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CASE REPORT

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Misdiagnosis of uncommon presentation of Angiolymphoid hyperplasia with Eosinophilia

Lai-lai Zhou | **Ru-zhi Zhang**

Department of Dermatology, The Third Affiliated Hospital of Suzhou University, Changzhou, China

Correspondence
Ru-zhi Zhang, Department of Dermatology, The Third Affiliated Hospital of Suzhou University, 185 Juqian Road, Changzhou 213003, China.
Email: zhangruzhi628@163.com

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Abstract

In 50-year-old female patients, the purple-red papules in the right upper arm were misdiagnosed as molluscum contagiosum or scars. The diagnosis of angiolymphoid hyperplasia with eosinophilia was determined on the basis of the clinicopathological features.

KEYWORDS

angiolymphoid hyperplasia, arm, eosinophilia

1 | INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a vasoproliferative disorder involving the skin and subcutaneous tissue. It is mainly manifested as solitary or multiple, reddish brown intradermal or subcutaneous papules and/or nodules. Approximately 85% of the lesions occur in the head and neck area, particularly the region around the ears, forehead, or scalp.[1] ALHE may also appear on the hand, shoulder, breast, tongue, eyelid,[2] conjunctiva,[3] penis, and scrotum.[4] Here, we report a case where the lesions appeared on the upper arm and were misdiagnosed as molluscum contagiosum or scar.

2 | CASE PRESENTATION

A 50-year-old female complained of some painless papules and nodules on her right arm for 1 year. The lesions had been misdiagnosed as molluscum contagiosum and had been treated with a carbon dioxide laser at a local hospital in June 2017. The lesions did not fade, and their number has increased slightly. Six months later, the patient saw a doctor in our department, but refused the doctor's advice for pathological and hematological tests. A dermatologist considered these lesions as scars only by visual inspection and prescribed a topical corticosteroid ointment. However, the number of rashes slowly increased and the individual grew up gradually, with occasionally mildly local pain. In February of this year, the patient finally agreed to perform the relevant tests. She had no history of preceding trauma or inflammatory dermatosis. Physical examination revealed several dark reddish papules and nodules measuring 0.2-0.7 cm on her right upper arm (Figure 1), without evidence of axillary lymphadenopathy. Laboratory investigations including blood and urine analysis, erythrocyte sedimentation rate, coagulation function, and a syphilis serum test did not reveal any abnormality. A skin biopsy taken from a nodule showed a slightly hyperplastic epidermis and the proliferation of lymphatic vessels and small blood vessels in the dermis. Those vessels were lined by enlarged endothelial cells, and some of which protruded into the lumen (Figure 2A,B). There was a mixed inflammatory infiltration mainly of lymphocytes, with a few eosinophils and histiocytes around the vessels and appendages (Figure 2C). Hyperplastic vascular endothelial cells can be seen under high magnification, such as histiocytes or epithelioid endothelial cells, with a large nucleus, eosinophilic cytoplasm, columnar or cubic shape, protruding

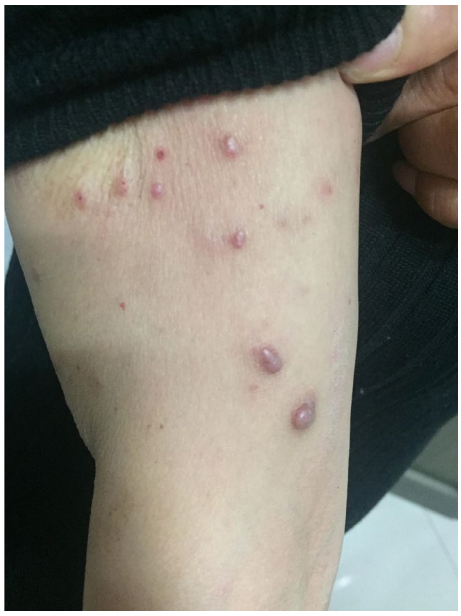

FIGURE 1 Several dark reddish papules and nodules measuring 0.2-0.7 cm on the right upper arm

to the vascular lumen, and sometimes vacuoles can be seen in the cytoplasm (Figure 2D). Hence, a final diagnosis of ALHE was made. The dermatologist explained to the patient about the occurrence and prognosis of the disease and suggested that long-acting corticosteroids be injected into the lesions. However, the patient insisted on waiting for 3 months before deciding whether to undergo treatment or not. Three months later, the patient told the doctor on the phone that the skin lesions had tended to decrease and subside and declined further treatment.

3 | DISCUSSION

Angiolymphoid hyperplasia with eosinophilia is a benign vasoproliferative disorder involving the skin and subcutaneous tissue, and was first described by Wells and Whimster in 1969.[5] Most cases are idiopathic, and several factors implicated in the pathogenesis include insect bites, infections, trauma, immunologic factors, and hyperestrogenic states.[6] The disease usually presents as solitary or multiple dome-shaped papules or nodules ranging in the sizes from 2-3cm and varying in color from brown to pink. Over half of ALHE patients present with a single lesion. Generally, the lesions are localized and unilateral in distribution. The most common locations are the ear, periauricular area, face and scalp, and lesions are occasionally reported on the hand, shoulder, breast, eyelid, as well as tongue conjunctiva, nostrils, penis, and scrotum.[2-4] The pathogenesis of ALHE is still controversial. Currently, the widely accepted hypothesis is that ALHE is a reactive vascular hyperplasia to various stimuli.[7] Clinically, insect bite reactions, pyogenic granuloma, cylindroma, hemangioma, or other diseases may resemble ALHE. The main differential diagnosis of ALHE is Kimura's disease in the Eastern literature, which was described by Kimura in 1948. Kimura's disease mainly presents with one or multiple asymptomatic masses involving the subcutaneous tissue and salivary glands in young Asian males, often accompanied by some systemic manifestations. The histopathological characteristics of Kimura's disease display lymphoid follicles and a higher eosinophil infiltration.[8,9] In contrast, ALHE appears predominantly in middle-aged women as multiple small papules or erythematous nodules. Histopathologically, ALHE is characterized by vascular proliferation and diffuse

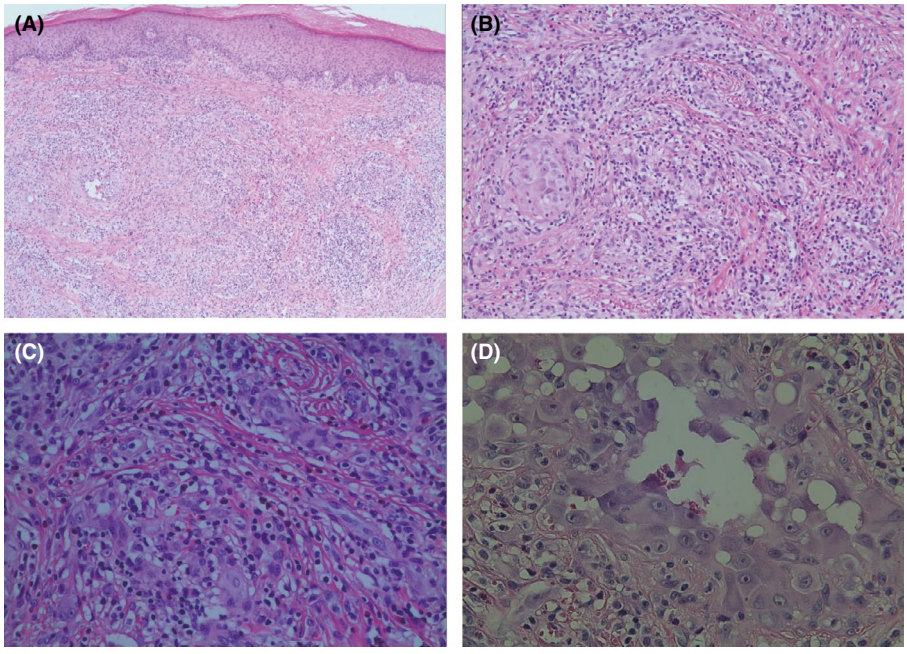

FIGURE 2 A, Diffuse lymphohistiocytic infiltrate with numerous eosinophils in a fibrous stroma (HE × 40). B, Vascular proliferation with prominent (plump) endothelial cells and occasional cytoplasmic vacuoles (HE × 100). C, Vascular proliferation with an intense eosinophilic infiltrate (HE × 200). D, Vascular proliferation with prominent (plump) endothelial cells and occasional cytoplasmic vacuoles (HE × 400)

lymphocytic infiltrates with eosinophils. We describe here a distinctive case of ALHE with regard to its location and multiple lesions. This patient was misdiagnosed as molluscum contagiosum or a scar. Since this disease is rare, attention should be paid to raising awareness.

Treatment of ALHE is often to relieve symptoms and address cosmetic concerns. Surgical excision is commonly used, but relapse is also common. Other therapeutic options include ablative laser therapy, cryotherapy, intralesional radiofrequency, topical or intralesional corticosteroids, isotretinoin, topical timolol, topical imiquimod, tacrolimus, interferon α-2a and anti-interleukin 5 antibody, photodynamic therapy, oral propranolol, or methotrexate.[10] Spontaneous resolution has also been reported, and the spontaneous regression rate was 2.9% (17 out of 593 cases) in a systematic review of the literature conducted by Adler et al[11] Our patient had not received any treatment, but was careful to avoid any irritation. After three months, the lesions had tended to subside.

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CONFLICT OF INTEREST

None declared.

AUTHORS' CONTRIBUTIONS

Lailai Zhou: was responsible for manuscript writing (First Author); Ruzhi Zhang: was responsible for whole project design (Corresponding Author); All authors: read and approved the final manuscript.

ORCID

Lai-lai Zhou https://orcid.org/0000-0003-0891-7895

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