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ANGIO LYMPHOID HYPERPLASIA OF THE UPPER LIP: A RARE CASE REPORT

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Abstract
Angio lymphoid hyperplasia with eosinophilia (ALHE) is a rare benign vascular lesion that commonly presents with pink to brown papules or nodules and predominantly occurs in the skin and subcutaneous tissues of head and neck and very rarely involve oral cavity. This is an angio-proliferating subcutaneous tumor with rich inflammatory infiltrate by lymphocytes and eosinophils. The etiology of ALHE is unclear, with proposed mechanisms extending from reactive vascular hyperplasia to vascular malformation or tumor. It is most commonly found in middle-aged women and is a rare condition in children. We describe a very atypical size and morphology of ALHE localized to the philtral area of the upper lip in an adult male.
KEYWORDS: Angio lymphoid hyperplasia, Eosinophilia, Lip, Excision

INTRODUCTION

The disease was first described by Wells and Whimster in 1969 as Angio lymphoid hyperplasia with eosinophilia (ALHE). (1) Other names are epithelioid hemangioma, inflammatory angiomatous nodule, atypical granuloma, pseudo pyogenic granuloma and histiocytoid hemangioma. It is a non-malignant field of vascular lesion, appearing as painless nodules and papules, solitary or multiple, localized in the dermal and subcutaneous tissues of the head and neck and, particularly affecting the pinna but generally sparing the oral mucosa. It is common in young and middle aged females, but less common in children
(2-5) (3,6–9) It is uncommon in male Asian population. (10) Its histologic features consist of reactive proliferation of blood vessels of varying size with plump endothelial cells with an inflammatory infiltrate containing eosinophils, lymphocytes, and plasma cells [10]. (2,9) We report the unusual presentation as ulcerative nodule in the philtrum of upper lip in a adult Asian male.

Case Report

A 49 years old male presented with swelling over mid upper lip since past 6 months. It appeared to be spontaneous and slowly progressive to its current size. The swelling was painless and had blood-stained discharge. He is a known case of diabetes mellitus and hypertension on regular treatment/medication. On examination, there was a 2 x 1 cm polypoidal pinkish coloured lesion in the philtral area of the upper lip with smooth surface and well-

defined margins arising from the skin. (Fig. 1) A -Afebrile, -Nontender, -No Induration, -No Regional Lymphadenopathy We then did surgical excision with primary closure of the skin. Under general anaesthesia, the lesion was excised, and defect closed in layers primarily with 4-0 vicryl and 6-0 ethilon. (Fig. 2) The lesion was excised for histopathological examination(s). Microscopic examination depicts epidermis with parakeratosis and focal ulceration covered with acute fibrinopurulent exudate, Dermis and subcutaneous show poorly demarcated lesion with proliferation of blood vessels with lymphoid follicles and dense infiltrates of inflammatory cells consisting of lymphocytes, plasma cells and eosinophils, compatible with angiolymphoid hyperplasia with eosinophilia. (Fig. 3 & 4)

Fig. 1 – Clinical photograph of the lesion

Fig. 2 – Photograph after excision and primary closure

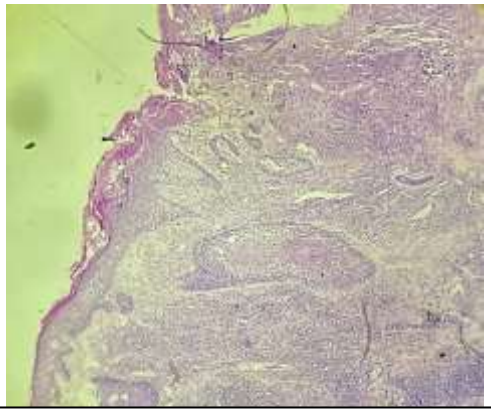

Fig. 3 - H and E sections, (10x) section showing skin with epidermis exhibiting parakeratosis and focal ulceration covered by acute fibrinopurulent exudate. Dermis and subcutaneous tissue show an ill-defined lesion composed of a proliferation of blood vessels with lymphoid follicles and dense inflammatory cell infiltrates consisting of lymphocytes, plasma cells, and eosinophils

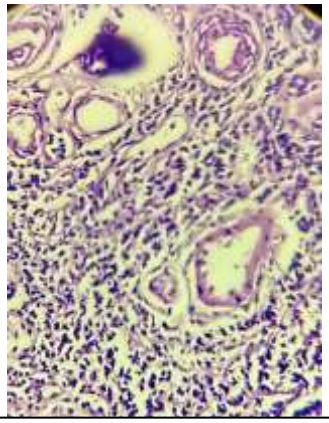

Fig. 4 - H and E sections (40X) section shows proliferation of blood vessels with lymphoid follicle admixed with inflammatory cells comprising of lymphocytes, plasma cells and eosinophils

DISCUSSION

Single or multiple, brown papules or nodules in females are typical of ALHE, especially young to middle-aged adults. They often involve head and neck areas, especially around the ear, forehead and scalp. The exact etiopathogenesis of ALHE is still poorly defined though can be a neoplastic process, a reactive process (hypersensitivity reaction) or inflammatory vascular reaction. It is well established that ALHE can have vaccine related or high oestradiol induced causes, infections and may be secondary to trauma. (2,7,9) In mutational change of TEK gene that encodes an endothelial cell tyrosine kinase receptor Tie-2, involvement was postulated in the pathogenesis of ALHE. (2) ALHE occurs most often in Asians and least often in black people. (9,11)

ALHE is a vascular proliferative disorder with its characteristic pathology being proliferating small blood vessels, an atypical expansive histological feature with a surrounding larger endothelial cell population displaying large vesicle nuclei and representative abundant eosinophilic cytoplasm around a central vascular unit. Lymphocytes and eosinophils are the predominant cell types in the inflammatory infiltrate around the vessels. About 20% of the patients have blood eosinophilia. Lymphocytes diffuse infiltration with or (folicle formation. (2,9) KADE is often mistaken for KD (Kimura's disease). (12) Both of these disorders are very alike and present in head and neck region particularly the periauricular region. (Kimura's disease is a rare chronic inflammatory disorder that most commonly occurs in young Oriental men and manifests as asymptomatic solitary or multiple, large tumors in the subcutis or salivary glands. It is often characterized by regional lymphadenopathy, peripheral blood eosinophilia, hyperimmunoglobulinaemia (serum IgE) and nephrotic syndrome. By contrast, the ALHE commonly presents in young to middle-aged females with a predominance to Caucasians and is characterised by multiple mild pruritic erythematous dermal papules or nodules and an absence of lymphadenopathy or laboratory abnormalities.

The histopathological features of ALHE consist of proliferation of blood vessels of variable size lined by dilated endothelial cells while prominent features of KD are florid lymphoid follicles with germinal centre involvement with eosinophilic infiltrate and fibrosis. (2,7,9,13) Other differential diagnosis includes angiosarcoma, epithelioid hemangioendothelioma, neurilemmoma, leiomyoma, hemangioma, fibrosarcoma. (14)

The first approach is surgical excision, with infrequent recurrence. Systemic medical treatment with intralesional corticosteroids, isotretinoin, topical imiquimod, tacrolimus, interferon α-2a and radiotherapy is always with poor success. Surgical excision, on the other hand, has a relatively low disease Recurrence rate compared to medical intervention, which has demonstrated 80%–100% failure rate. ALHE is an indolent chronic lesion, good prognosis, malignant transformation has also never been reported. (2,9,10,13,15)

Conclusion

While ALHE is a rare disease, it should be in the differential diagnosis for a solitary erythematous papule on the head and neck. Histopathological examination of the always excised tissue should be performed. The ALHE patient should be counselled properly about benign behaviour and chronic course of this disease and about the best modality of treatment is surgical excision with a very low rate of recurrence.

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