Angiolymphoid Hyperplasia with Eosinophilia (Pseudopyogenic Granuloma) Among Iraqi Patients

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ABSTRACT:

BACKGROUND:

Angiolymphoid hyperplasia with eosinophilia (ALHE) (pseudopyogenic granuloma) is a benign locally proliferating lesion composed of vascular channels with a surrounding infiltrate of lymphocytes and eosinophils.

OBJECTIVE:

ALHE is a rare disease but in Iraq it apparently seems to be more common than expected, for this reason the present work was arranged to evaluate the clinical and histopathological aspects of the disease.

METHODS:

This case descriptive study was conducted in the Department of Dermatology & Venereology- Baghdad Teaching Hospital during the period between June 1982- March 2005. Sixteen patients with this skin problem were included in this study. All demographic points related to the disease were obtained from each case through detailed history, close clinical and histopathological means.

RESULTS:

Sixteen patients with ALHE were evaluated. Their ages of patients ranged from 20-50 years with a mean ± SD of 35.73 ± 8.72 years, they were 14 females and two males. The duration of the disease ranged from 1-15 (6.4 ± 4.42) years.

The clinical picture consisted of multiple dull to pinkish red angiomatous papules and nodules affecting the head only mostly around the scalp and ears. The histopathological examination revealed a proliferation & ectasia of blood vessels with eosinophilic infiltrate of the dermis.

CONCLUSION:

This study revealed that angiolymphoid hyperplasia with eosinophilia (ALHE) in Iraq, is a disease predominantly of adult females only affecting the head, with no lymphadenopathy and typical histopathology without lymphoid follicle formation. This is the first report describing this disease in Iraq and seems to be more common than European countries.

KEYWORDS: angiolymphoid hyperplasia with eosinophilia (ALHE), pseudopyogenic granuloma, Iraqi patients.

INTRODUCTION:

Angiolymphoid hyperplasia with eosinophila (ALHE) also called pseudopyogenic granuloma is a chronic, uncommon and an apparently benign locally proliferating tumor composed of vascular channels with a surrounding infiltrate of lymphocytes and eosinophils, whose etiology and pathogenesis is under debate [1-8].

When angiolymphoid hyperplasia with eosinophilia was first described in Western Europe, similarities to Kimura's disease as reported in the Far East were noted[9,10]. Indeed, many authors thought that both conditions might be part of one disease spectrum [8,10]. However, more recently most authorities emphasize differences between the two entities (Table 1) [3-5, 9,12-20].

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Table- 1: A Comparison between Angiolymphoid Hyperplasia with Eosinophilia and Kimura's Disease.

	ALHE	Kimura's disease
Gender	Female Predominance	Almost exclusively males
Age	3 – 4th decade	All ages
Lesion characteristic	Small & superficial inflammatory red angiomatous nodules & plaques or grape like clusters, no regional lymphadenopathy	Large, translucent dermal and or subcutanous nodules & tumors may involve salivary or parotid glands with regional lymphadenopathy
Site	Head (mostly ear & scalp)	Head, neck, oral mucosa, breast, .. etc.
Symptom	Asymptomatic	Pain, itching, hearing defect.
Lymphoid follicle	Rarely present in old lesions	Prominent
Mast cell number	Commonly increase	Rarely increased
Eosinophils & IgE level	Rarely increased	Commonly increased
Vascularity	Abundant Angiomatoid proliferation with uncanalized masses of endothelial cells	Capillary proliferation, large thickened vessels, swollen canalized endothelial cells
Fibrosis	Absent or present only at lesional edges	Prominent
Recurrence	Common after excision	Common

• Compiled from [3-5, 8, 9,12-20].

Although ALHE is a rare disease, but in Iraq it apparently seems to be more common than expected when compared with other countries, for this reason the present work was carried out, to evaluate the clinical , histopathological aspects of the disease and to review of the literatures.

PATIENTS & METHODS:

This case descriptive study was done in the Department of Dermatology & Venereology-Baghdad Teaching Hospital during the period June 1982- March 2005. Sixteen patients with this skin problem were included in this study.

A detailed history was carried out regarding the following points: age, sex, age of onset, duration of disease, family history, history of medical or surgical interference, recurrence after treatment and any associated symptoms. While close clinical evaluation was done including: location, size, number, tenderness, bleed easily, regional or systemic lymphadenpathy and any associated signs.

Biopsies were performed from all patients for histopathological examination. Assessment of IgE and complete blood film were done to 10 patients with especial attention to eosinophils count.

RESULTS:

Fifteen patients with ALHE were evaluated. Their ages ranged from 20 – 50 years with a mean ± SD of 35.73 ± 8.72 years while the duration of the disease ranged from 1 – 15 (6.4 ± 4.42) years . The age of onset was ranged between 5-47 years with a mean ± SD of 29.33 ±10.89 years. Females (13) were mostly affected compared to two males with a female: male ratio 6.5: 1.

The scalp was the site of predilection in 10 (66.66%) patients, followed by ear and retrouricular area in 6 (40 %) patients while two patients had two sites. The nose was the site of involvement in one (6.66%) patient.

The lesions were mostly asymptomatic apart from 4 patients who presented with history of itchy, tender lesions that affected the scalp, while 5 patients complained from conductive deafness plus tenderness in the lesions that affected ear. Three cases had history of bleeding from their scalp lesions (Table-2). All lesions showed gradual increase in size with time and recurrence after excision occurred in 7 patients.

The size of each lesion ranged from 1 – 2 cm with tendency to enlarge over time. The lesion(s) were either single or multiple, dull to pinkish red angiomatous papules, nodules, and plaques or grape like clusters some of which peduniculated. There was no associated lymphadenopathy in all patients (Fig-1).

The histopathological examination (Fig.-2) revealed that the epidermis was normal apart from marked hyperplasia in some areas. Regarding the dermal changes, there was a band of chronic inflammatory cells in the upper dermis with a clear grenz zone from the epidermis.

The chronic inflammatory cells consisted of mainly lymphocytes and macrophage, with variable number of eosinophils.

In this band there were numerous capillary vessels which were lined by big swollen endothelial cells. In some capillaries, endothelial cells were markedly prominent into the lumen. The deep dermis was composed of collagen bands among them, there were large dilated vascular spaces lined by normal endothelial cells. The lumens were full of red blood cells. Also in some foci these dilated big vascular spaces were present in the upper dermis. No lymphoid follicles were observed in any sections.

Blood sampling from 10 patients showed a high eosinophil count in 7 (70%) cases.

Table -2: The frequency distribution of the cases of Angiolymphoid hyperplasia with eosinophila (ALHE) by their complaint.

	No.	%
Itching*	4	26.66
Pain*	7	46.66
Bleeding*	3	20
Hearing defect*	4	26.66
Total	16	100

* The figure is more than total because some cases had itching, pain, and hearing defect.

DISCUSSION:

There is a controversy whether Kimura's disease or angiolymphoid hyperplasia with eosinophlia are one entity or separate diseases [8,10]. Although there are many overlapping features between them, Kimura's disease is reported to be common in Japan while rare in other countries [2,5,9].

To the best of our knowledge this condition was firstly diagnosed in Iraq in 1982(Sharquie – Personal observation). Since then, all cases were collected & evaluated, clinically and histopathologicaly the present work in favor of ALHE rather than Kimura's disease for the following reasons: The disease was more commonly found in females rather than males, affecting commonly the middle age group, and the site of involvement was head (scalp, ear, nose).The majority of our patients were asymptomatic, and there was no associated lymphadenopathy and no lymphoid follicle formation in any patients. Also, the typical histopathological picture of angiolymphoid hyperplasia with eosinophlia (ALHE). Therefore our cases are similar to ALHE cases reported in Western European countries [3-5, 8, 9,12-20].

This disease unfortunately is unfamiliar among dermatologists and is often misdiagnosed and mistreated as a Kaposi's sarcoma or other related conditions. This is the first report describing this disease in Iraqi patients.

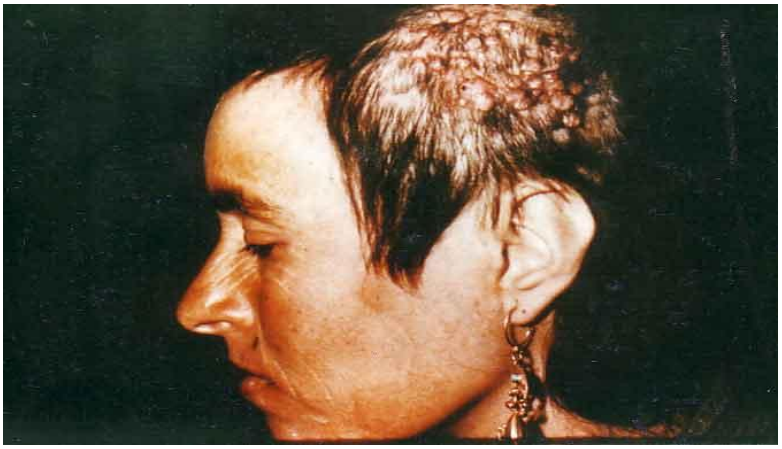

Fig-1A: Showing Angiolymphoid hyperplasia with eosinophila (ALHE)on the scalp of 30 years female.

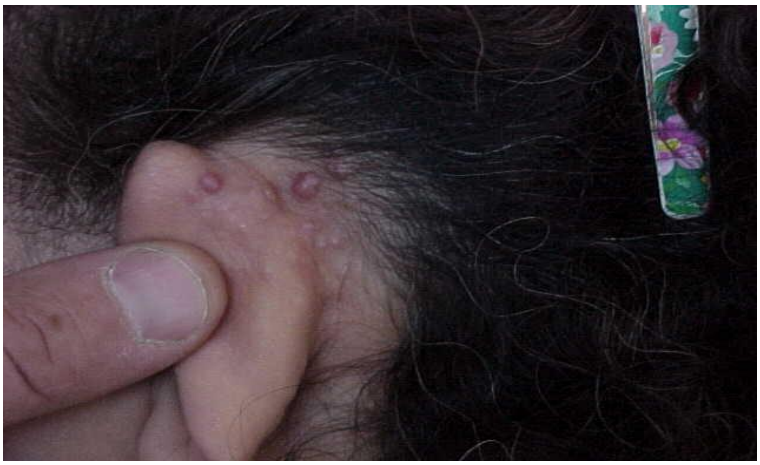

Fig-1B:Showing Angiolymphoid hyperplasia with eosinophila (ALHE)on the retrouricular of 27 years female.

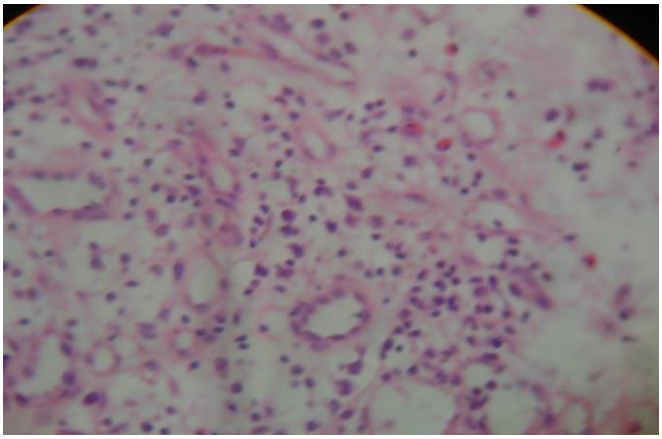

Fig.2:Light microscopic features of angiolymphoid hyperplasia with eosinophila (ALHE) showing multiple dilated blood vessels with perivascular inflammatory cells infiltration.(Hematoxylin-eosin stain; original magnification x 200).

REFERENCES:

1. Wells GC & Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. Br. J. Derm. 1969; 81, 1-15.
2. Mackie RM. Soft-Tissue Tumors: In Textbook of Dermatology. Champion RH; Burton JL; Burns DA; and Breathnach SM. 6th ed Oxford Blackwell Science Ltd. Ed. Offices 1998;3, 55: 2347-72
3. Chan JKC; Hui PK; Ng CS; Yuen NM; Kung IT& GWI E. Epithelioid haemangioma (angiolymphoic hyperplasia with eosinophilia) and Kimura's disease in Chinese. Histopathology 1989; 15 , 557-74.
4. Calonje E& Wilson- Jones E. Vascular Tumors and Tumor- Like Conditions of Blood Vessels and Lymphatics. Lever's Histopathology of the Skin. 8th ed. Elder D; Elenitsas R., Jaworsky C. and Johnson Jr.B. Lippincoott-Philadelphia. 1997; 34, 889-932.
5. Leiferman KM, Peters MS & Gleich GJ .Eosinophils in Cutaneous Diseases. Fitzpatrick's Dermatology In General Medicine, 6th ed. Freedberg IM, Eisen AZ, Wolffk K, Austen KF, Goldsmith LA & Katz SI, New York McGrow-Hill Companies, 2003,1,96:959-66.
6. Mariatos G; Gorgoulis VG; Laskaris G & Kittas C. Epithelioid hemangioma (aniolymphoid hyperplasia with eosinophia) in the oral Mucosa. A Case report and review of the literature. Oral– oncolo 1999; 35 , 435-8.
7. Requena L & Sangueza OP. Cutaneous Vascular Proliferation. Part II Hyperplasias and neoplasms. J. Am. Acad. Dermatol. 1997 ; 37, 887- 919.
8. Kung ITM; Gibson JB & Bannatyne PM. Kimura's disease a Clinico– pathological Study of 21 cases and its distinction from angiolymphioid hyperplasia with eosinophilia. Pathology 1984 ; 16 ,39-44.
9. Odom RB; James WD & Berger TG. Dermal and Subcutaneous Tumors, Andrew's Diseases of the skin: Clinical Dermatology; 9th ed. W.B Sanders-Philadelphia 2000,733-99.
10. Olsen TG & Heling EB. Angiolymphiod hyperplasia with eosinophplia. J. Am. Acad. Derma. 1985; 12, 781-96.
11. Chun S & Ji HG. Kimura's disease and angiolymphioid hyperplasia with eosinopholia: Clinical and histopathologic difference. J. Am. Acad. Derma. 1992; 27, 954.
12. Urabe A; Tsuneyoshi M & Enjoji, M. Epituelioid hemaryioma versus Kimura's disease. A comparative clinicopathologic study. Am. J. Surg. pathol. 1987 ; 11,758- 66.
13. Googe PB ;Harris NL &Mihm MCJR. Kimura's disease and angiolymphiod hyperplasia with eosinopholia Two distinct histopathological entities. J. Cutan. Pathology 1987; 14,263.
14. Kuo TT; Shih LY & Chan. Kimura's disease. Involvement of regional lymph nodes and distinction form angiolymphoid hyperplasia with eosinophplia. Am. J. Surg. Pathol. 1988; 12 , 843-54.
15. Ann HJ & Lee KG. A clinicopathological study of Kimura's disease and epitheliod hemangioma. Vonsei– Med. J. 1990; 31, 205-11.
16. Nair M; Aron M & Sharma MC. Angiolymphiod hyperplasia with eosinophilia (epthelioid hemangioma) of the breast: Report of a case. Surg. Today 2000; 30 ,747-9.
17. Delgandio JM; Myers MW & Teliun SA. Angiolymphiod hyperplasia with eosinophplia involving the external auditory canal. Otolaryngology Head Neck Surgery 1994; 111,669.
18. Allen PW; Ramakrishna B & MaCormae LB. The Histocytoid haemangionas and other controversies. Pathol. Am.1992; 27,51.
19. Tsang WYW & Chan JKC. The family of Epithoeliod Vascular Tumors. Histol. Histopathool. 1993; 8,187.
20. Kawada A. Kimura's disease Demonstration of the disease and its differential diagnosis. Hautarzt. 1976 ; 27 ,309-17.