Angiolymphoid Hyperplasia with Eosinophilia in the Lower Eyelid: A Case Report

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ANGIOLYMPHOID HYPERPLASIA WITH EOSINOPHILIA IN THE LOWER EYELID

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Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE), also called epithelioid hemangioma is a benign vascular tumor first described by Wells and Whimster in 1969. It has a female predominance with most women presenting in the third or fourth decade. It usually occurs in the head and neck area, especially the periauricular region and rarely in other parts of the body, including the trunk, extremities, hands, colon, oral cavity and genitals. It presents as single or multiple flesh to plum-colored papules or nodules, measuring a few to several centimetres. We present an unusual case of a single, firm papule of ALHE presenting on the lower eyelid of a 39-year-old female, making the diagnosis challenging.

Case Report

A 39-year-old female presented to the ophthalmology clinic one year ago with a papule on the right lower eyelid. The mass was excised but no biopsy was sent for histopathological examination.The patient presented 10 days ago with a lesion in the same region. It was a painless, non-pruritic, hemorrhagic and erythematous 5 mm mass with an overlying black area. (Figure 1) The patient reports that the mass bled on several occasions. She also complained of blurry vision. The patient has no history of trauma or any medication use.

On gross examination, a single skin-covered piece of tissue measuring 1.0 x 0.7 x 0.5 cm was seen. There was an overlying blackish area, measuring 0.3 x 0.3 cm. Microscopy revealed a number of lymphoid aggregates (Figure 2) surrounded by edematous stroma and inflammatory infiltrate rich in eosinophils. (Figure 3). Plump endothelial cells were also identified around vessels which are characteristic of ALHE. PAS was negative for fungal organisms. There was no evidence of malignancy. Hence a diagnosis of ALHE was made.

Figure 1: Clinically patient presented with a 5 mm papule in the right lower eyelid.

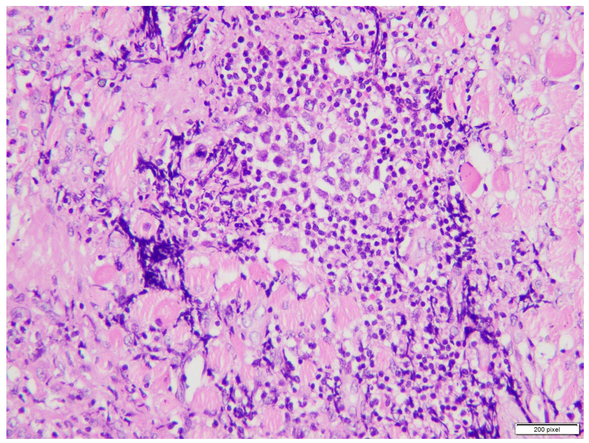

Figure 2: Photomicrograph showing lymphoid aggregates.

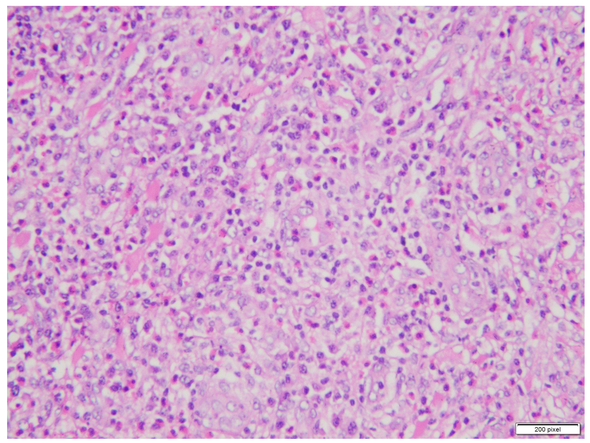

Figure 3: Photomicrograph showing dense inflammatory infiltrate with lymphocytes and numerous eosinophils.

Discussion

ALHE presents as a papule or subcutaneous nodule, which is usually painless but may be painful, hemorrhagic or pruritic.. A definitive diagnosis is made on histopathological examination.

ALHE on histopathological examination presents as a vascular bundle surrounded by plump, vacuolated endothelial cells, which are also called ‘epithelioid’ or ‘histiocytoid’ due to their appearance and are a hallmark feature of ALHE. The lesion is also known as an ‘epithelioid hemangioma’ because of these endothelial cells.

The cells are ‘hobnailed’ due to the cytoplasmic vacuoles in them. The cytoplasm protrudes into the lumina of the blood vessel. (Figure 4) There is a dense inflammatory infiltrate, which includes lymphocytes, prominent eosinophils and mast cells. The lymphocytes may aggregate to form follicles composed of germinal centers. There may be a peripheral eosinophilia. ALHE usually recurs if excision is incomplete. Its etiology is unknown but there are currently a few suggested hypotheses: a neoplastic, reactive, or infectious process associated with HIV. Arteriovenous shunts, elevated serum estrogen levels and trauma also seem to be contributing factors. Our patient denied any trauma or taking oral contraceptives.

Angiolymphoid hyperplasia with eosinophilia is a benign condition that must be differentiated from Kimura’s disease.

The other differential diagnoses include a hemangioma, angiosarcoma, Kaposi sarcoma, pyogenic granuloma, subcutaneous metastases and eccrine hydrocystoma.

Surgical excision is the treatment for ALHE with recurrence in one-third of the cases in case of incomplete excision. Other treatment options are oral and intralesional steroids, combined pulsed dye and CO2 laser, cryotherapy, chemotherapy, imiquimod, tacrolimus, methotrexate and cyclophosphamide. Our patient was given intralesional steroids and the mass was excised subsequently. Recent studies suggest that as it is a benign condition, observation is recommended for spontaneous regression for 3-6 months before surgical excision. The lesion may ulcerate and bleed, hence treatment is required for both aesthetic and functional reasons.

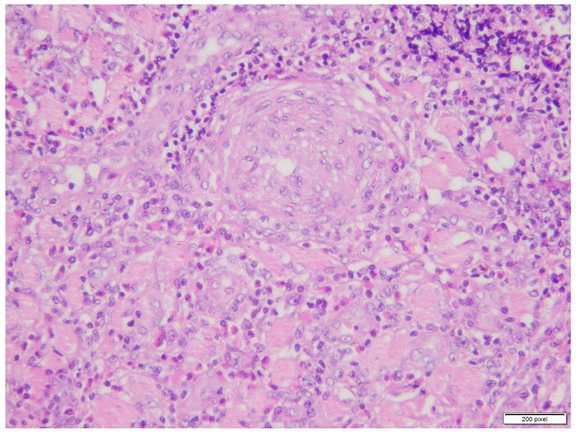

Figure 4: Photomicrograph showing epithelioid endothelial cells in a blood vessel, surrounded by lymphocytes and eosinophils.

Conclusions

ALHE has a recurrence of 33% if not completely excised. Although a rare occurrence, it should be considered in the differential diagnosis of an eyelid mass.

References

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Disclosure: NOTHING TO DISCLOSE