle.

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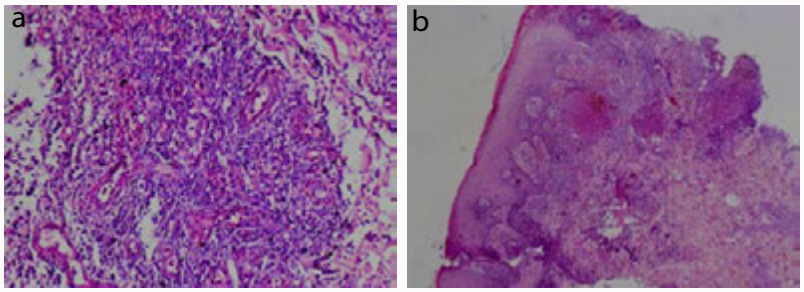

Figure 2: Histopathological features: 27-year-old Tunisian man that presented with nodular ulcerated lesion on the right ankle (a) many irregular hyperplastic vessels with hypertrophic endothelium in the dermis. Endothelial cells were protruding into the lumen assuming a spiked appearance (hematoxylin and eosin, original magnification x10); (b) diffuse inflammatory infiltrate consisting primarily of eosinophils and lymphocytes with nodular architecture (hematoxylin and eosin, original magnification x10).

but other diseases such as cutaneous leishmaniasis, angiosarcoma, or skin lymphocytoma may clinically look like ALHE. Kimura's disease occurs mainly in Asian young men with multiple asymptomatic masses involving the subcutaneous tissue and salivary glands [2]. In contrast to ALHE, Kimura's disease is typically accompanied by regional lymphadenopathy, peripheral blood eosinophilia, increased serum IgE and proteinuria. Histopathology of Kimura's disease demonstrates hyperplastic lymphoid follicles containing prominent germinal centers associated with a few vessels with non-epithelioid endothelium. The diagnosis of ALHE disease in our case was based on histological findings, the absence of lymph nodes and the absence of laboratory abnormalities.

The treatment of choice for ALHE is surgical excision. Cryotherapy, local radiotherapy, topical or intralesional corticosteroids, are also alternative therapies [5]. Spontaneous regression is possible too. In our case, as the surgical excision can't be indicated due to the size and the location of the lesion, we opted for cryotherapy.

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