e - ISSN - 2349 - 8005

INTERNATIONAL JOURNAL OF ADVANCES IN CASE REPORTS

Journal homepage: www.mcmed.us/journal/ijacr

ANGIOLYMPHOID HYPERPLASIA WITH EOSINOPHILIA (EPITHELOID HEMANGIOMA) OF THE PERIANAL REGION: AN UNUSUAL PRESENTATION

Jayakar Thomas[1*], Bhairavi M[2], Najad Najeeb[2] and Manoharan D[3]

[1]Professor & Head, [2] Junior Residents, [3]Professor,
Department of Dermatology, Sree Balaji Medical College & Bharath University, Chennai 600044, Tamilnadu, India.

Corresponding Author: - **Jayakar Thomas**
E-mail: jayakarthomas@gmail.com

Article Info
Received 15/06/2015
Revised 27/06/2015
Accepted 12/07/2015

Key words:
Angiolymphoid hyperplasia with eosinophilia (ALHE), Epitheloid Haemangioma, clinical, histological.

ABSTRACT
Angiolymphoid hyperplasia with eosinophilia (ALHE) is a unique entity that usually occurs as less than 3 cm pink to reddish brown dermal nodules of the head & neck. Here we report an unusual occurrence in size, site and appearance of ALHE in the groin of an adult male.

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE), epitheloid haemangioma, inflammatory angiomatous nodule, atypical granuloma, pseudopyogenic granuloma, and histiocytoid hemangioma are synonyms for this rare, benign, reactive vasoproliferative disease, essentially as a malformation of blood vessels due to an underlying ateriovenous shunt, as a result of damage to and repair of an artery or vein. It presents as painless, vascular dermal and subcutaneous nodules of the head and neck, commonly around the ear and lip [1, 2]. ALHE has also been reported in the oral mucosa [3], [orbits] [4] and the conjunctiva [5]. ALHE has a distinctly noteworthy histopathology, marked by a proliferation of blood vessels with large endothelial cells, accompanied by an inflammatory infiltrate that includes eosinophils. ALHE occurs more in females than males [7]. It can persist for years, but serious complications (e.g., malignant transformation) do not occur and have never been reported. We report an unusual case of a large ALHE in a male in the perianal region.

CASE REPORT

An otherwise healthy 40-year-old Indian male presented to our department with a 2cm × 5 cm solitary mass in the perianal region, lying superficial to the skin. It had slowly enlarged over the last one year. There were few erosions, but no obvious cutaneous ulceration, crusting or discolouration which are commonly associated with ALHE. The mass was non-tender, fixed, with a lobulated surface possessing a solid consistency when palpated [Fig 1]. No regional lymphadenopathy was observed. Routine blood and urine investigations were within normal limit.

Chest screening showed no abnormality. Complete excision biopsy of the mass was performed under local anaesthesia. The excised specimen was examined histopathologically. Histological examination showed proliferation of small blood vessels lined by plump endothelial cells and surrounded by a lymphoid infiltrate with formation of germinal centres. Large numbers of eosinophils were seen [Figs 2 & 3). A diagnosis of ALHE was made based on the clinical and histopathological features.

Figure 1. Clinical photograph showing a 2cm × 5 cm solitary mass in the perianal region, lying superficial to the skin.

Figure 2. Histopathology showing proliferation of small blood vessels lined by plump endothelial cells and surrounded by a lymphoid infiltrate with formation of germinal centres.

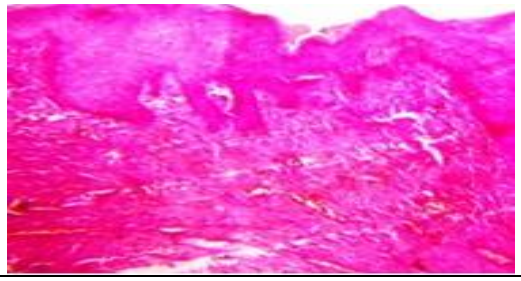
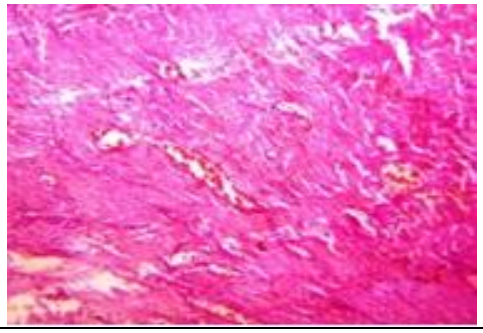


Figure 3. High power microscopic view showing blood vessels lined by cuboidal endothelial cells and numerous eosinophils in the background.



DISCUSSION

Angiolymphoid hyperplasia with eosinophilia (ALHE) Angiolymphoid hyperplasia with eosinophilia (ALHE) is a benign, locally proliferating lesion composed of vascular channels lined by endothelial cells with abundant pink cytoplasm and vesicular nuclei. It is an unusual cutaneous entity affecting arteries, of the head and neck. It was first described in 1969 by Wells and Whimster [1]. The formation of ALHE is quite unsolved and multiple postulations and prepositions have been put forward. The main debate is, if ALHE is an atopic hypersensitivity reaction or due to vascular neoplastic changes, the former receiving more support .The possibility that ALHE may follow a primary trauma or infection has also been reported [6].

Initially it was thought to be related to Kimura's disease but newer research shows that clinically the lesions tend to be more superficial and the vascular component are more prominent in AHLE, whereas in Kimura's disease lesions are deeper and there is higher incidence of peripheral blood eosinophilia, but the lymphoid features dominate and there is lack of the distinctive endothelial cells. Histologically the lesions are characterized by proliferation of small- to medium-sized blood vessels often showing a lobular architecture.

Many of these vascular channels are lined by greatly enlarged (epitheloid) endothelial cells, leading to the terms "histiocytoid" or, more recently "epitheloid" haemangioma [8]. A perivascular inflammatory cell infiltrate composed mainly of lymphocytes and eosinophils is present, and among these the eosinophils predominate. In subcutaneous lesions, the inflammatory cell infiltrate is usually more massive, with a central, poorly circumscribed nodule that replaces the fat.

Blood eosinophilia has been reported in up to 15 % of cases and is not required to make the diagnosis. Some patients with ALHE have also been found to have renal disease, urinalysis could be considered. Radiologic examinations such as MRI or angiography may be required to determine the extension of the lesions. Positive diagnosis is based upon histological findings.

Other differential diagnosis based on clinical and/or histopathologic findings, includes cutaneous lymphoid hyperplasia, pyogenic granuloma, hemangioma, soft tissue tumours, angiosarcoma, Kaposi sarcoma, bacillary angiomatosis, bartonellosis and metastatic carcinoma. Many therapeutic procedures have been used. The first line of management includes excision being primary, topical corticosteroids, intralesional

corticosteroids. The second line include topical tacrolimus ointment, systemic corticosteroids, cyclosporine, local radiation therapy and vinblastine, the latest being the usage of the Nd-YAG laser ablation [9].

Despite the basket of therapeutic modalities, frequent recurrences have been noted. Although our patient exhibited all of the fundamental and differentiating histopathological features of ALHE, the initial diagnosis of ALHE was difficult because of the atypical site and lobulated surface.

CONCLUSION

Angiolymphoid hyperplasia with eosinophilia is a condition with a challenging diagnosis and treatment. In spite of the benign nature of this disease, it causes a diagnostic and therapeutic confusion because of the aesthetic disfigurement, resistance and recurrence post treatment. ALHE has rarely been reported in the perianal region and to exceed 3 cm in diameter, but based on our case report; it should be a reasonable differential diagnosis when confronting dermal tumours of the perianal region.

.

REFERENCES

1. Botet MV and JL Sanchez. (1978). Angiolymphoid Hyper plasia with Eosinophilia: Report of a Case and a Review of the Literature. *Journal of Dermatologic Surgery and Oncology*, 4, 931-936.
2. Suzuki H, Hatamochi A, Horie M, Suzuki T, Yamazaki S. (2005). A case of angiolymphoid hyperplasia with eosinophilia (ALHE) of the upper lip. *J Dermatol*, 32(12),991–5
3. Park Y, Chung J, Cho CG. (2002). Angiolymphoid hyperplasia with eosinophilia of the tongue: report of a case and review of the literature. *Oral Oncol*, 38(1), 103–6.
4. Fernandes BF, Al-mujaini A, Petrogiannis-haliotis T, Al-kandari A, Arthurs B, Burnier Jr MN. (2007). Epitheloid hemangioma (angiolymphoid hyperplasia with eosinophilia) of the orbit: a case report. *J Med Case Reports*, 1, 30.
5. Huang M, Lloyd WC, O'Hara M. (2008). Angiolymphoid hyperplasia with eosinophilia: an unusual presentation in a child. *J Amer Assoc Ped Ophthalmol Strabismus*, 12(3), 302–4.
6. T. G. Olsen and E. B. Helwig, (1985), Angiolymphoid Hyper- plasia with Eosinophilia: A Clinicopathologic Study of 116 Patients, *J Amer Acad Dermatol*, 12, 781-796.
7. Rajendran R, Padmakumar SK, Kothawar S, Balaraman NM, (2005), Angiolymphoid hyperplasia with eosinophilia (ALHE). *J Oral Maxillofacial Pathol*, 9, 24–6.
8. Chong OWS, Thomas A and Goh CL. (2006). Kimura's Disease and Angiolymphoid Hyperplasia with Eosinophilia: Two Disease Entities in the Same Patient. Case Report and Review of the Literature. *Inter J Dermatol*, 45, 139-145.
9. Ramchandani PL, Sabesan T and Hussein K. (2005). Angiolymphoid hyperplasia with eosinophilia masquerading as kimura disease. *Brit J Oral Maxillofacial Surg*, 43, 249-252.