www.jsstd.org

Journal of Skin and Sexually Transmitted Diseases

Case Report

Angiolymphoid hyperplasia with eosinophilia

S. Pradeep Nair

Department of Dermatology and Venereology, Government Medical College Trivandrum, Kerala, India.

***Corresponding author:**
Dr. S. Pradeep Nair,
Department of Dermatology and Venereology, Government Medical College,
Trivandrum –Kerala, India.

dvmchtvm@yahoo.co.in

Received : 19 December 18
Accepted : 08 January 19
Published : 22 April 19

DOI
10.25259/JSSTD_1_2019

Quick Response Code:

ABSTRACT

A 45-year-old male presented with multiple discrete and confluent erythematous plaques, distributed on the malar area and bridge of nose of 1-year duration those were blanching on diascopy. There was no history of photosensitivity. There were no skin lesions elsewhere. Blood examination was normal and antinuclear antibody profile was negative. Skin biopsy showed the upper dermis, mid dermis, and subcutaneous tissue packed with vascular spaces lined by epithelioid endothelial cells with eosinophilic infiltrate, diagnostic of angiolymphoid hyperplasia with eosinophilia. The patient was treated with Nd-Yag laser resection.

Keywords: Angiolymphoid hyperplasia with eosinophilia, Kimura's disease, Epithelioid hemangioma

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a benign vascular proliferative disorder of unknown etiology. Epithelioid hemangioma and pseudopyogenic granuloma are the other names for this entity. Some workers consider it to be a reactive process due to an unknown stimulus. Trauma and otitis externa may be precipitating factors.[1] The present concept is that there is no relation between ALHE and HHV-8, unlike Kaposi's sarcoma. It presents with erythematous papules and plaques on the head and neck region with lesions becoming confluent in the periauricular region. Usually, there are no systemic associations. Even though the disease name incorporates eosinophilia, peripheral eosinophilia is seen only in 10% of the cases.[2] However, tissue eosinophilia is seen. Its presentation on the face may mimic discoid lupus erythematosus (DLE) or Jessner lymphocytic infiltrate. We are reporting here a rare case of ALHE without ear lesions.

CASE REPORT

A 45-year-old male presented with multiple discrete and confluent reddish lesions distributed on the malar area and bridge of the nose of 1-year duration. The lesions first started on the bridge of the nose and later involved the malar region. There was no exacerbation of these lesions with sunlight or spicy foods. The lesions were slightly pruritic. There was no history of any drug intake before the onset of the skin lesions. On examination, the patient had multiple discrete and confluent erythematous plaques distributed on the bridge of the nose and malar region which blanched on diascopy [Figure 1]. The ears were not involved. There were no oral lesions or skin lesions elsewhere. Dermatoscopy examination showed prominent blood vessel architecture. Systemic examination was unremarkable.

Blood hemogram, liver function tests, and renal function tests were within normal limits. Serological tests for syphilis, hepatitis B and C, and HIV were negative. Antinuclear antibody (ANA) profile was negative. Chest X-ray and ultrasound abdomen were within normal limits. Skin biopsy showed the upper and mid dermis, packed with vascular spaces and some extension into the subcutaneous tissue

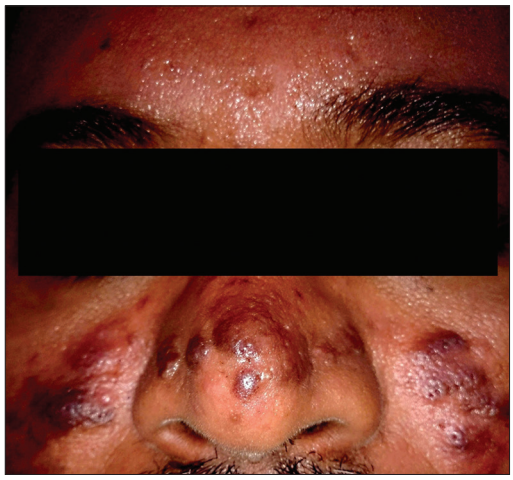

Figure 1: Erythematous plaques on nose and malar area of angiolymphoid hyperplasia.

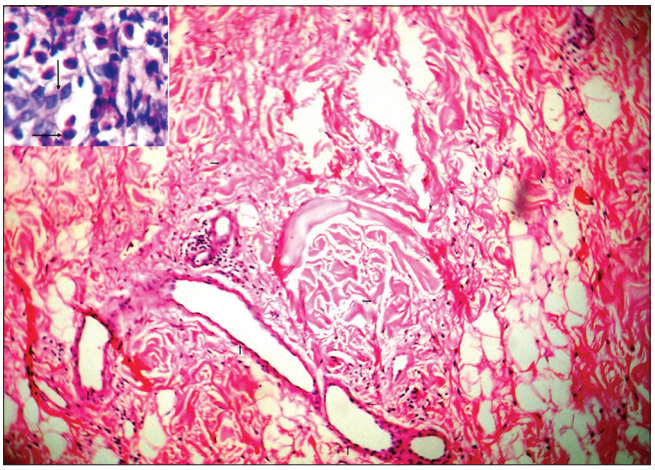

Figure 2: Skin biopsy showing the upper dermis, mid-dermis, and subcutaneous tissue filled with vascular spaces and eosinophilic infiltrate, H and E ×100; inset: Showing high power of skin biopsy with epithelioid endothelial cells (down arrow) and eosinophils (straight arrow), H and E ×400.

[Figure 2]. There was infiltration by eosinophils. High-power view showed vascular spaces lined by epithelioid endothelial cells with eosinophilic infiltrate, diagnostic of angiolymphoid hyperplasia with eosinophilia [Figure 2, inset]. The patient was treated with Nd-Yag laser resection.

DISCUSSION

Our patient presented with erythematous plaques on the nose and malar region. Dermatoscopy showed a predominant vascular architecture. ANA profile was negative. Histopathology showed proliferating vascular spaces lined by epithelioid endothelial cells with eosinophilic infiltrate. Therefore, we made a final diagnosis of ALHE. Even though the patient did not have peripheral eosinophilia, there was tissue eosinophilia which is the usual feature of ALHE.

Table 1: Salient clinical and histological differences between ALHE and Kimura's disease.

ALHE	Kimura's disease
Head and neck region	Head, neck, and other areas
Lymph node involvement rare	Lymph node involvement common
No parotid gland involvement	Parotid gland involvement
Dermis and subcutaneous tissue	Predominantly subcutaneous tissue
Angiomatous component more	Lymphoid component more
IgE levels normal	IgE levels are increased
Not seen	Eosinophilic abscesses are seen
Not seen	Fibrosis around blood vessels seen

ALHE: Angiolymphoid hyperplasia with eosinophilia, IgE: Immunoglobulin E

An interesting feature of our patient is the lack of lesions on the ear lobes which is considered to be the classical site. Hence, we had to investigate for other causes which were negative. However, there have been reports of ALHE presenting without ear lesions.[2] There has been some confusion regarding ALHE and Kimura's disease. The present concept is that both are different entities. The salient clinical and histological differences between ALHE and Kimura's disease are given in Table 1. The histopathology of ALHE is very specific. The dermis and sometimes the subcutaneous tissues are filled with vascular spaces lined with plump endothelial cells resembling epithelioid cells which protrude into the lumen of the blood vessels giving a "cobble stone" appearance, and hence, it is also known as epithelioid hemangioma.[3] In between the vascular spaces, there will be infiltrate by eosinophils. Since the presentation is on the face and neck region, the important differentials are DLE, sarcoidosis, Jessner lymphocytic infiltrate, and granuloma faciale. Histopathology can clinch the diagnosis. The skin lesions may also be confused with bacillary angiomatosis and pyogenic granuloma. Pyogenic granuloma has a characteristic clinical presentation with angiomatous papules and plaques with a peripheral collarette of epidermis which bleeds on touch and has a characteristic histopathology. Bacillary angiomatosis presents with painful angiomatous lesions on the upper extremities and trunk with the involvement of organs including the liver and bone marrow. Histopathology with special stains can confirm the diagnosis. The oral mucosa, tongue, lymph nodes, bones, and testes are the other rare sites reported in literature to be involved in ALHE. There are no definite treatment modalities for ALHE. Nd-Yag laser, cryotherapy, radiofrequency ablation, isotretinoin, Interferon-α 2a, oral propranolol, topical tacrolimus, intralesional steroids, timolol, and imiquimod are reported to be effective.[4] Recently, anti-interleukin 15 antibody, mepolizumab was found to be effective.[5] Our case was successfully treated with Nd-Yag laser.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names

and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Lade NR, Banode P, Dhope N, Tharafdar P, Saoji V. Angiolymphoid hyperplasia with arterial ectasia and arteriovenous malformation: Response to intra-arterial embolization. Indian J Dermatol Venereol Leprol 2016;82:413-5.
2. Gupta LK, Bansal NK. Multiple grouped nodules around the right ear in a young male. Indian J Dermatol Venereol Leprol 2011;77:399-401.
3. Shastry V, Vijaya. Multiple erythematous papules on the pinna. Indian J Dermatol Venereol Leprol 2010;76:83-4.
4. Zaraa I, Mlika M, Chouk S, Chelly I, Mokni M, Zitouna M, *et al*. Angiolymphoid hyperplasia with eosinophilia: A study of 7 cases. Dermatol Online J 2011;17:11-15.
5. Damarla SV, Chintagunta SR, AraSkkal GK. Recurrent unilateral angiolymphoid hyperplasia with eosinophilia in pregnancy. Indian J Dermatol Venereol Leprol 2018;84:325-7.

How to cite this article: Nair SP. Angiolymphoid hyperplasia with eosinophilia. J Skin Sex Transm Dis 2019;1:32-4.