

Case Report: Angiolymphoid Hyperplasia with Eosinophilia of the Nose

Van Ratingen AR*, MMD van der Linden and Sillevius Smitt JH

Academic Medical Center, Department of Dermatology, University of Amsterdam, Amsterdam, Netherlands

Abstract

We present a case of a 62-year-old woman with a 3-year history of a red indurated plaque on the right ala of the nose, which did not respond to therapy for rosacea. A skin biopsy showed a conglomerate of thick walled vessels with a wide variation of the vessel wall diameter and a cobblestone appearance. Furthermore there were some small infiltrates with eosinophilic granulocytes. The diagnosis of ALHE was made and the patient was referred to a plastic surgeon for excision of the tumour.

ALHE is a rare entity mostly found around the ears. ALHE on the nose is even more rare and could be misdiagnosed as phymatous rosacea. Misdiagnosis can lead to unnecessary treatment with antibiotics and delay of appropriate therapy.

Keywords: Rosacea; Tumour; Antibiotics

Report of a Case

A 62-year-old woman was referred to our dermatology department because of a 3-year history of a red indurated plaque on the right ala of the nose. According to the patient the lesion initially started as a little erosion that would not heal, since then it was swollen, pulsating and painful. There was no trauma in advance, specifically no piercing. The colour of the lesion fluctuated during the day from erythematous to intense red. A former skin biopsy suggested the diagnosis phymatous rosacea. Based on this histology treatment was started with doxycycline 100 mg a day for 3 months and metronidazol 1% and azelaic acid 20% topically. This treatment did not lead to resolution of the lesion, although she experienced less pain, swelling and redness.

The patient otherwise felt well and did not have any other complaints, especially no complaints of her eyes and no facial papules or pustules. Her medical history showed a nephrectomy of the left kidney because of sclerosis. She did not use any other medication besides the ones mentioned above.

On examination we saw a swollen, indurated and erythematous right ala of the nose (Figures 1 and 2). There was no pulsation palpable. The dimensions of the lesion were 1.5 × 1.5 cm. The remainder of the facial skin was normal. angiolymphoid hyperplasia with eosinophilia (ALHE) was suspected.

Histopathological analysis of a new skin biopsy revealed a conglomerate of thick walled vessels with a wide variation of the vessel wall diameter and an increase of micro vessels. A few of these vessels showed a cobblestone appearance. Furthermore there were some small infiltrates with eosinophilic granulocytes. All of these findings together are compatible with the diagnosis ALHE (Figure 3).



Figure 1: Swollen, indurated and erythematous right ala of the nose.



Figure 2: Detail of the lesion.

Magnetic Resonance Imaging was performed to determine the depth of the tumour and a possible afferent arteriole. This revealed a soft tissue swelling on both sides of the nasal bone of the right ala without extension in the depth or the cavum nasi. There was no afferent arteriole identifiable.

With the diagnosis ALHE the patient was referred to a plastic surgeon for excision of the tumour. The result of plastic surgery was excellent and patient's satisfaction was high (Figure 4). Finally she was told that unfortunately, recurrence would be possible.

Discussion

ALHE was first described by Wells and Whimster [1] who defined it as a late stage of Kimura disease. Since then there has been a lot of controversy about these two conditions. Nowadays it is widely accepted to be different entities.

ALHE is a rare, benign, vasoproliferative disease of unknown

***Corresponding author:** Anne R van Ratingen, Academic Medical Center, Department of Dermatology, University of Amsterdam, Meibergdreef 9, 1105 AZ Amsterdam, Netherlands, Tel: 0031 20 5662587; E-mail: a.r.vanratingen@amc.uva.nl

Received: October 27, 2015; **Accepted:** January 13, 2016; **Published:** January 15, 2016

Citation: Van Ratingen AR, Linden V, Sillevius Smitt JH (2016) Case Report: Angiolymphoid Hyperplasia with Eosinophilia of the Nose. Dermatol Case Rep 1: 105.

Copyright: © 2016 Van Ratingen AR, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

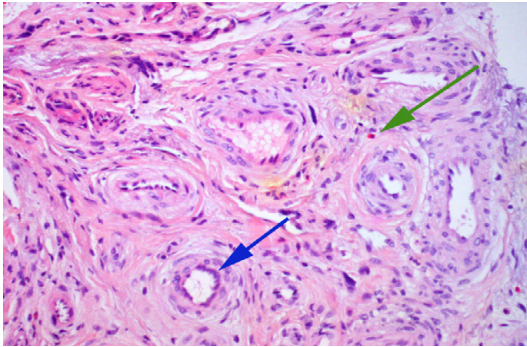


Figure 3: Histology: HE stains 200x. A conglomerate of thick walled vessels with a variation of the vessel wall diameter and a cobblestone appearance (blue arrow). Furthermore few small infiltrates with eosinophilic granulocytes (green arrow).



Figure 4: The result after plastic surgery.

origin. It is characterized by intradermal or subcutaneous single or multiple reddish-brown papules or nodules, mainly on the head and neck, especially around the ears [2,3]. Less often lesions of ALHE have been described in the mouth, on the trunk, extremities, vulva and penis.

Patients, mostly young to middle-aged females of any race, may complain about itching, tenderness or pulsation of the lesions. They do not have lymphadenopathy or peripheral eosinophilia in comparison with Kimura disease [2,3,4].

Histopathologically ALHE is characterized by prominent vascular hyperplasia. There are numerous irregular thick-walled blood vessels lined up with "epithelioid" endothelial cells. These cells have large nuclei and abundant eosinophilic cytoplasm. Sometimes cytoplasmic vacuoles are present which protrude into the lumina resulting in a "cobblestone" appearance.

The inflammatory infiltrate consists of lymphocytes, eosinophils, mast cells, and plasma cells. In some cases lymphoid follicles with germinal centers are present [2,3,5].

The pathogenetic mechanism of ALHE is unknown. In some cases

there is a preceding trauma, a recently published article described ALHE after an ear piercing [6]. Some reporters suggest it is related to infection and high estrogen status. Nevertheless, there seems to be consensus that ALHE is a reactive rather than a neoplastic process [3,5,7].

The treatment of ALHE is challenging because of the high recurrency rate, however surgical excision remains the treatment of choice. Many other therapeutic options for ALHE have been discussed in the literature including radiotherapy, indomethacin farnesil, laser therapy (CO₂, argon, copper vapour, pulsed dye, Nd: YAG), photodynamic therapy, corticosteroids, cryotherapy, electrodesiccation, pentoxifylline, intralesional chemotherapeutic agents, Interferon- α , thalidomide, oral isotretinoin and acitretin. Unfortunately ALHE tends to recur. Because it is a benign disease and malignant change never has been reported so far, interventions should not be too aggressive [5,7].

Our patient presented with a single lesion on her nose, a location rarely described in the literature [8,9]. Because of the exceptional location of the lesion it will be more difficult to recognize. Concerning the differential diagnosis it is easily mistaken for phymatous rosacea; a type of rosacea characterized by a swollen nose with sebaceous gland hyperplasia and fibrosis. Other differential diagnoses could be granuloma faciale, angioma, pyogenic granuloma, angiosarcoma, Kaposi sarcoma and Kimura disease.

It is important to take a representative skin biopsy to discriminate between the different diagnoses. Misdiagnosis can lead to unnecessary treatment with antibiotics (in our case) and delay of appropriate therapy.

References

1. Wells GC, Whimster IW (1969) Subcutaneous angiolymphoid hyperplasia with eosinophilia. *Br J Dermatol* 81: 1-14.
2. Olsen TG, Helwig EB (1985) Angiolymphoid hyperplasia with eosinophilia. A clinicopathologic study of 116 patients. *J Am Acad Dermatol* 12: 781-796.
3. Requena L, Sangueza OP (1997) Cutaneous vascular proliferations. Part II. Hyperplasias and benign neoplasms. *J Am Acad Dermatol* 37: 887-919.
4. Soo IC, Hye GJ (1992) Kimura's disease and angiolymphoid hyperplasia with eosinophilia: clinical and histopathologic differences. *J Am Acad Dermatol* 27: 954-958.
5. Chong WS, Thomas A, 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. *Int J Dermatol* 45: 139-145.
6. Okman JS, Bhatti TR, Jackson OA, Rubin AI (2014) Angiolymphoid hyperplasia with eosinophilia: a previously unreported complication of ear piercing. *Pediatr Dermatol* 31: 738-741.
7. San Nicoló M, Mayr D, Berghaus A (2013) Angiolymphoid hyperplasia with eosinophilia of the external ear: case report and review of the literature. *Eur Arch Otorhinolaryngol* 270: 2775-2777.
8. Chiu SC (2013) An unusual case of angiolymphoid hyperplasia with eosinophilia of the nose. *Ear Nose Throat J* 92: E10-11.
9. Panduranga Kamath M, Bhojwani KM, Bhandarkar AM, Pai RR, Rent NH (2014) Angiolymphoid hyperplasia with eosinophilia of root of nose: a rare phenomenon. *J Clin Diagn Res* 8: 144-145.