KNOW YOUR FIELD

A rare case of angiolymphoid hyperplasia with eosinophilia in the submental region

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ABSTRACT

Angiolymphoid hyperplasia with eosinophilia is a rare reactive, angio-proliferative lesion which is usually found in the subcutaneous tissue of the head and neck. The lesion rarely arises from a native blood vessel. It is of unknown etiology but may be related to some benign, localized form of vasculitis. While preauricular location is the most frequent site of occurrence as reported in literature, a lesion in submental region seems to be a first reported site of occurrence. We report such an occurrence in an individual with history of trauma twice in his lifetime at the affected site. Spontaneous regression is common, but persistent or recurrent lesions may require treatment. Several treatments have been reported but surgery is considered to be the most efficient one.

Key words: Angiolymphoid hyperplasia, eosinophilia, head and neck, lymphoid tissue

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a benign but potentially disfiguring vascular lesion. It is usually characterized by dermal and subcutaneous nodules, primarily in the head and neck region and is especially common in preauricular region. Although rare, this entity should be considered in the differential diagnosis of hypervascular masses of the superficial head and neck region. We present a rare site of occurrence of angiolymphoid hyperplasia with eosinophilia in the submental region.[1-13]

CASE REPORT

A 22-year-old man reported with a complaint of a hard disfiguring swelling in the chin region. The swelling had gradually increased in size to attain the present size and had hardened in consistency to attain the present consistency. Patient had first noticed the swelling 1½ years back and complained of mild associated pruritis. The patient gave a history of trauma to the same region twice, the first about 10 years ago, which was associated with significant bleeding and had required suturing and the second about 2 years ago. His medical history was non-contributory. He had no tissue abuse or tobacco abuse habit.

On extra-oral examination, a single, well-defined, non-tender nodular swelling measuring about 2.5 cm × 1.5 cm in size with firm to hard consistency was present in the submental region [Figure 1]. The nodule was movable in nature. The temperature over the nodule was normal. Intra-orally there was no abnormality detected apart from moderate gingival recession with lower left central incisor.

Periapical, occlusal and panoramic radiographs were obtained and were non-contributory. The laboratory investigations that were advised for the patient included complete blood count, bleeding time, clotting time, random blood sugar, hepatitis B surface antigen test and human immunodeficiency virus test. All the reports were within the normal limits except for differential leukocyte count, which showed marked increase in eosinophils to 8% (normal 1-6%). The lesion was surgically excised and sent for histopathological examination [Figure 2].

HISTOPATHOLOGY

The tissue section showed:

- Vascular proliferation [Figure 3]
- Lymphoid proliferation [Figure 4]
- Vascular proliferation with thickened blood vessel walls showing large plump endothelial cells having oval nuclei with vacuolated cytoplasm [Figure 5]

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- Dense stroma with inflammatory cells viz. abundant eosinophils with central necrosed areas and lymphocytes [Figure 6].

DIFFERENTIAL DIAGNOSES

- Angiolymphoid hyperplasia with eosinophilia
- Kimura's disease
- Cutaneous lymphoma
- Cavernous hemangioma
- Pyogenic granuloma
- Kaposi's sarcoma
- Bacillary angiomatosis.[13]

Cutaneous lymphoma

- Epidermotropism
- The intraepidermal cluster of lymphocytes (Pautrier's abscess)
- Angiodestruction and tissue infarction
- Infiltration into the subcutaneous fat (panniculitis-like cutaneous lymphoma) or blood vessels (intravascular lymphoma).[11]

Kimura's disease

- Produces large subcutaneous nodules in the head and neck region with normal overlying skin
- Often involves regional lymph nodes
- Is a chronic inflammatory condition
- Lacks "epithelioid" blood vessels
- Elevated serum immunoglobulin E.[11,14-20]

Angiolymphoid hyperplasia with eosinophilia

- Proliferation of small blood vessels many of which are lined by enlarged endothelial cells with uniform ovoid nuclei and intra-cytoplasmic vacuoles (cobblestone appearance)
- Perivascular and interstitial infiltrate composed primarily of lymphocytes and eosinophils (5-15% of infiltrate)
- Lymphoid aggregates with or without follicle formation. [1,4-10,12,16-18,21-23]

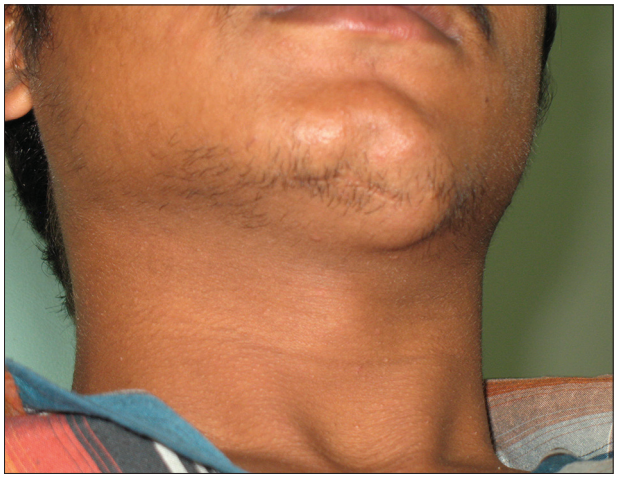

Figure 1: Clinical picture showing extraoral nodular swelling at the submental region

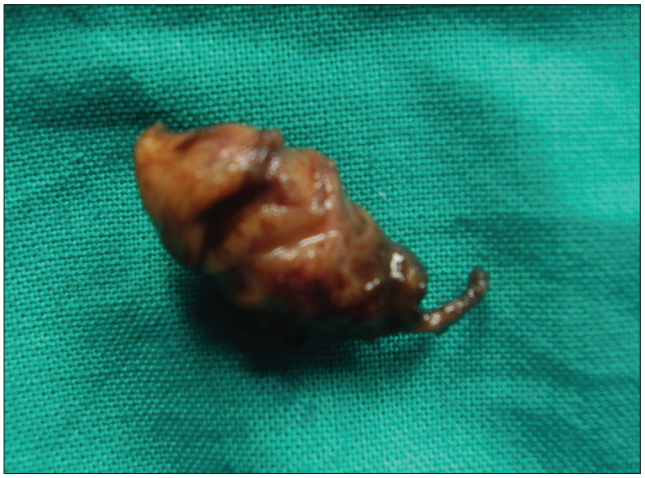

Figure 2: The excised specimen in toto obtained from the submental region

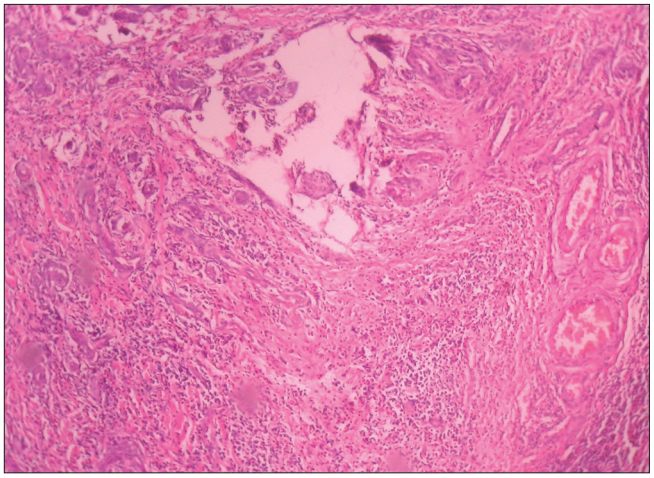

Figure 3: Vascular proliferation (H&E stain, ×100)

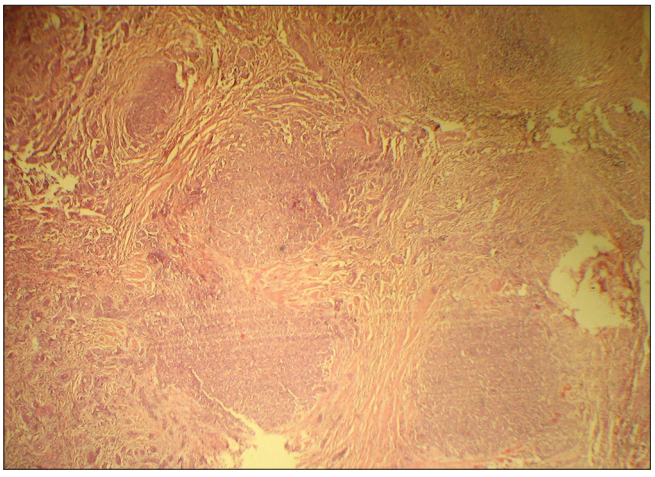

Figure 4: Photomicrograph showing lymphoid proliferation (H&E stain, ×100)

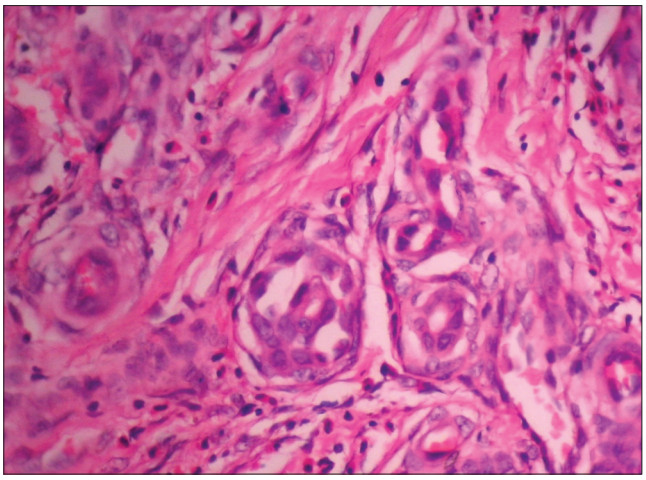

Figure 5: Vascular proliferation with thickened blood vessel walls showing large plump endothelial cells having oval nuclei with vacuolated cytoplasm (H&E stain, ×400)

Cavernous hemangioma

- Large dilated blood sinuses with thin walls, showing endothelial lining
- Sinusoidal spaces usually are filled with blood
- Lymphatic vessels occur in some instances.[2,11]

Pyogenic granuloma

- Lobular pattern of vascular proliferation with inflammation and edema similar to granulation tissue
- Thin and atrophic epidermis at the top with variable ulcerations
- Vast numbers of endothelium-lined vascular spaces
- Acanthosis and hyperkeratosis at sides
- Extreme proliferation of fibroblasts and budding endothelial cells
- Variable mitotic activity.[11]

Kaposi's sarcoma

- Proliferation of small veins and capillaries around one or more preexisting dilated vessels
- Slit-like vascular channels without a visible endothelial lining, typically interspersed with the spindle cells
- Vessels are lined by plump, mildly atypical endothelial cells
- Pronounced mononuclear inflammatory cells infiltrate, including mast cells, scattered erythrocytes and hemosiderin deposits may be present
- Variable mitotic activity.[2,3,11]

Bacillary angiomatosis

- The architecture resembles that of lobular capillary hemangioma, but the endothelial cells are often larger, polygonal and sometimes markedly atypical
- There is presence of neutrophils, leukocytoclastic debris and granular material (bacteria).[11]

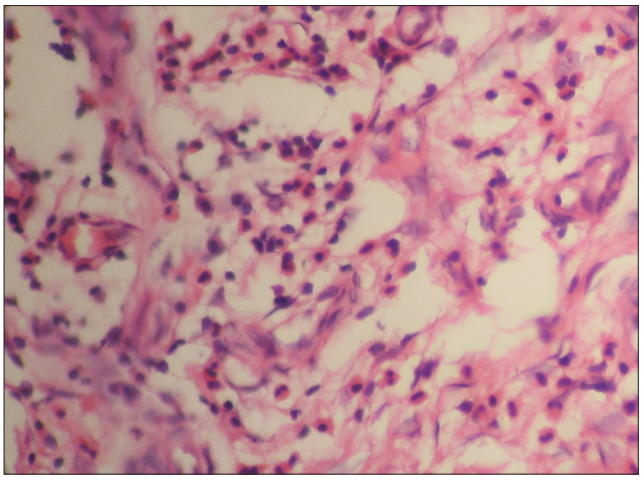

Figure 6: Dense stroma having abundant eosinophils (H&E stain, ×400)

FINAL DIAGNOSIS

Angiolymphoid hyperplasia with eosinophilia.

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