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# Angiolymphoid Hyperplasia with Eosinophilia in External Auditory Conduct, New Therapeutic Option

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## Abstract

It is an uncommon angioproliferative disorder, which clinical and histopathologic features are characteristic. It shows a young female patient with lesions on the left ear. She received a successful treatment with Imiquimod. Currently, there are many treatments for such rare disorder, although there is still a high rate of relapses.

**Keywords:** Angiolymphoid Hyperplasia with Eosinophilia; Imiquimod; External Auditory Conduct

**Abbreviations:** EAC: External Auditory Conduct; ALHE: Angiolymphoid Hyperplasia with Eosinophilia; TLRs: Toll receptors.

## Introduction

Angiolymphoid hyperplasia with eosinophilia is a dermatitis that rarely appears and represents a therapeutic challenge for all specialties. A great number of treatments were described; glucocorticoids (topic, intraregional or systemic), intraregional chemotherapy and ablatives techniques; surgery, electrocoagulation, among others.

## Objective

Is to report a case of ALHE in the External Auditory Conduct, successfully treated with Imiquimod 5% with minimal side effects and without reappearance of the lesions during the follow up.

## Materials and Methods

We report a case of a 41 years-old female patient, who had a 4 months history of pruritic lesions on the left ear. There were no previous records of traumatism, oral contra conception, atopy or recent pregnancy.

**Physical Examination:** We observed multiple dome shaped tumors, reddish-violaceous, smooth surface, hard elastic consistency and angiomatosous aspect, without fremitus, localized in the left auricular concha, in the tragus and in the helix root, which compromised the distal portion of External Auditory Conduct (EAC) (Figure 1).

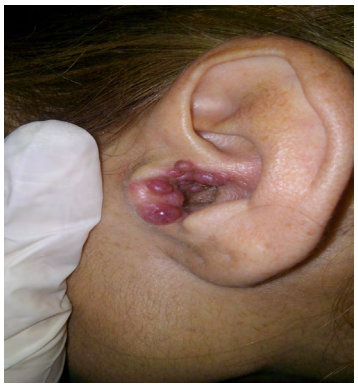

**Figure 1:** Multiple dome shaped tumors of angiomatosous aspect localized in the left auricular concha.

Skin biopsy was performed with the following findings: Vascular proliferation and endothelial hyperplasia and the presence of abundant eosinophil's (Figures 2 & 3).

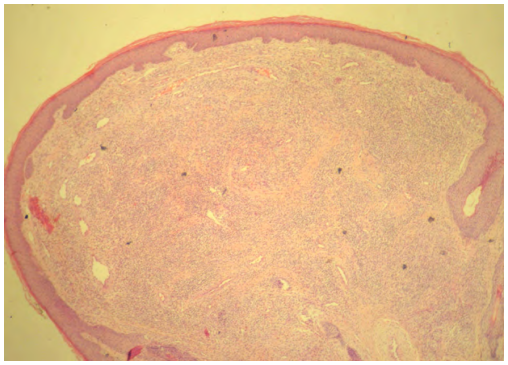

**Figure 2:** Vascular proliferation and endothelial hyperplasia and abundant presence of eosinophil (Eosin-hematoxylin x 40).

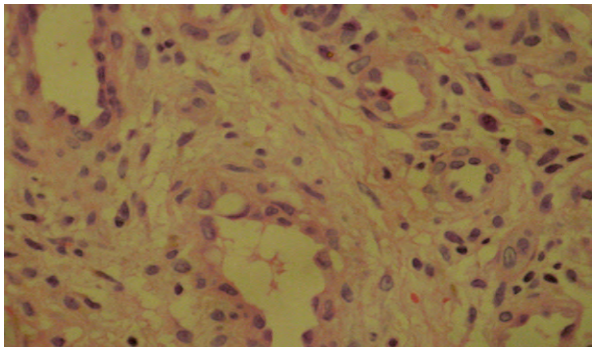

**Figure 3:** In better detail, the abundant presence of eosinophils is emphasized. (Eosin- Hematoxylin x 100).

**Treatment:** Imiquimod 5% cream applied five days a week for 5 months with high tolerance and vanishing tumors in the preauricular region and tragus (Figure 4).

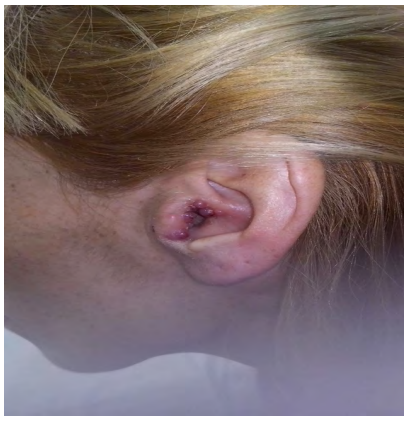

**Figure 4:** Improvement of the lesions after 45 days with Imiquimod 5%.

The application was limited to the lesions outside the concha due to the potencial risk of entrance in the EAC, which could cause inflammation and infection. Otomicroscopic control was kept during the treatment, local cleaning with aspiration, occasionally. During the first three weeks it was required the application of fluidic acid combined with betamethasone cream, to minimize irritation.

## Results

After 5 months of treatment, it was observed the absence of preauricular lesions in the helix root and in the tragus but the tumors persisted in the distal portion of the EAC (Figure 5). We followed up the patient for 24 months without showing new lesions.

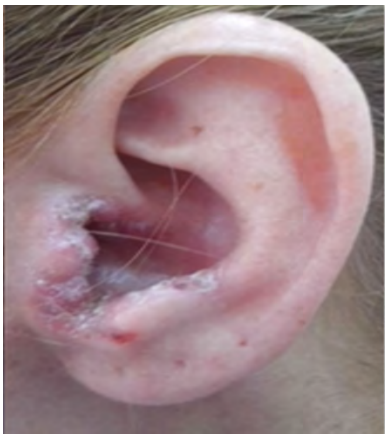

**Figure 5:** Complete resolution over the next 5 months of constant treatment.

## Discussion

Angiolymphoid Hyperplasia with Eosinophilia (ALHE) belongs to the group of eosinophilic dermatosis [1]. It was described by Wells and Whimster in 1969, they thought it was a late status of "Abnormal granuloma with lymphoid tissue proliferation" as described by Kimura and Cols in 1948 [2]. The etiology suggests that is a reactive process caused by different stimuli, which could be toxic, allergic, insect bites, infections or hormonal influence (oral contraception or pregnancy).

It is believed that the vascular proliferation would be the primary event and the inflammation would be the consequence of said proliferation. As well, it was postulated that is a lymphoproliferative disorder CD4 related to a predominant reactive angiogenic answer. The most accepted hypothesis is the presence of persistent shunts arteriovenous [3-4].

Different treatmens have been proposed: glucocorticoids (topics, systemic or intralesional) [2], propanolol, indomethacin, farnesyl, pentoxifylline, intralesional chemotherapy (bleomycin, vinblastine, fluorouracil), isotretinoin, acitretin, tacrolimus [5], interferon alpha-2a and mepolizumab [6], sclerotherapy [7]. Most of them had been reported with variable rate of response between 20 to 50%, the ablatives are the treatment of choice in anatomic areas outside the auricular zone and they include: surgery, electrodessication, CO2 laser [1], argon laser, ultra pulsed dye laser [9], electrocoagulation, radio frequency, and cryosurgery [10]. In the third part of the treated cases, particularly after the incomplete removal, have been reported recurrences [11].

Imiquimod is an inmunomodulator that acts mainly in Toll receptors (TLRs) which are in the surface of antigen presenting cells. It has antiangiogenic properties inhibiting pathological development of new vessels through the production of NFI. The increasing levels of IL 10 and 12, which decreases the cellular production of proangiogenic factors, such as fibroblast growth factor, IL 8 and the plasminogen activator urokinase [12].

Imiquimod reduces the cellular proliferation increasing apoptosis in tumoral tissue. The expression in the tissue of the metalloproteinase-1, angiogenesis inhibitor, is increased and that event diminishes the metalloproteinase-9 activity [13].

Redondo et al reported the first case of ALHE that obtained complete clinical resolution with Imiquimod cream 5% applied 5 times per week during 16 weeks without recurrences in the following 4 months [13]. In our case in the absence of recurrences during the follow up for 24 months Gencoglan, et al. detailed a case in which Imiquimod was applied twice a day for 5 days a week, during 2 weeks with an excellent evolution after 2 years of follow up [14]. A similar experience happened to our patient.

Isohisa, et al. reported another patient with multiple lesions successfully treated with Imiquimod 5% cream (5 times per week) with a rapid improvement of the itching and involuted in 22 weeks of therapy (the little ones) and in 47 weeks (the bigger ones) no adverse effects were reported [15]. In our patient the bigger lesions involutes after 20 weeks with the same modality of treatment.

In our experience, the use of Imiquimod 5% topic has the advantage of resolving short term lesions without developing subsequent recurrence in a period of 2 years of follow up with acceptable cosmetic results.

## Conclusion

Imiquimod could be an effective therapeutic alternative for ALHE and other cutaneous vascular tumors. However, this result must be confirmed with larger number of cases to support this evidence [11].

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