Case Report:

Angiolymphoid hyperplasia with eosinophilia

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Abstract

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare benign reactive inflammatory lesion causing blood vessel proliferation and a dense eosinophilic inflammatory infiltrate presents as subcutaneous nodules in middle-aged women's head and neck region. It commonly presents reddish pruritic nodules and papules of the ear and the periauricular region, which is difficult to eradicate. Orbit, colon, peripheral arteries, lacrimal gland, parotid gland, and throat are the extracutaneous sites.

Keywords: Angiolymphoid hyperplasia with eosinophilia, (ALHE)

Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare vascular disorder described in 1969 by Wells and Whimster.[1] This idiopathic condition presents in adults as isolated or grouped papules, plaques, or nodules in the skin of the head and neck. Lesions in the skin of the periauricular region, forehead or scalp are the most presented sites. Hands, shoulders, breasts, penis, oral mucosa and the scrotum are the rarely involved site. Extracutaneous sites have also been noted to include the orbit, peripheral arteries, the colon, the mandible, the lacrimal gland, the parotid gland and the throat.[2-4] It by preference involves females between 20 and 40 years old, being of benign character. Vascular malformation or local trauma are considered as the potential etiological factors. ALHE usually manifests as a cluster of lesions on the skin of the head and neck with classic red-brown nodules. Similar cutaneous lesions may also appear on the trunk, extremities, and genitalia.[2-4] Though the lesions are benign, they may be hemorrhagic (25%) in addition to causing itching (37%) or pain (20%). Peripheral eosinophilia has been found in up to 20% of cases.[5-6] As the name implies, the diagnosis is made histologically by the presence of vascular proliferation (angio-hyperplasia), lymphoid follicles (lymphoid hyperplasia) and a prominent eosinophilic infiltrate (eosinophilia).[7]

Case Report

A 22-year-old patient presents an unremarkable medical history. Initially, she consulted another institute with multiple pruritic right ear auricle skin lesions for one and a half years. The lesions started as reddish papules that gradually enlarged to form nodules with the development of new papules. There was mild bleeding after itching. She noticed ulcerations and discrete bleeding after scratching caused by pruritus. On examination, discrete, well-defined, erythematous papules and nodules of different sizes were found over the right auricular and post-auricular region and the nodules were firm and tender. She didn't suffer local trauma or any constitutional symptoms (Pic. 1 & 2). Her physical examination proved insignificant, without any lymphadenopathies or salivary gland enlargements. Dermoscopy revealed Yellow to orange areas on the lesions intersected by fine horizontal telangiectasia. Consequently, a laboratory workup and an excisional biopsy of one dermatologic lesion were conducted. The Biopsy exhibited Mild acanthosis in the epidermis, dense perivascular infiltration of lymphocytes and eosinophils with thick-walled blood vessels in the dermis.

The patient underwent electrofulguration for the skin lesions, which resulted in complete resolution with no recurrence. This successful outcome demonstrates the potential effectiveness of this treatment method for ALHE. Follow-up: Six-month monthly follow-ups were done,

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followed by six-month follow-ups every six months for any possible progression, the appearance of new lesions, and potential therapeutic changes. No new lesion was seen up to 1 year.

Discussion

ALHE is a benign, slowly growing tumour with a still unclear etiopathogenesis. It can be self-limited and characterised by intense vascular proliferation.[7] More clarity exists in the literature regarding ALHE and Kimura's disease. Both present nodules, preferably on the head and cervical region but in ALHE, they tend to be much more erythematous than in Kimura's disease, where the lesions are normochromic. They also have similar histopathological features, such as involvement of the dermis and subcutaneous infiltrate comprising lymphocytes and eosinophils, proliferation of endothelial cells and the absence of adnexal structure involvement.[3,7] Asian males are more prone to Kimura's disease, which has a triad of painless subcutaneous masses, usually unilateral, in the head and neck region. There is peripheral eosinophilia and a significant increase in immunoglobulin E levels in the tissues. Salivary gland enlargement may also occur. Histopathologically Kimura's disease differs from ALHE for being mainly a disorder of the lymphoid follicles, without irregular blood vessels and with non-protruding endothelial cells in the vascular lumen but always presenting an increased number of eosinophils that may extend to the muscular fascia.[8]

Surgical excision is indicated where relapse is common. Other reported treatments include cryotherapy, local radiotherapy, topical or intralesional corticosteroids, imiquimod, acitretin and laser therapy (dye laser, CO2 laser). While these treatments may be effective, they also risk potential complications or side effects, which should be considered when choosing a therapeutic approach.

Follow-up: Six-month monthly follow-ups were done, followed by six-month follow-ups every six months for any possible progression, the appearance of new lesions, and potential therapeutic changes.

Conclusion

Angiolymphoid hyperplasia with eosinophilia presents a therapeutic dilemma due to the wide variety of proposed treatments, but there is a lack of comprehensive data on most of them. Although the disease is not deadly by itself, it usually presents with disfiguring lesions that can severely affect the patient's quality of life. This underscores the urgent need for further research and concerted efforts to find an effective cure and a unified therapeutic approach.

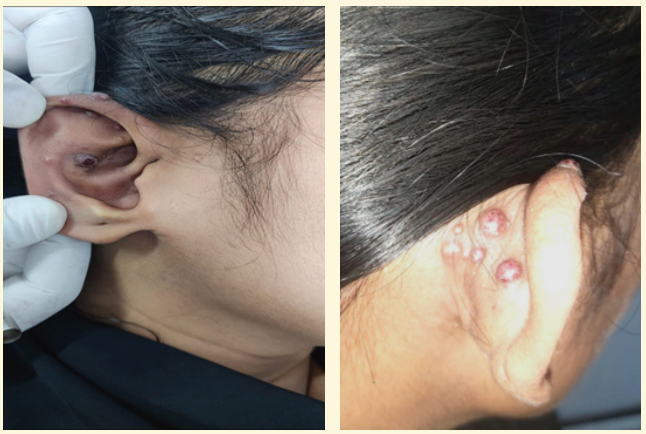

fig 1. Discrete, well-defined, erythematous papules and nodules on right auricle (a) and post auricular region (b) before treatment

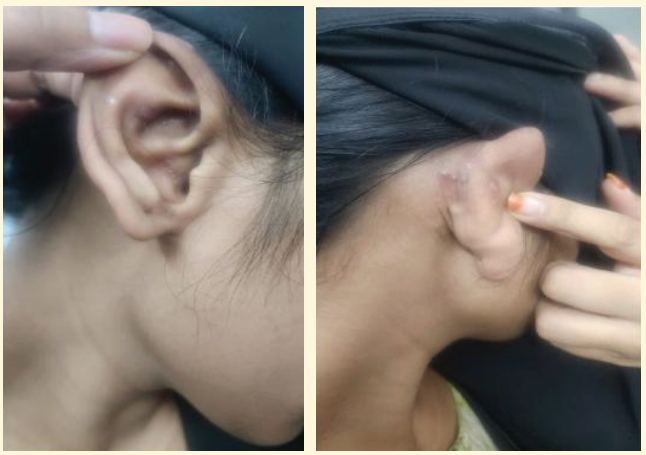

fig 2. post-treatment follow up after one year

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