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Angiolymphoid Hyperplasia with Eosinophilia: A Case Report

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Abstract: ***Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon benign vascular neoplasm with unclear pathogenesis that typically occurs in adults. Usually presents as a group of several red-brown or violaceous skin papulo-nodules in a localized area. Usually occurs on the head or neck, and the ears are a particularly common location. In our case we describe an elderly male with more than 50 in number of papules and nodules involving a wide area of the scalp and neck range from 0.3 mm to around one cm in diameter. Histological examination of skin lesion showed dermal vascular proliferation, lined with plump endothelial cells and surrounded by inflammatory cells. Clinical and histological examination was consistent with ALHE.The management was done by removing most of lesions by curettage and cauterization of the base. Recurrence is expected with any treatment modality but in this case in addition to recurrence, new lesions appeared.***

Keywords: Angiolymphoid hyperplasia, eosinophililia

1. Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE), also known as epithelioid hemangioma (EH). It is an uncommon benign vascular neoplasm. The etiology is currently unknown. Various hypotheses have been proposed, including a reactive process, a neoplastic process, and infectious mechanisms. Most commonly presents in young to middle-aged adults but it can occur at any age. [1] There is no sex or ethnic predominance among patients with ALHE. [2]

Usually present as a single or a group of several red-brown or violaceous papules or small nodules in a localized area on the skin. ALHE usually involves the head or neck; with the ears are common site. Other less common locations are: trunk, extremities, hands, penis, oral mucosa, and colon. [1] Although EH is usually superficial, deeper soft tissues and large blood vessels can be involved. [3] Lesions are usually asymptomatic but pain, pruritus, pulsation and bleeding may occur. [1, 2]

2. Case Report

We report a case of a 71-year old man present with multiple papules and nodules involving his head and neck asymptomatic apart from bleeding with manipulation developed over one year. No history of trauma to the area. The patient was diagnosed with hypertension and diabetes 2 years ago.
Clinical exam revealed multiple papules and nodules more than 50 in number, purple to dull red in color, distributed widely on the left part or the scalp and neck posteriorly with few lesions anteriorly, range from 0.3mm to around one cm in diameter, some of them topped with crustation. They are firm with smooth surface, non-pulsatile, no associated lymphadenopathy (figure 1, 2).

Also we noticed small papules and purpura like lesions involving the base of the tongue which is also asymptomatic. (Figure 3)

His laboratory result were unremarkable, Histological examination of skin lesion showed dermal vascular proliferation, lined with plump endothelial cells and surrounded by inflammatory cells (Figure4) and (Figure 3). No atypical mitotic figures were observed. We could not take biopsy of his oral lesions because of the patient refusal. So we are not sure whether it is consistent with the same diagnosis.

The management plan was to refer the patient for laser therapy but because the patient could not afford it, so we removed most of lesions by curettage and cauterization of the base, no complications was faced during or after the treatment. Three months later the patient developed new lesions in addition to recurrence of some of the removed lesions(figure 2), no farther intervention was taken.

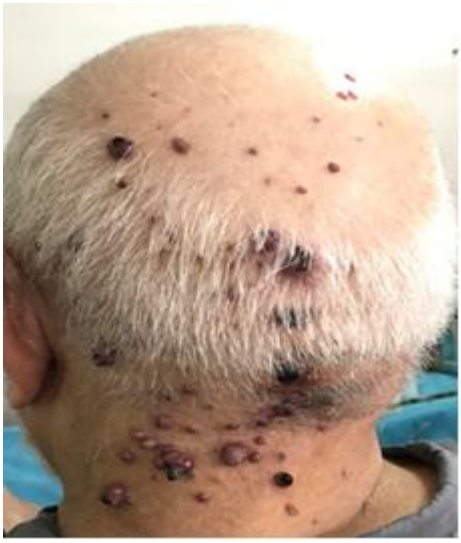

Figure 1: Mutable papules and nodules involving the scalp and neck

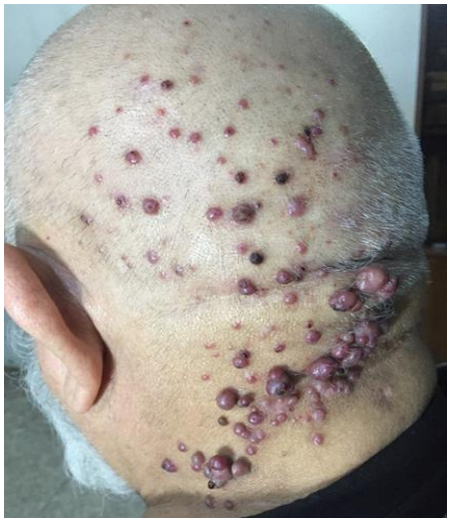

Figure 2: Recurrence of the removed lesions and appearance of new ones.

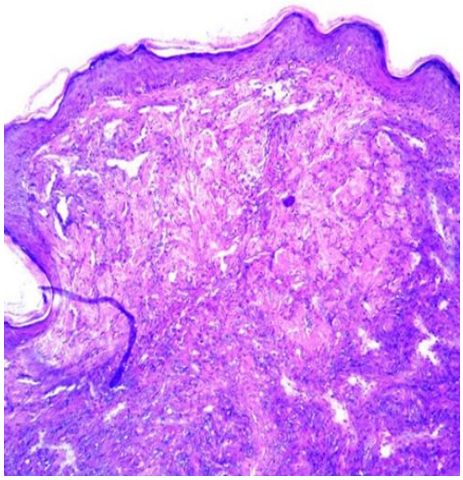

Figure 3: Low power shows vascular proliferation with inflammatory infiltrate.

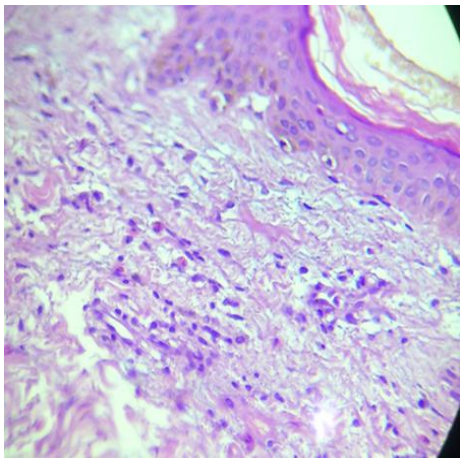

Figure 4: Higher power shows vessels lined by plump endothelial cells and inflammatory cells mainly eosinophils and lymphocytes.

3. Discussion

Clinical and histopathological features need to be present to diagnose ALHE and differentiate it from other benign and malignant conditions. Peripheral eosinophilia is not essential for diagnosis as it present in only 20% of cases. [1, 4, 5]

Clinically present as red-brown or violaceous papules or nodules most involve dermis, subcutaneous, deeper tissues or arise from vessels. head and neck is most common location but other sites can be involved .[1, 2, 5] Approximately half of patients have multiple lesions, generally in a clustered pattern.[2]

Histologically two defining features to diagnose ALHE: 1-proliferation of blood vessels of varying sizes lined by plump endothelial cells.[1]

2-Inflammation: Lymphocytes and varying amounts of eosinophils diffusely surround and may infiltrate the blood vessels.The lymphocytes may be diffusely present or may form distinct follicles with germinal centers. [1]

the vascular or inflammatory component may predominate depending on the age of the lesion: Early or actively growing lesions show predominance of vascular component, the vessels is immature with prominent epithelioid endothelial cells, while in in late stages lymphocytes become more prominent, and the endothelial cells lining the maturing vessels become smaller and less epithelioid. [1, 5]
Clinical differentials that have clinical or histological similarity to ALHE include: - Pyogenic granuloma, Kaposi sarcoma, multiple cylindromas, lymphoma cutis, sarcoidosis, epithelioid angiosarcoma, epithelioid hemangioendothelioma, Richly vascularized metastatic tumors.[1, 2, 4, 5, 6]

Treatment of ALHE is challenging because of the high recurrence rate that is why many therapeutic options have been tried. Surgical excision is the treatment of choice with recurrences is observed in 44.2%.in our patient surgical excision was inappropriate due to multiple lesions and involvement of wide area. Laser therapies (Combination of pulsed dye laser and carbon-dioxide lasers) recurrence can be prevented by maintenance of treatment.Other options include photodynamic therapy, cryotherapy, electrodesiccation, and radiotherapy, brachytherapy, Sclerotherapy with polidocanol or bleomycin.

Systemic treatments include methotrexate, thalidomide, and oral propranolol alone or before surgery to reduce the size or number of lesions. Local treatment options have variable outcome including: Topical and intralesional corticosteroids were the primary therapeutic agents, but high recurrence rates remained. Topicalimiquimod 5%, topical tacrolimus 0.1%, and intralesional interferon alpha-2b. Topical timolol also showed some success in the management of ALHE. [7]

4. Conclusion

The case is being presented due to presence of numerous lesions involving large area of the scalp and neck. Recurrence is expected with any treatment modality but in this case in addition to recurrence, new lesions appeared.

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