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Case Report

Spontaneous regression of angiolymphoid hyperplasia with eosinophilia (AHLE): A case report

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ABSTRACT

Introduction: and importance - Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare benign microvascular proliferation. It is clinically characterized by intradermal or subcutaneous papules and/or nodules. Treatment consists mainly of surgery or laser therapy, with a high recurrence rate. Spontaneous regression is rare but possible.

Case presentation: A 72-year-old man presented for an erythematous plaque on his scalp. Physical examination demonstrated a large angiomatous indurated subcutaneous plaque. The lesion was not pulsatile. The doppler ultrasound revealed no underlying vascular malformation. The biopsy revealed an ALHE. No further treatment was carried out. During the follow-up, the lesion gradually subsided. After one-year follow-up, the lesion had regressed with no sign of recurrence.

Clinical discussion: There are different treatment modalities for AHLE management, including surgical excision, cryotherapy and lasers. However, none of these modalities has provided consistent results and recurrence after treatment is common.). Rare instances of spontaneous regression have been reported.

Conclusion: Given the rarity of cases of spontaneous resolution, ALHE may be considered as a chronic condition, especially since recurrences are frequent. A "wait and see" approach can be proposed in some specific situations.

1. Background

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare benign microvascular proliferation tumor of unknown etiology tumor [1]. It presents as red to brown papules or nodules, located in the head and neck region, especially around the ears and on the scalp. Microscopically, ALHE consists of plumped epithelioid or histiocytoid endothelial cells and accompanying eosinophils and lymphocytes. Treatment consists mainly of surgery or laser therapy. Recurrence after treatment is common [2]. Spontaneous regression is rare but possible. We report a case of spontaneous regression of ALHE after two-years follow-up. The work has been reported in line with the SCARE 2020 criteria [3].

2. Case report

A 72-year-old man, with no significant past medical history, was referred to our department for evaluation of a rapidly enlarging, mildly itchy erythematous plaque on his scalp. The lesion had first been noticed 6 months ago. There was no known traumatic event, but bleeding after minor frictional trauma was reported by the patient. Physical examination demonstrated a large angiomatous indurated subcutaneous plaque, located on the occipital region of the scalp (Fig. 1). The lesion was not pulsatile. There was no regional lymph node enlargement. Laboratory findings revealed no peripheral eosinophilia. Prior to biopsy, our differential diagnosis were ALHE, Kimura disease, lymphoma cutis, angiosarcoma, metastatic tumor and sarcoidosis. The doppler ultrasound revealed no underlying vascular malformation. The biopsy revealed vascular hyperplasia with epithelioid endothelial cells, as well as lymphocytic and eosinophilic infiltrate with a few plasma cells and rare giant cells. No atypical mitotic figures were observed (Figs. 2 and 3). The diagnosis of ALHE was confirmed by clinical presentation and histology. No further treatment was carried out (either local or systemic corticosteroid administration, or excision) because the patient lacked any disabling symptoms and he did not prefer such treatment. During

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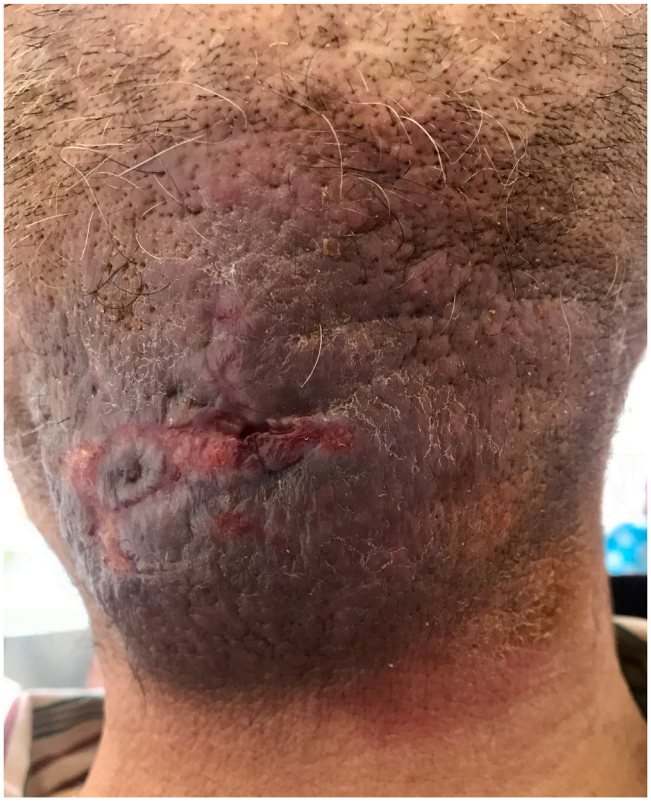

Fig. 1. Angiomatous subcutaneous plaque on the scalp (AHLE).

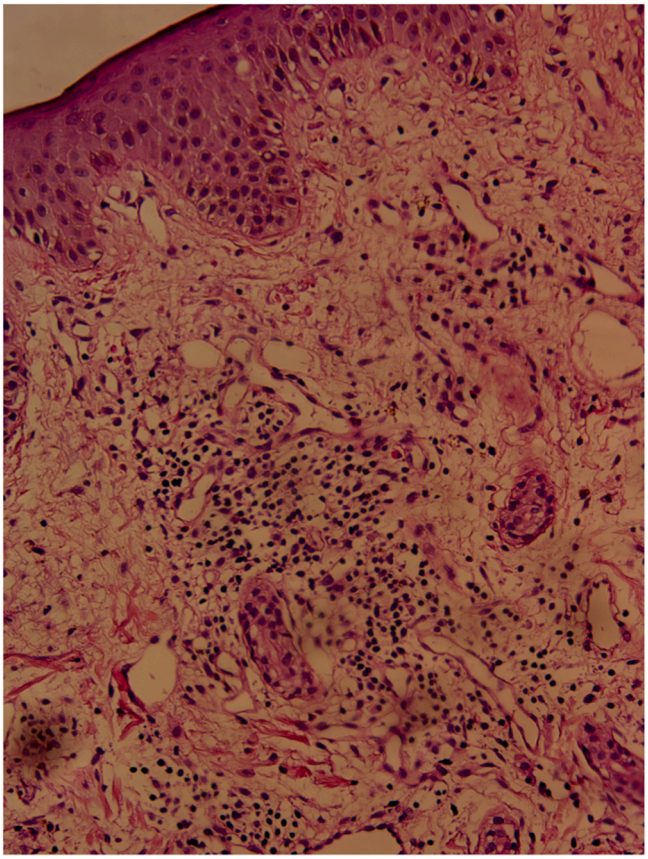

Fig. 2. Hematein-eosin tissue section (x20).

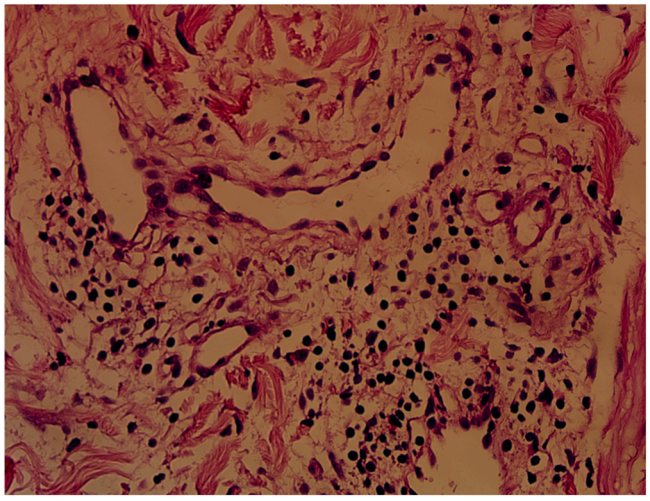

Fig. 3. Hematein-eosin tissue section (x40).

the follow-up, the lesion gradually subsided. After one-year follow-up, the lesion had regressed with no sign of recurrence (Fig. 4).

3. Discussion

Both the cell of origin and the pathogenesis of ALHE remain controversial. The World Health Organization (WHO) and the International Society for the Study of Vascular Anomalies (ISSVA) classify ALHE as a benign vascular tumor [4]. However, this lesion has several features that imply a significant reactive, as opposed to neoplastic, component.

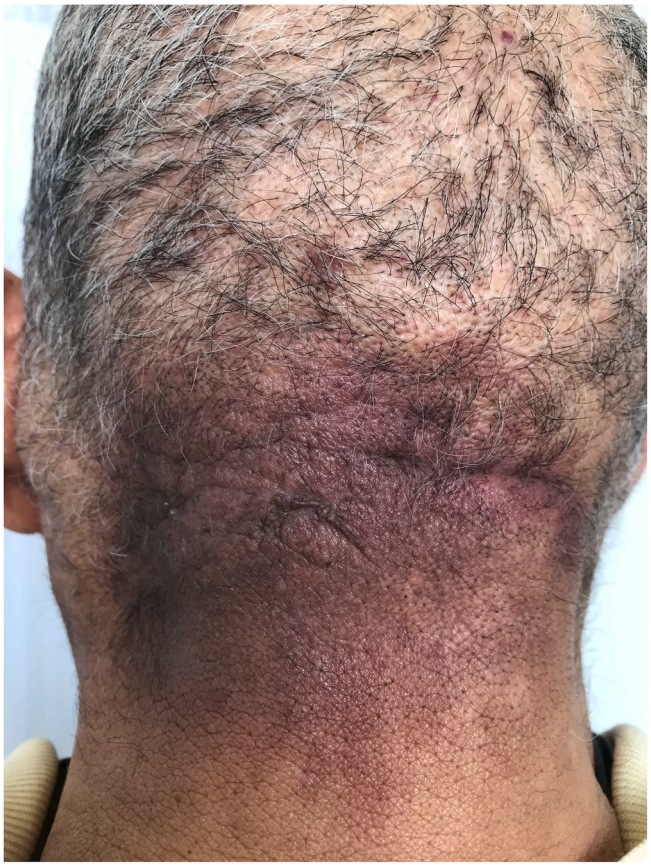

Fig. 4. Spontaneous regression of the lesion without recurrence after one-year follow-up.

Furthermore, ALHE or a subset of ALHE cases harboring a clonal T-cell population have been reported and may represent a T-cell lymphoproliferative disorder of a benign or low-grade malignant nature [5]. ALHE occurs in young to middle-aged adults without gender or ethnic predilection. Previous reports of a female predominance are not supported by more recent series, including our case report [6].

Past reports emphasized pruritus as the most common symptom associated with ALHE, as in our patient. Pain is as an uncommon symptom. Spontaneous bleeding over the lesion in our patient is consistent with the vascular nature of the lesion (7).

The clinical lesions appear as single or multiple red-brown papules or as subcutaneous nodules with a predilection for the head and neck region. ALHE is characterized histologically by proliferations of capillary-sized vessels with epithelioid endothelial cells surrounding larger thick-walled vessels, accompanied by eosinophils and lymphocytes. The differential diagnosis of ALHE includes Kimura's disease, angiomatous lymphoid hamartoma, hemangioma, pyogenic granuloma, Kaposi sarcoma, lymphoma, and epithelioid hemangioendothelioma Unlike Kimura's disease, peripheral eosinophilia is inconstant [8]. It was absent in our patient. There are different treatment modalities including surgical excision, cryotherapy, chemotherapy, intralesional and systemic corticosteroids, curettage, electrodesiccation, radiation, and lasers. However, none of these modalities has provided consistent results and recurrence after treatment is common [9].

According to the literature, cases of ALHE with earlier age of onset, longer duration, and multiple lesions are linked to higher recurrence rates [6]. Rare instances of spontaneous regression have been reported (Table 1). On 1969, Wells and Whimster published the first report describing 9 cases of ALHE characterized by marked vascular proliferation with infiltration of eosinophils. One of them, a 23-year-old man, had a nodule for 3 months in the left nasolacrymal region that was excised. One year later, a nodule recurrence was found but no further treatment was carried out. Follow up after 4 years showed that there was no longer any nodle at the original site and no fresh lesion [8]. In a study of 116 cases of ALHE, Olsen et al. reported spontaneous resolution without any form of therapy in 2 patients [7]. Fays et al. described the case of a 32-year-old male patient having five angiomatous lesions of the face consistent with ALHE, that showed spontaneous regression with no recurrence after three years follow-up [10]. Given the rarity of cases of spontaneous resolution, ALHE may be considered as a chronic condition, especially since recurrences are frequent. Together with previously published reports, the case presented here suggests that in some specific situations, a "wait and see" approach with close monitoring can be proposed to patients suffering from ALHE as a first step.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Conflicts of interest

None.

Table 1
Review of literature of spontaneously regressive AHLE cases.

Author	Number of patients	Age	Findings	Follow-up
Wells et al. [8]	1	23	Recurrence of an AHLE of the left nasolacrymal region that was excised the year before	4 years
Olsen et al. [7]	2	Not precised	Not precised	Not precised
Fays et al. [10]	1	32	Five angiomatous lesions of the face	3 years

I have no financial and personal relationship with other people or organisations that could inappropriately influence my work.

Sources of funding

None.
I have no source of funding.

Ethical approval

Ethical Approval has been given by the ethics committee of our faculty.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

I, Yasmine Slimani, corresponding author of the manuscript, accept full responsibility for the work and the conduct of the study, had access to the data, and controlled the decision to publish.

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The following information is required for submission. Please note that failure to respond to these questions/statements will mean your submission will be returned. If you have nothing to declare in any of these categories then this should be stated.

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Authors must obtain written and signed consent to publish a case report from the patient (or, where applicable, the patient's guardian or next of kin) prior to submission. We ask Authors to confirm as part of the submission process that such consent has been obtained, and the manuscript must include a statement to this effect in a consent section at the end of the manuscript, as follows: "Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request".

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Author contribution

Please specify the contribution of each author to the paper, e.g. study concept or design, data collection, data analysis or interpretation, writing the paper, others, who have contributed in other ways should be listed as contributors.

- Drafting of the manuscript: Yasmine Slimani
- Examination of the biopsy: Cheikh Sid'Ahmed Tolba, Farida Marnissi
- Critical revision of the manuscript for important intellectual content: Fouzia Hali, Soumiya Chiheb

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The Guarantor is the one or more people who accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

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Declaration of competing interest

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102376.

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