# Case Report:

# Angiolymphoid hyperplasia with eosinophilia of the penis

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#### **ABSTRACT:**

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare, benign entity of unknown pathogenesis. It often presents as painful or pruritic intradermal or subcutaneous red to brown papules or nodules on the head and neck of young adults. Rare sites of involvement are hands, PENIS, conjunctiva, breast, oral mucosa, orbit. We report a case of 36 year old HIVpositive male who presented with history of swelling over ventral aspect of prepucial skin of 5 months duration. Clinically differential diagnosis of primary chancre/Carcinoma penis/Donovanosis was considered. Pathological findings showed nodular lesion with dermal proliferation of anomalous blood vessels lined by plump, epithelioid endothelium with a significant perivascular inflammatory infiltrate composed of lymphocytes, histiocytes and many eosinophils, consistent with the diagnosis of ALHE. Although recurrence is common, surgical excision is still regarded as a treatment of choice. We discuss about this exceptional entity for its rarity of site and should be considered in differential diagnosis of penile lesions.

Keywords: Angiolymphoid hyperplasia with eosinophilia

#### INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon, benign, idiopathic, vasculo-proliferative lesion, first reported by Wells & Whimster in 1969 based on study of 9 cases. [1,2] It is also known as Epithelioid Hemangioma [3]. It is a distinctive vascular lesion which usually occurs during early & mid adult life (20-50yrs) & clinically presents as single or multiple pink to red brown papules & nodules on face, scalp & ear.

Rare sites of involvement are hands, breast, PENIS<sup>[1,3,4,6]</sup>, shoulders, oral mucosa & orbit.

# CASE REPORT

36 yr old HIV positive male presented with slowly growing ,non tender nodule of 5 months duration over ventral aspect of prepucial skin. On physical examination ,3x2 cm firm, brown subcutaneous mass was found on the prepucial skin. No other symptoms or signs were noted. Lab tests were within normal limits. Clinically differential diagnosis of primary chancre/Carcinoma

penis/Donovanosis were considered. Incisional biopsy followed by circumcision was done.

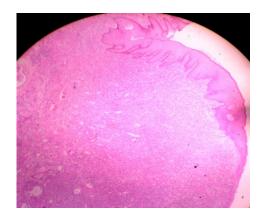
Grossly, specimen was well circumscribed, nodular mass of 3cm in diameter present over prepucial skin flap of 4.4 cm in length. Cut surface of mass was grayish white.(fig.1,2)

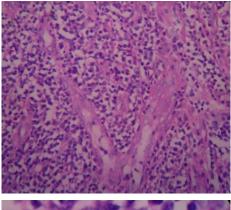
Histopathological examination revealed a nodular lesion lined by stratified squamous epithelium with focal areas of ulceration(fig.3). Underneath is seen nodular appearing proliferation composed of haphazardly arranged, small calibre sized, irregularly shaped blood vessels accompanied by patchy inflammatory infiltrate.

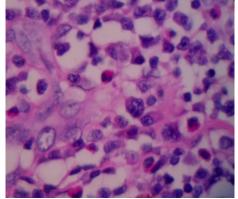
The vascular spaces are lined by plump appearing pleomorphic, epithelioid endothelial cells with hyperchromatic nuclei, inconspicuous nucleoli & copious eosinophilic cytoplasm. Inflammatory infiltrate is Characterised by admixture of mature lymphocytes, numerous eosinophils & scattered histiocytes (fig. 4,5).











#### DISCUSSION

ALHE is a rare, benign entity of unknown pathogenesis occurring most frequently over head, neck, periauricular region or scalp. Very few cases have been reported in the penis. Previously it had been described as pseudo- or atypical pyogenic granuloma, subcutaneous angioblastic lymphoid hyperplasia with eosinophilia, and papular angioplasia <sup>[3,7]</sup>. Initially thought to be related to Kimura's disease, a condition occurring in male Asians sharing some of the same clinical and histological features, ALHE is now considered a distinct entity <sup>[3,8,9]</sup>.

Association of ALHE with HIV, trauma has been reported in the literature. ALHE can persist for yrs but serious complications (e.g. malignant transformation) do not occur. ALHE should be differentiated from Kimura Disease, epithelioid hemangioendothelioma & epithelioid Angiosarcoma by its superficial location ,lobular architecture,

predominant vascular component with epithelioid endothelial cells, characteristic inflammatory infiltrate ,lack of frankly infiltrative growth,lack of extensive mitotic activity & absence of significant nuclear atypia. Local surgical excision is the treatment of choice with low rate of recurrence.

We present this case to expand anatomical distribution of this rare lesion.

#### CONCLUSION

Penis is a rare site for occurrence of ALHE & ALHE should be considered in the differential diagnosis of penile lesions in HIV patients.

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Date of submission: 18 December 2013

Date of Provisional acceptance: 29 December 2013

Date of Final acceptance: 19 February 2014

Date of Publication: 04 March 2014

Source of support: Nil; Conflict of Interest: Nil