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Angiolymphoid Hyperplasia with Eosinophilia

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Abstract: Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon idiopathic condition that manifests in adults as isolated or grouped papules, plaques, or nodules in the skin of the head and neck. It manifests with lesions in the periauricular region, forehead, or scalp. Rare sites include the hands, shoulders, breasts, penis, oral mucosa, and orbit. We have come across a case of ALHE in our hospital with the patient presenting popular lesions on the skin of the neck and head.
Keywords: Angiolymphoid hyperplasia with eosinophilia, Hemangioma, Histology

INTRODUCTION

In 1969, Wells and Whimpster described Angiolymphoid hyperplasia with subcutaneous eosinophilia (ALHE) after studying nine patients with persistent subcutaneous nodules in the head and neck [1]. Wilson-Jones and Bleehen described similar lesions in the same year [2]. In 1983, Enzinger and Weiss suggested the expression "epithelioid hemangioma" for this uncommon and distinct vascular formation [3].

ALHE is also known as epithelioid haemangioma, pseudopyogenic granuloma, inflammatory angiomatous nodule, papular angioplasia, subcutaneous angioblastic lymphoid hyperplasia with eosinophilia and lymphofolliculosis, intravenous atypical vascular proliferation and histiocytoid haemangioma [4].

Lesions may also involve the inside of the mouth or genitals. They may be brown or red in color, asymptomatic, itchy or painful. Individual nodules rarely exceed 2-3 cm in diameter, although occasionally they are larger and extend more deeply [5]. Some cases clear up without treatment, after a variable period of time. Frequency in the United States is unknown. ALHE is uncommon but not rare. Although frequency is unknown, cases have been reported worldwide. It may be more common in Japan than in other countries [6].

Though ALHE can persist for years, but serious complications do not occur [7]. A few cases of nephropathy have been reported in patients, although the association is not strong [8]. ALHE is slightly more common in females; however, a male predominance has been noted in selected Asian studies. ALHE presents most commonly in patients aged 20-50 years, with mean onset of 30-33 years [9]. This condition is rare in elderly patients and in the non-Asian pediatric population [7]. The cause is unknown, but antigenic stimulation following insect bites has been postulated [4]. It sometimes follows an injury [5].

CASE REPORT

In our case we had the biopsy findings of sub epidermal lymphoid aggregates with good number of eosinophils. (Fig.1). There was blood vessel proliferation with prominent sub epidermal lymphocytic infiltration with eosinophils in the interstitium (Fig. 2). The peripheral blood smear examination revealed increased number of eosinophils (16%).

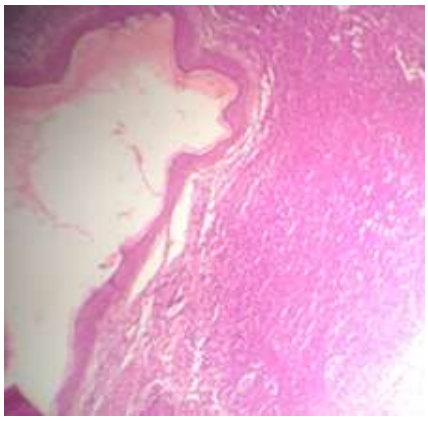

Fig. 1: Showing subepidermal lymphocytic aggregation

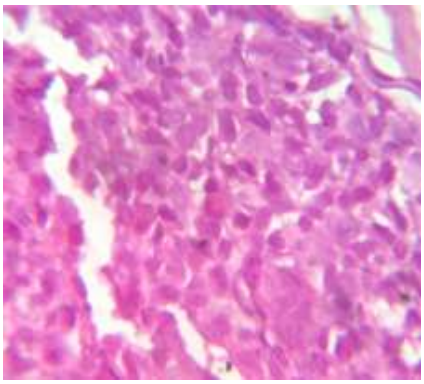

Fig. 2: Showing eosinophils in the subepidermal area

DISCUSSION

Differential diagnoses of ALHE includes granuloma faciale, insect bites, Kimura disease, lymphocytoma cutis, pyogenic granuloma (Lobular Capillary Hemangioma), sarcoidosis [10].

Granuloma faciale may present as reaction to localized persistent immune complex deposition or hypersensitivities to a persistent antigen. There may be neutrophilic infiltration with vasculitis and few eosinophils.

Kimura disease is a rare chronic inflammatory disease that presents as a tumor-like swelling in the head and neck region and is often associated with regional cervical lymphadenopathy. Histopathologically, this condition is characterized by lymphocytic inflammatory infiltrate, forming lymphoid follicles interspersed with aggregates of eosinophils and variable fibrosis in a richly vascular stroma. Eosinophilia in the peripheral blood and in tissues and marked increase in serum levels of immunoglobulin E (IgE) [11, 12].

Lymphocytoma cutis is a benign cutaneous lymphoid hyperplasia due to an excessive immune response to antigen. Skin biopsies show superficial and deep angiocentric, neurotrophilic and eccrinotropic lymphocytic infiltrates, often accompanied by plasma cells and eosinophils, the former at the periphery and the latter in the center of lesions.

Pyogenic granuloma is a common proliferative lesion, often occurs shortly after a minor injury or infection of the skin. The typical lesion presents as a polypoid mass of angiomatous tissue protruding above the surrounding skin. Inflammation is usually slight in the deeper part of the lesion. It may be absent when the epidermis is intact. The angiomatous tissue tends to occur in discrete masses or lobules, resembling a capillary hemangioma [13].

ALHE shows characteristic histologic features that include a proliferation of small blood vessels, many of them are lined by enlarged endothelial cells with uniform ovoid nuclei and intracytoplasmic vacuoles. These distinctive endothelial cells have a cobblestone appearance. In addition, a perivascular and interstitial infiltrate composed primarily of lymphocytes and eosinophils is present [14].

CONCLUSION

The etiology of ALHE is still unknown, and proposed pathogenesis includes a neoplastic process, a hypersensitivity reaction, inflammatory vascular reaction or a tissue reaction to a previous trauma as seen in cases of acquired traumatic A-V fistulas.

Histopathologically, the conditions can be distinguished at first with a low microscopic magnification and other characteristics detected with higher magnification are useful to confirm diagnosis and enable the differentiation.

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