Angiolympoid Hyperplasia with Eosinophilia of the Forearm: A case report

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Abstract: A 35 years old female patient presented with swelling of the forearm. Histopathological evaluation confirmed the diagnosis of angiolympoid hyperplasia with eosinophilia. It is a benign vascular proliferative disease also called as histiocytoid or epitheloid haemangiomma mainly occurring in head and neck(around the ear). Etiology of the lesion is unknown. Various treatment modalities have been described. We present a case successfully treated with surgery.

Keywords: Angiolympoid Hyperplasia with Eosinophilia (ALHE); Epithelioid Haemangiomma; Histiocytoid Haemangiomma; Kimura’s Disease.

I. Introduction
Angiolympoid hyperplasia with eosinophilia is also called as histiocytoid haemangiomma or epitheloid haemangiomma. It is characterised by one or more purplish, brownish papule or subcutaneous nodule with a predilection for head and neck region (around the ear).1

II. Case report
A 35 years old brown skinned female patient presented in the surgery department complaining of a nodular lesion on the ventral side of forearm just below elbow on the medial side. The lesion caused slight pain to time to time but there was no significant past medical history. On examination the overlying skin did not show any abnormality, nontender, soft to firm in consistency with restricted mobility and subcutaneous in location. No hepatosplenomegaly and lymphadenopathy was observed. CT angiography showed hypodense enhancing hypervascular mass with multiple internal vessels in it. An excisional biopsy was performed and lesion was removed en masse. Formalin(10%) fixed whitish tissue was received. The size of the specimen was 2.5×2×1.5 cm with smooth external surface. Cut surface was grey white with grey brown areas in between and without necrosis (figure 1). Microscopy showed a benign vascular lesion characterised by proliferation of blood capillaries with thick vascular walls (figure 2) and considerable quantity of eosinophils. There are lymphoid follicles with germinal centres (figure 2) and epitheloid or histiocytoid appearance of endothelial cells (figure 3). Variable amount of inflammatory cells like eosinophilic (figure 4), lymphocytes, plasma cells were present. The cellular atypia and mitoses were not identified. The diagnosis was straightforward on histopathological evaluation and did not require immunohistochemical confirmation. The patient is still in clinical follow up and has not suffered a relapse.

III. Discussion
Angiolympoid hyperplasia with eosinophilia is an uncommon benign vasoproliferative disease. It presents most commonly in the patients aged 20-50 years with a mean onset of 30-33 years2. It is more common in Asians, followed by Caucasians. Although less commonly blacks too can develop the disease. It is rare in elderly patients and in non asian paediatric population3. It mainly occurs in head and neck region but other tissues such as orbit, heart, bone, liver, and spleen may also be involved. It is more common in middle aged females. Contrary to the name suggests, peripheral blood eosinophilia is not a constant feature4. It is a benign slowly growing tumor that can be self limited and characterised by intense vascular proliferation. Much confusion exist between angiolympoid hyperplasia with eosinophilia and kimura’s disease as they both present as nodules preferably on the head and cervical region, but in angiolympoid hyperplasia with eosinophilia they tend to be much more erythematous as opposed to kimura’s disease, where lesions are normochromic. They also share histopathological similarities, such as involvement of dermis and subcutaneous infiltrate comprising of lymphocytes and eosinophils, proliferation of endothelial cells and absence of adenexal structure involvement5(6). Kimura’s disease shows typical lymphoid follicles. It is associated with lymphadenopathy and is always accompanied by peripheral blood eosinophils and these two findings are absent in ALHE7. Other major
alternative to rule out in differential diagnostic are – salivary gland tumors, haemangioma, kaposi sarcoma, lymphoma, insect bites and pyogenic granuloma. It is proposed that the lesion occur with greater frequency in atopic individuals like asthmatics. Candida albicans skin testing demonstrates immediate reaction with elevation of IgE in patient sera. Surgical excision is the preferred method for the treatment of ALHE. Alternative therapies include electrodissection, curettage, radiotherapy, cryotherapy, chemotherapy, corticosteroids and agents like IF α 2b. Spontaneous remission in such cases is possible with in months but recurrences are frequent. Treatment is necessary in symptomatic cases and in situation that alter patients appearance. In this cases, surgical excision was preferred.

Figures

(Fig. 1)

(Fig. 2)
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IV. Conclusion

Angiolymphoid hyperplasia with eosinophilia is a benign vascular lesion occurring more commonly in middle aged females. There is no peripheral blood eosinophilia. Surgical excision is the treatment of choice. There is no evidence of malignant transformation.
Acknowledgements

My gratitude to Dr. Neelu Gupta HOD & Professor , Department of Pathology , SPMC Bikaner for allowing me to publish this case report . Thanks are also due to Dr. Sunita Kulheri for her assistance with the photografhic work.

References