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Clinical Record

Angiolymphoid hyperplasia with eosinophilia of the auricle: progression of histopathological changes

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Abstract
Angiolymphoid hyperplasia with eosinophilia is a rare condition that demonstrates dermal or subcutaneous proliferation of endothelial cells associated with an inflammatory cell infiltrate. A case is reported, with emphasis on the histopathological features on repeated biopsies. The report serves to stress the importance of considering this condition in the differential diagnosis of lesions in and around the ear.

Key words: Angiolymphoid Hyperplasia with Eosinophilia; Kimura Disease; Ear, External

Introduction

Angiolymphoid hyperplasia with eosinophilia is a rare vascular disorder of unknown pathogenesis. It is characterized by intradermal or subcutaneous reddish brown papules and/or nodules, typically occurring in the head and neck region.[1] Histologically, it is composed of a proliferation of vascular channels with a surrounding infiltrate of lymphocytes, macrophages and eosinophils.[2] The term was first described in the English literature in 1969 by Wells and Whimster.[3] It is related to Kimura's disease, described in Japan and China and first reported in the Japanese literature in 1948.[4]

Although the ear and periauricular areas are the most typical sites of development in all races,[5,6] the author could not find a report of this condition in any otolaryngological journal. The aim of this report is to document a case with this rare condition and to illustrate the progression of the histopathological features over time.

Case report

The patient was a 50-year-old, male, insulin-dependent diabetic and hepatitis C carrier who worked in the car industry. He first presented to our institution in February 2000 with a small, red nodule at the floor of the right auricular concha. There was no previous history of trauma or insect bite. The lesion failed to resolve with conservative measures. Two months later, a biopsy was taken under local anaesthesia. The histopathological features were non-encapsulated, dermal vascular proliferation and lymphocytic and histocytic infiltration, with areas of fibrosis and hyalinization. There were minimal eosinophils in the specimen (Figure 1). The condition was diagnosed as an inflamed fibroepithelial polyp. The patient refused surgery and did not attend follow up.

The patient was later seen in September 2004, by which time the lesion had enlarged to about 2.5 cm in diameter. It involved the tragus and blocked the ear canal (Figure 2). The lesion was intensely pruritic and frequent episodes of bleeding were noted. There was no cervical lymphadenopathy. A full blood count revealed a normal eosinophil count and liver function tests showed normal values. A chest radiograph showed no radio-opaque lesions. A further biopsy was taken, the histopathology of which differed from the first biopsy in revealing intense eosinophil cellular infiltrate and lymphoid cells arranged in follicles without germinal centres around the proliferating blood vessels (Figure 3). The condition was diagnosed as angiolymphoid hyperplasia with eosinophilia. Intralesional steroid injections were successful in reducing the size of the lesion, and further resolution of the swelling was achieved by argon laser treatment at the Laser Institute. At the last follow-up appointment, nine months after starting laser treatment, the condition had completely resolved.

Discussion

Although cutaneous vascular tumours are relatively uncommon, they have received much attention due to the acquired immunodeficiency syndrome (AIDS) epidemic. Because one of the early signs of AIDS is the appearance of Kaposi sarcoma, it became necessary to distinguish this disease from other vascular lesions.[7]

The common denominator in all cases reported as angiolymphoid hyperplasia with eosinophilia is the appearance of proliferating endothelial cells. There is controversy as to whether the condition is reactive or neoplastic. Burrall *et al.* (1982)[8] postulated that the condition may initially be reactive, followed by neoplastic proliferation of stimulated endothelial cells. *In vitro* studies have demonstrated a direct antiproliferative effect of recombinant human interferon alpha, beta and gamma on human dermal microvascular endothelial cells.[9] Interferon alpha has recently been successfully used clinically for alleviation of this condition.[1,5]

In the above patient, the progression of appearance of the inflammatory cell infiltrate showed that, over time, the lymphocytes became arranged into follicles around the blood vessels. The follicles showed no germinal

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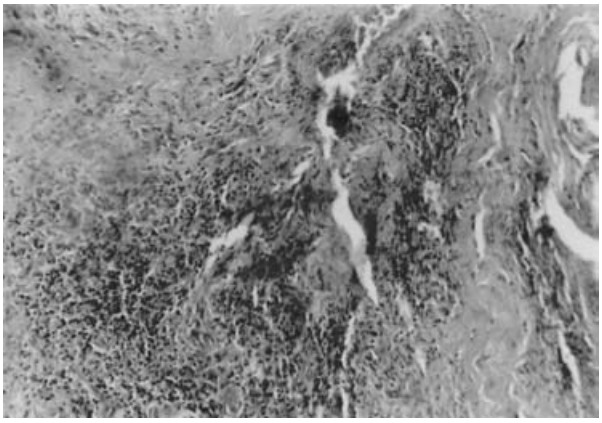

FIG. 1
Histopathology of lesion at early stage, showing vascular proliferation and lymphohistiocytic infiltrate (H & E; × 100).

centres and there were no plasma cells, suggesting that the lymphocytes may have been mostly T-lymphocytes. Eosinophils became much more numerous in the lesion but there was no peripheral blood eosinophilia. The absence of peripheral eosinophilia in repeated blood examinations, despite the long course of the disease, may indicate that angiolymphoid hyperplasia with eosinophilia is a different condition from Kimura's disease, the latter being associated with marked peripheral eosinophilia.[10]

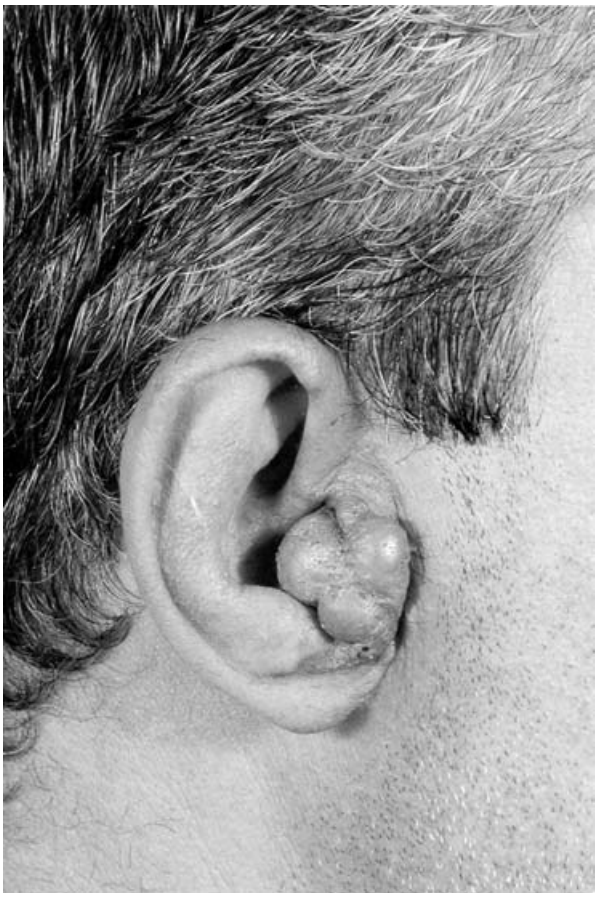

FIG. 2
The lesion in 2004 (reproduced with permission of the patient).

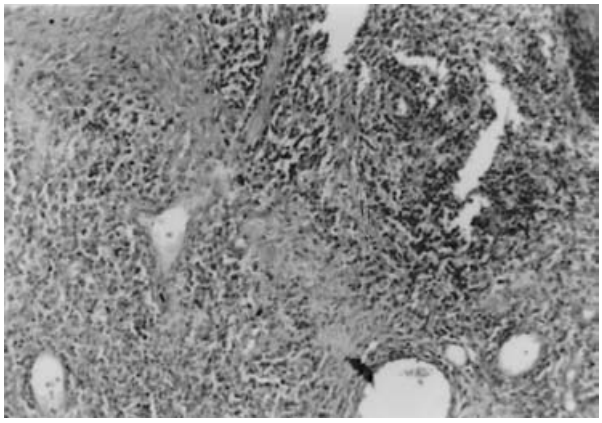

FIG. 3
Histopathology of late stage lesion, showing vascular proliferation, eosinophil infiltration and lymphoid follicles (H & E; × 100).

Several treatments have been reported for angiolymphoid hyperplasia with eosinophilia. These include surgical excision, laser, irradiation, corticosteroids (intralesional and systemic), cryotherapy and chemotherapy.[11,12] The patient in question refused surgery, even in the early stage of the disease. He was successfully managed by intralesional steroids and laser therapy.

Conclusion

Angiolymphoid hyperplasia with eosinophilia is a distinct clinical and pathological condition. Clinically, it presents as a red, papulonodular lesion, typically in the region of the auricle. Pathologically, the central feature of the disease is vascular proliferation. The surrounding inflammatory process develops over time into lymphoid follicles and local eosinophilia.

- **Angiolymphoid hyperplasia with eosinophila is a rare condition characterized by dermal or subcutaneous proliferation of endothelial cells, with an inflammatory infiltrate**
- **This report describes a case affecting the floor of the right concha. The author discusses the clinical and pathological features**

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