CASE REPORT

Angiolymphoid hyperplasia with eosinophilia in a 27 year old male: A case report in Jos, North Central Nigeria

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Abstract

Background: Angiolypmphiod hyperplasia with eosinophilia (AHLE), also known as epithelioid hemangioma or histiocytoid hemangioma is a very rare vasoproliferative disorder with an obscure aetiopathogenesis. It mostly affects the head and neck regions of young women. It appears as a dark hyperpigmented or erythemathous nodular subcutaneous swelling depending on skin colour of the individual. Histologically it appears as proliferating small to medium sized blood vessels with epithelioid like endothelial lining cells within a lymphoid background punctuated by eosinophils. Even though it is mostly self limiting, surgery is one of the good modalities for its treatment.

Case presentation: A 27 year old male Nigerian presenting with dark hyperpigmented nodular growths on the right pinna and the left posterior auricular area. Had no history of ear piercing or injury prior to the onset of the growths. The masses measured about 4 by 2 by 1.5 cm and non tender. No regional lymphadenopathy felt on palpation or peripheral blood eosinophilia. Complete surgical excision of the two masses was done and specimen taken for histological evaluation. A diagnosis of ALHE was made. There was no recurrence of the growth two years after the surgery.

Conclusions: AHLE is very rare lesion and might be mistaken for other lesions that share gross or histological features with it. Histological diagnosis of the lesion is pertinent before definitive treatment. This will allow choice of preferred modality by the patient and the clinician. There are several treatment modalities for this lesion, but surgical excision is currently recommended.

Keywords: Angiolymphoid, Hyperplasia, Eosinophilia, Hemangioma, Surgery, Histomorphologic

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Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) was first described in 1969 by Wells and Whimster as a rare benign vasoproliferative disorder with an unknown aetiopathogenesis. The lesion is frequently confused with Kimura's disease, due to some salient similarities in both their gross and histological appearances.[1,2] ALHE is also known as epithelioid haemangioma or histiocytoid haemangioma and was considered to be a reactive phenomenon in the past, but now regarded as a true benign neoplasm. It commonly occurs in women between the ages of 20 to 40 years.[2,3,4] It has a predilection for the head and neck regions, most especially the face and the peri-auricular area. This lesion may also be seen occasionally on the trunk, the upper limbs and the genitalia.[5,6] It grossly appears as a dark hyperpigmented swelling or erythematous to violaceous papulonodular mass depending on the patient's skin colour. The modalities for treatment include intra-lesional corticorsteroids, laser therapy, radiotherapy, surgical excision of the lesion and among others. Surgical excision is the preferred choice with rare reported recurrence.[6]

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Case Report

A 27 year old Nigerian male presenting in the hospital with a slowly growing ovoid shaped nodular lesions on the right ear lobe and the left posterior auricular area of approximately 4 years duration. There was no history of ear piercing, injuries around the ear or the peri-auricular area. On examination, the lesions were hyperpigmented, soft to firm, painless and measured about 4x2x1.5 cm each. There was no regional lymphadenopathy. The peripheral blood film did not show eosinophilia.

Excision biopsies of the left peri-auricular and right ear lobe nodules were done 16 months apart. The biopsy specimens were sent for histopathological evaluation. Grossly the tumours appear as fairly nodular grayish-white masses measuring about 3.5cm in diameter that are partially covered by a negroid skin. Histologically they showed a keratinizing stratified squamous epithelium overlying a highly collagenized fibrous stroma within which were circumscribed intra-dermal masses composed of numerous proliferating blood vessels of various sizes. These blood vessels comprise of capillaries and medium sized arteries that were lined by plump epithelioid endothelial cells giving them hobnail or tombstone appearances. The background was formed by lymphocytic cell collections which formed lymphoid follicles with reactive germinal centres. These were punctuated by remarkable eosinophilic polymorpho-nuclear cell infiltrates. There were normal skin adnexal

structures. Based on these features, a histological diagnosis of Angiolymphoid hyperplasia with eosinophilia (ALHE) was made.

The patient had reconstruction of the right ear lobe and was followed up. There was no recurrence of the lesions at two year post surgery.

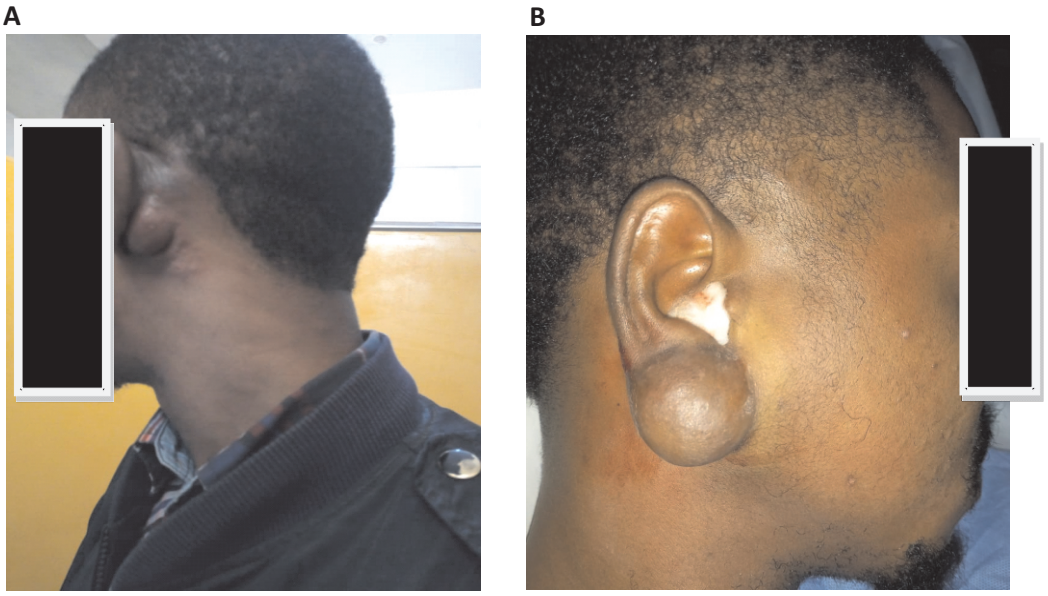

Figure 1. A, Show a nodular hyperpigmented left posterior auricular swelling while **B,** Show a hyperpigmented growth of the right pinna of the same case.

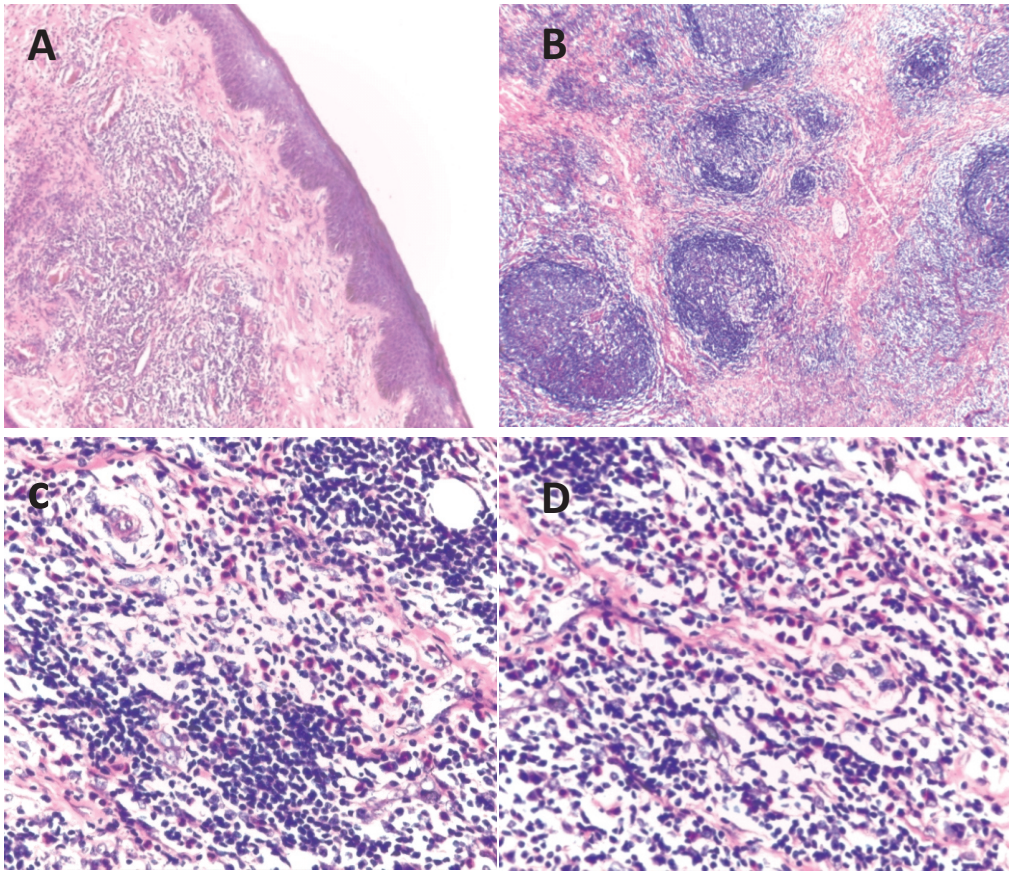

Figure 2. A, Shows dermal collections of lymphocytic cells and proliferating blood vessels (H&E, ×40). B, shows lymphoid follicles with reactive germinal centres (H&E, ×40). C and D,Show Presence of remarkable eosinophils within the lymphocytic collections and the presence proliferating blood vessels lined be epithelioid endothelial cells (H&E, ×100).

Discussion

ALHE is a rare benign slowly growing tumor with an unknown aetiopathogenesis. It was considered to be the same with Kimura disease as both lesions present as nodules and are located commonly on the head and neck regions.[8, 9] They are however now regarded as separate entities. Lesions in Kimura disease are not erythematous as opposed to that of ALHE which are erythematous in light skinned individuals.[10] They are deeply located, often associated with localized regional lymphadenopathy, peripheral blood eosinophilia, histologically more fibrotic stroma and less blood vessels with epithelioid looking endothelial cells.[4, 9] AHLE is commoner in females, but this case occurred in a 27 year old male. The age and the sites of the lesions follow the known pattern.

Histological evaluations of incisional biopsies taken from these lesions could have been done preferably before the commencement of definitive treatment. This is due to the cosmetic concerns associated with the lesions and their site of predilection. This will give opportunity for both the patient and the clinician to choose the therapy with the best treatment and aesthetic outcome and also acceptability to the patient. In this case complete excision of the lesions was done before histological diagnosis.

Majority of ALHE cases regress spontaneously after a variable period of time ranging from 3-6 months.[10] Progressive lesions may require complete surgical excision, which is the treatment of choice even in the presence of less invasive therapeutic modalities. This is due to higher rate of recurrence after treatment with the other modalities like Intra-lesional corticosteroids, laser and radiotherapy.[6, 10] Surgial intervention alone was curative in this index case. There was no recurrence after two years and the patient was comfortable with the cosmetic outcome.

Conclusion

Globally, AHLE is a very rare lesion and this is the first reported case in our centre. A high index of clinical suspicion and a subsequent histological evaluation is necessary before definitive treatment. Surgery remains a reliable treatment option for this lesion, not only in other parts of the world but also in our domain. Every treatment modality must be by a patient's consent no matter the prognosis of the condition

References

1. Wells GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. Br J Dermatol. 1969; 81:1–5.
2. Kede MPV, Alves MFGS, Ramos-e-Silva M. Hiperplasiaangiolinfóide com eosinofilia. An Bras Dermatol. 1993; 68:263–265.
3. Chan JKC, Hui PK, Ng CS, Yuen NWF, Rung ITM, Gwi E. Epithelioid haemangioma (angiolymphoid hyperplasia with eosinophilia) and Kimura's disease in Chinese. Histopathology. 1989; 15:557–574.
4. Fletcher CDM. Selected summaries: soft tissue tumours. Journal of Pathology. 1989; 159:341–344.
5. Youssef, A., Hasan, A.R., Youssef, Y, Al-soufi L, Elshimali Y, Alshehabi Z. Angiolymphoid hyperplasia with eosinophilia: a case report. J Med Case Reports. 2018; 12:89.
6. Nogueira A, Filho JWA, de Castro MCR, Maceira JP, Ramos-e-Silva M. Hiperplasiaangiolinfóide com eosinofilia – relato de doiscasos. An Bras Dermatol. 2003; 78:79–85.
7. Zaraa I, Mlika M, Chouk S, Chelly I, Mokni M, Zitouna M et al. Angiolymphoid hyperplasia with eosinophilia: a study of 7 cases. Dermatol Online J. 2011; 17:1.
8. Mehregan AH, Shapiro L. Angiolymphoid hyperplasia with eosinophilia. Arch Dermatol. 1971; 103:50–57.
9. Demitsu T, Nagato H, Inoue T. Angiolymphoid hyperplasia with eosinophilia: its character and therapy. Skin Surg. 2000; 9:8–16.
10. Kennedy CTC. Angiolymphoid hyperplasia with eosinophilia. In: Champion RH, Burton JL, Burns DA, Breathnach SM, editors. Rook/Wilkinson/ Ebling Textbook of dermatology. 6th ed. Blackwell Science Ltd.; London: 1998:3027–3028.