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Subcutaneous Angiolymphoid Hyperplasia With Eosinophilia: A Diagnostic Dilemma

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Abstract:
Background*: Angiolymphoid hyperplasia with eosinophilia (ALHE), also named epithelioid hemangioma, is an uncommon, benign vascular proliferation. It is characterised by dermal or subcutaneous red or brown papules or nodules, most commonly on the head and neck region. Histologically, it is characterised by a vascular proliferation with epithelioid endothelial cells, accompanied by a surrounding lymphocytic and eosinophilic infiltrate.*
Conclusion: *Angiolymphoid hyperplasia with eosinophilia is a rare epithelioid vascular tumour with a challenging clinical and histological diagnosis. Despite its benign nature, ALHE causes a therapeutic dilemma. Hence, a high index of clinical suspicion is warranted for the diagnosis and appropriate treatment. To avoid overtreatment, it is important to consider the diagnosis of angiolymphoid hyperplasia with eosinophilia in such cases as the implications regarding prognosis and the mode of therapy vary considerably.*
Keywords: *Angiolymphoid hyperplasia with eosinophilia, wide local excision, Kimura disease, split thickness skin graft, squamous cell carcinoma of the pinna, rare ear swelling*

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I. Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare vascular tumor with a tendency to occur in the head and neck of middle-aged adults. Although benign, the tumor may cause pain, bleeding, or pruritus. Surgical excision is considered the most effective treatment among many options, but treatment failure still exceeds 40% with surgery. Here we report a case of ALHE that was treated with wide local excision with split-thickness skin graft reconstruction.

II. Case Report

A 70-year-old male presented to our Outpatient department, with multiple swellings over the left pinna for the past 4 years following a finger nail prick injury a few months prior. It was gradually progressing, associated with itching and bleeding on scratching. On examination, an exophytic mass in the left conchal bowl of pinna extending to external auditory canal and the symba concha and two swellings approximately 4cmx3cm and 3cmx2cm in the medial aspect of the pinna, firm in consistency, non-tender, and no palpable neck nodes (Fig.1A, 1B). Routine baseline investigations were found to be within normal limits. Contrast CT showed large multilobulated, heterogeneously enhancing predominantly hyperdense mass in skin and subcutaneous plane of left external auditory canal extending to outer surface of pinna as well as skin over left mastoid. FNAC suggestive of Invasive squamous cell carcinoma-moderately differentiated. Patient underwent widelocal excision of the mass. After excision, the medial aspect of the pinna repaired by primary suturing. The defect in the lateral aspect of the pinna was reconstructed by split thickness skin graft harvested from anterolateral aspect of left thigh of the patient under general anaesthesia (Fig.3A, 3B). Histopathological examination showed eosinophilic plump and vacuolated endothelial cells in small groups and sheets within a fibrous background, accompanied by lymphoid

aggregates and eosinophils. (Fig.4A, 4B). On 8th post operative week wound was completely healed, and patient was followed up for 30 months during which no recurrence was noted.

III. Discussion

In 1969 Wells and Whimster coined the term Angiolymphoid hyperplasia with eosinophilia (ALHE).[1] It is characterized by a florid proliferation of blood vessels lined by plump endothelial cells and admixed with a dense inflammatory infiltrate of lymphocytes, eosinophils, and mast cells. Other names include histiocytoid hemangioma, angiomatous nodule, pseudopyogenic granuloma, inflammatory angiomatous nodule,[2,3] but angiolymphoid hyperplasia with eosinophilia and epithelioid hemangioma remain the current, unanimously accepted terms for this entity. More common in female but some studies also showed male preponderance.[4] Size ranges from a few millimetres to 10 cm. There does not appear to be any racial preponderance, although the majority of cases have been reported in the Japanese or Chinese literature. In our case, it was a male patient. The aetiology is currently unknown. Various hypotheses have been put forth, including the reactive process, neoplastic process, infectious mechanisms with possible association with human immunodeficiency virus, arteriovenous shunting, insect-vector-induced organism, immunologic reactions, local trauma, hormonal factors like hypothyroidism, pregnancy, elevated serum estrogen levels, increase in lesion size during menstruation, is reported suggests a hormonal mechanism.[3,5,6,7,8,9] However, none have proven to be conclusive or definitive. Our patient developed the lesion after finger nail trauma. They are frequently located on the head, which is involved in (81.8%) of cases in previous reports. The most common locations are periauricular area (36.3%), face (28.2%), and scalp (17.3%).[10] Other locations such as trunk, lower extremities, hands, penis, oral mucosa, and colon.[5,10–13] There are usually no associated symptoms. However, owing mainly to the vascular nature of the lesions, tenderness, pulsation, pruritus (36.8%), ulceration, bleeding (25.3%), either spontaneously or after minor trauma, may occur in some patients. Peripheral blood eosinophilia and regional lymphadenopathy may also be present.[1,2,3] The length of time from onset of lesions to 1st clinical visit has ranged from a few months to many years (1 month to 7 years). Histologically, the lesions are characterized by two major elements: (1) the vascular, which preponderates in early, more active cases, and (2) the lymphoid, which preponderates in the later quiescent stages. Other important diagnostic features include diffuse mast cell and eosinophil infiltration of the dermis and subcutaneous tissues, as well as serum eosinophilia 10–20%. IgE is often normal, one study showed an elevated level was noticed in 11% of the cases. ALHE can be intradermal or subcutaneous. The subcutaneous form is the florid proliferation of large epithelioid endothelial cells that may become so exuberant as to form solid intraluminal nodules or clusters. Thus, it may be perfidious and challenging to diagnose in that these masses may obscure the vascular nature of the lesion. In contrast, dermal form tends to be less circumscribed, smaller, and composed of more mature open vessels lined by smaller, less epithelioid endothelial cells.[1,2,3] Differential diagnosis includes Kimura disease, Angiosarcoma, Kaposi sarcoma, eosinophilic granuloma, malignant lymphoma, granuloma pyogenicum, angiomatous lymphoid hamartoma, persistent reaction to insect bites, cutaneous lymphoma, skin metastases, sarcoidosis, keloid.[14,15] Spontaneous recovery was rarely reported. Surgical excision is the treatment of choice.[2,3] One-third of cases that are incompletely excised do recur, either at the same site or distant from it, but still typically along the course of the affected vessel. Other types of procedures, such as Mohs micrographic surgery, pulsed-dye laser, carbon dioxide laser, and argon laser, cryosurgery, irradiation have been applied and reported. Adequate local surgical excision seems to be the treatment of choice, but the highly vascular nature of the lesion often makes margins difficult to identify and thus CO_2 laser under microscopic control provides a good and dry field with accurate microscopic delineation of the margins.[8] The nonsurgical methods include steroids (local and systemic), methotrexate, pentoxifylline, propranolol, isotretinoin, thalidomide, phototherapy, as well as new therapies targeting cytokines including topical imiquimod, mepolizumab, bevacizumab, and interferon alpha-2a.[15] According to Adler et al.,[10] treatment failure was low with excision (40.8%), pulsed dye laser (50.0%), or carbon dioxide laser (54.6%); intermediate for argon laser (66.7%), intralesional corticosteroids (79.1%), or cryotherapy (80.5%); and high for systemic (87.8%) and topical (98.2%) corticosteroids, oral pentoxifylline, and isotretinoin (100%). Besides, higher recurrence rates are seen in ALHE with earlier age of onset, longer duration, multiple lesions, and symptomatic lesions.[10]

IV. Conclusion

In conclusion, our study demonstrates that although ALHE is a benign epithelioid vascular tumor, it represents a challenging diagnosis as in our preoperative FNAC showed moderately differentiated invasive squamous cell carcinoma. hence requires high index of clinical suspicion despite rarity of the disease and intraoperative finding to avoid radical surgeries. Surgical excision with or without reconstruction of the defect is the most effective treatment. Post operative follow up is must since the lesion carries risk of recurrence.

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Figures

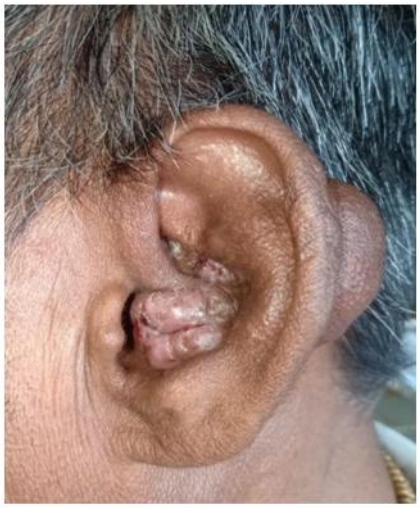

Fig. 1A Exophytic mass in the left conchal bowl of pinna extending to external auditory canal

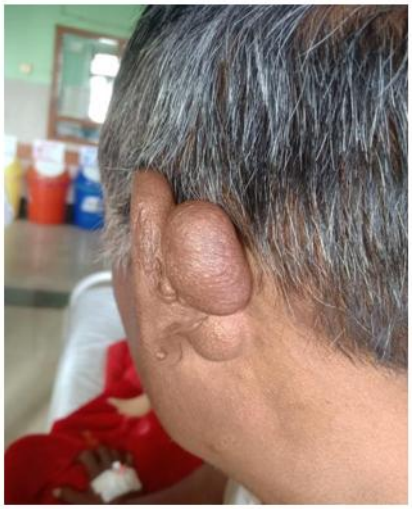

Fig. 1B Two soft to firm swelling in the medial surface of the left pinna

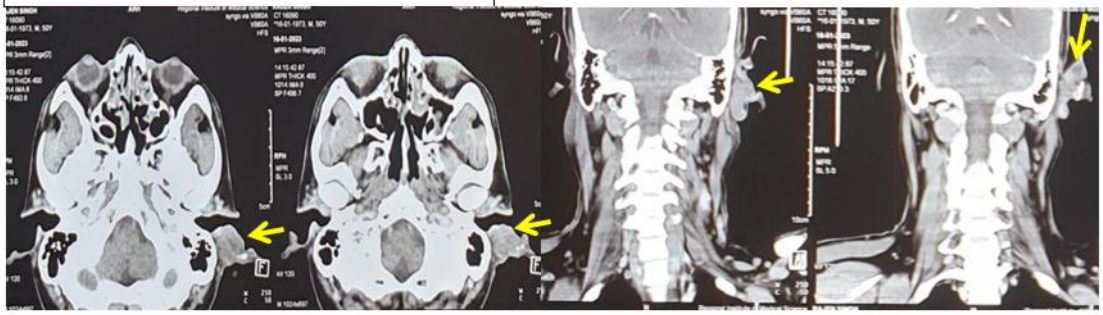

Fig. 2 Contrast CT showing, multilobulated, heterogeneously enhancing mass in skin and subcutaneous plane of left external auditory canal extending to outer surface of pinna (Yellow arrows)

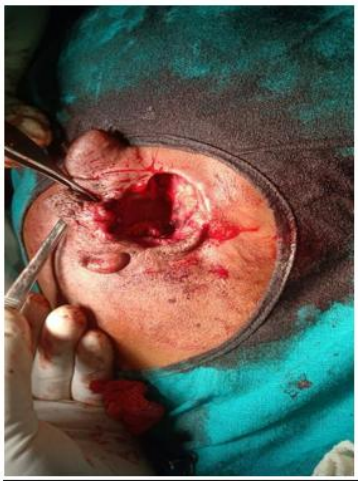

Fig. 3A Intraoperative picture: after the wide local excision of the lesion with defect

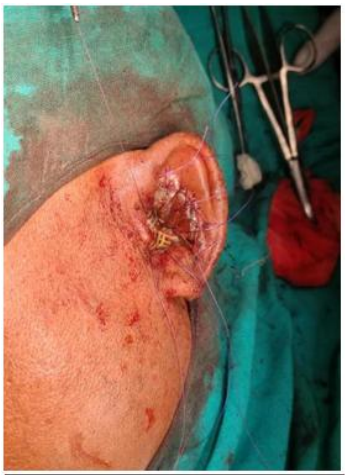

Fig. 3B lateral aspect of pinna defect was repaired with Split thickness skin graft

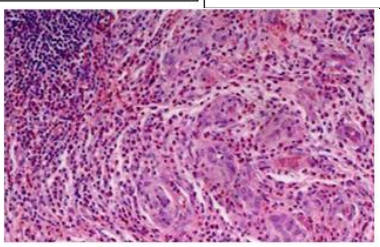

Fig. 4A Eosinophilic plump and vacuolated endothelial cells in small groups and sheets within a fibrous background, accompanied by lymphoid aggregates and eosinophils