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Case Report

Angiolymphoid hyperplasia with eosinophilia of the inguinal region–unusual location of rare entity

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Abstract

Angiolymphoid hyperplasia with eosinophilia (ALHE), also called epithelioid hemangioma (EH), is an unusual, benign proliferation of vessels characterized by inflammation with high levels of eosinophils. In the current literature, there is very less evidence of ALHE occurring on the lower extremities. We report a rare instance of Angiolymphoid Hyperplasia with Eosinophilia (ALHE) in a 49-year-old male who presented with a pruritic, non-tender, palpable lump on the right inguinal area. Fine needle aspiration cytology was done and was suggestive of abscess. However, histopathological examination revealed increased vascular proliferation with dense infiltrate of eosinophils, and confirmed it to be a case of Angiolymphoid hyperplasia with eosinophilia. (ALHE).

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

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1. Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a unique, benign vascular entity characterized by the growth of small, angiomatous-like bumps or nodules, typically in the head and neck area.[1,2] It is very rarely seen in Inguinal region with only single case being previously reported.[3,4] ALHE is also referred to as epithelioid hemangioma because of abnormal proliferation of endothelial-like cells.[4] The etiology of ALHE is not clear; though, it has been reported in pregnancy and post trauma in some patients.[4,5]

ALHE and Kimura's disease (KD) have many similar clinical and histopathological features. They were initially considered as sequential stages of a same disease but now they have been recognised as separate entities.[5] Histopathological features of ALHE and KD are same, with both lesions having a dermal involvement, prominent proliferation of endothelium and eosinophilic infiltrate. However, they can be distinguished by features of endothelial cell morphology. Kimura's disease is related with elevated levels of peripheral blood eosinophils and increased serum immunoglobulin E (IgE) levels.[5,6] Notably, approximately 20% of patients with ALHE have eosinophilia without elevated IgE levels, making it an important criteria to make the diagnosis of ALHE.[6,7]

The differential diagnosis for ALHE encompasses a range of conditions like epithelioid hemangioendothelioma, injection site granuloma, eosinophilic cellulitis, eosinophilic leukemia and eosinophilic pustular folliculitis.[7]

We hereby report a rare case of ALHE in a 49 year old male presenting with a small palpable lump in the inguinal region.

2. Case Report

A 49 year-old male came to the Surgery OPD with a two weeks history of painless swelling in right inguinal region. Patient had history of travelling to a village four months back and bathing in a pond, after which he noticed that skin started exfoliating on both his feet. Patient had complaints of pruritus not associated with pain or bleeding. The patient did not receive any treatment for the skin lesions. Three months later

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he noticed a swelling at inguinal area for which he consulted our hospital. On physical examination, a lump of size 1.5 x 1.5 cm was identified at right inguinal region. The lump was soft to firm, fixed, and non-tender. Patient had no history of fever. The patient was initially treated with antibiotics for one week, however the swelling did not subside and later excision was planned.

Ultrasound revealed significant raised echogenicity, skin thickening, subcutaneous edema in right inguinal region with small hypoechoic area measuring 15mmx 14 mm (? abscess) - suggestive of inflammatory/infective etiology. Other findings were mild right hydrocele, left undescended testis, mild ascites, grade II fatty liver and left renal type II BOSNIAK cyst.

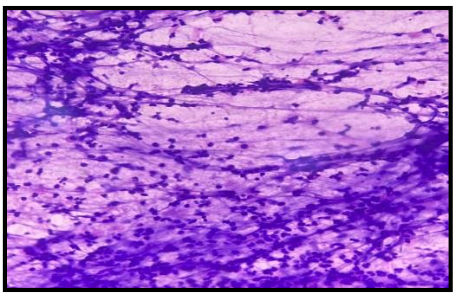

Figure 1: Photomicrograph from inguinal lesion showing neutrophils with few eosinophils in a necrotic background. (Giemsa stain, 40x)

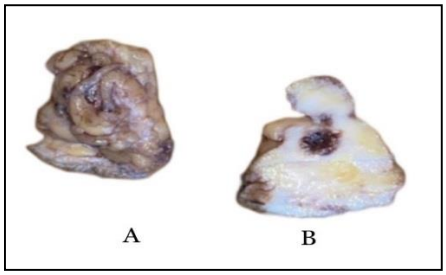

Figure 2: Gross images of excised inguinal swelling; (**A**): External surface showing fibrofatty tissue piece with skin ellipse; (**B**): Cut surface showing grey yellow area with foci of hemorrhage.

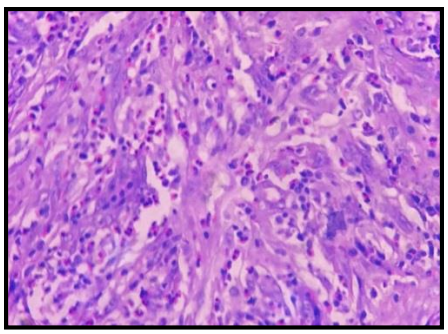

Figure 3: Photomicrograph from inguinal swelling showing dense infiltrate of eosinophils. (H&E, 40x)

Pus culture however showed no growth after 48 hours of incubation at 37°C. No eosinophilia was seen on the peripheral blood. Fine needle aspiration cytology of the swelling was performed and yielded pus like thick material. FNA smears examined were cellular and showed inflammatory cells both viable and degenerated predominantly comprising of polymorphs, few eosinophils, lymphocytes and occasional histiocytes in a necrotic background. Cytological examination revealed no evidence of granulomas or atypical cells, and the Ziehl-Neelsen (ZN) stain for acid-fast bacilli was negative. Cytological features indicated Abscess. **[Figure 1]** Swelling was excised and sent for histopathological examination. Grossly, fibrofatty tissue piece with attached skin ellipse measuring 4×3×2cm was received. **[Figure 2A]** On cut section, a grey yellow area was noted along with foci of haemorrhage. **[Figure 2B]** Microscopic examination revealed tissue lined by stratified squamous keratinized epithelium with acanthosis at places. Subepithelial tissue showed a lesion composed of lymphoid follicles with prominent pale germinal centres along with dense infiltrate of eosinophils **[Figure 3]** few neutrophils and plasma cells. Proliferation of vascular channels of variable calibre was seen having plump endothelial cells with epithelioid features. **[Figure 4]** Intervening thick collagen bundles with haemorrhage infiltrating into the deeper adipose tissue was also present. There were no features of granuloma or malignancy.

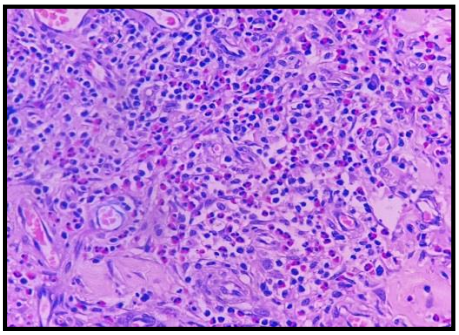

Figure 4: Photomicrograph showing proliferation of prominent endothelial lining. (H&E, 40x)

A histomorphological diagnosis of Angiolymphoid hyperplasia with eosinophilia was given.

3. Discussion

Angiolymphoid hyperplasia with eosinophilia (ALHE), also called epithelioid haemangioma was narrated by Wells and Whimster in 1969 and was later regarded as a late stage manifestation of Kimura's disease, a condition previously documented in Japanese medical literature.[7,8] Nowadays, it is believed that Kimura's and ALHE are separate disease entities with a different clinico-pathological findings.[9]

The symptoms of ALHE include pain, itching, pulsating sensations and unprovoked bleeding. [9,10] It is predominantly

seen in mid aged women, with a typical age of onset of 30-33 years.[10]

Origin of ALHE is unknown and is believed to be unusual reactive process. Underlying arteriovenous shunting could have a role in its causation, which is supported by histopathological evidence of damaged blood vessels. The development of Angiolymphoid Hyperplasia with Eosinophilia (ALHE) has been linked to various factors, including trauma, increased blood levels of estrogen in pregnancy, use of oral contraceptive pills and infection.[10,11]

Angiolymphoid Hyperplasia with Eosinophilia (ALHE) typically presents as red to purple-colored papules, plaques, or nodules with 0.1-2 cm in size located in the dermal tissues of the head and neck region most commonly found in the ear and surrounding areas.

Less frequently it affects the abdomen, limbs, intestine, and oral mucosa.[11]

Inguinal region is rarely involved. Current literature describes very few reported cases of ALHE involving the lower limbs. Sezer et al[4] presented a case of 58-year old male having four year history of multiple pink plaques over the inguinal folds and scrotum. The lesions resembled bowenoid papulosis clinically, however, histopathology confirmed ALHE. Dewan et al[7] reported a case of ALHE in a 47 year old male with 8 month history of red nodules on glans penis and scrotum. Park et al[8] in 2009 presented a case on 17 year old male with 15 day history of papules and nodules on penis and scrotum. Histopathological examination of excisional biopsy showed dermal tissue infiltrate of lymhocytes, eosinophils and marked vascular proliferation, diagnosis was confirmed as ALHE.

In our case, ALHE involved the inguinal region, an unusual clinical presentation. Cytological findings were of abscess with mixed infiltrate of polymorphs, lymphocytes and eosinophils. Histopathological findings, however, confirmed it to be ALHE.

ALHE and Kimura's disease can be differentiated on microscopy. ALHE is characterized by the occasional lymphoid follicles whereas Kimura's disease is known for well-formed lymphoid follicles in addition to dense eosinophilic infiltrates. [11] In our case, there were occasional lymphoid follicles, proliferation of vessels and dense eosinophilic infiltrate.

Therefore a key differential diagnosis to consider is Kimura's disease which is notably more prevalent in Asian males. In Kimura's disease, the patient presents with peripheral eosinophilia, lymphadenopathy, immunologically mediated systemic involvement like membranous nephropathy and increased serum IgE levels. Histological examination shows involvement of deeper part with numerous lymphoid follicle formation, eosinophil microabscess and dense fibrosis. Whereas ALHE shows many abnormal vascular proliferation with dense aggregates of eosinophils and lymphocytes. These capillaries have plump endothelial cells with hobnailing.[11,12] In our case there was no increase in IgE levels.

The differential diagnosis that can be considered are bacillary angiomatosis, cylindroma, angiosarcoma, injection site granuloma, epithelioid hemangioendothelioma, eosinophilic cellulitis, eosinophilic leukemia and eosinophilic pustular folliculitis.

Surgical excision is the preferred treatment option.[12]

4. Conclusion

ALHE is an uncommon disease entity characterized by endothelial cell proliferation surrounded by an inflammatory cell infiltrate comprised of mainly eosinophils. The head and neck region is the most frequent site for the development of Angiolymphoid Hyperplasia with Eosinophilia (ALHE) lesions. Rarely ALHE is seen at inguinal region or lower limbs. Recurrence is possible, and the likelihood of recurrence does not diminish over time without treatment. Therefore, an accurate diagnosis is crucial to guide appropriate management and intervention. Through this exceptional case, we underscore the imperative for clinicians to broaden their diagnostic purview and consider Angiolymphoid Hyperplasia (ALHE) as a potential entity in the inguinal region, thereby augmenting their index of suspicion and facilitating timely diagnosis.

5. Source of Funding

None.

6. Conflict of Interest

None.

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