Case Report
Iranian Journal of Otorhinolaryngology, Vol.38(5), Serial No.148, Sep-2026

Angiolymphoid Hyperplasia with Eosinophilia in the Auricle and External Ear: A Case Report

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Abstract

Introduction:

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare, benign condition of unknown cause. It presents with red, purple, or brown bumps, patches, or lumps that develop in the skin or under the skin .It often appears in the head and neck area and near the ears.

Case Report:

A 42-year-old woman presented with multiple red lesions in the area around the ear .The patient had a history of treatment with steroid cream and steroid injections into the lesion, but there was no improvement. The initial biopsy was folliculitis, and the final pathology report reported ALHE.

Conclusion:

Surgical treatment resulted in improvement. ALHE should be considered as a potential diagnosis when a patient presents with isolated, well-defined bumps that look like blood vessels and may have a broken surface (ulcer) or crust. Despite ongoing research, questions regarding the etiology and pathogenesis of ALHE remain unanswered, and it requires further studies.

Keywords: Angiolymphoid hyperplasia with eosinophilia, External auditory canal, Auricle.

Received date: 29 Apr 2026
Accepted date: 29 Jun 2026

Please cite this article; *Jalali MM, Akbari M, Ammar A. Angiolymphoid Hyperplasia with Eosinophilia in the Auricle and External ear: A Case Report. Iran J Otorhinolaryngol. 2026:38(5): 347-351. Doi: 10.22038/ijorl.2026.95304.4143*

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Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare and benign disease of unknown etiology. This disease appears as red, purple, or brown lesions, spots, or lumps on the skin or under the skin. These lesions can be single or grouped, and sometimes they may even itch.

About 85% of the time, they appear on the head and neck, especially near the ears. The cause of ALHE is not well known, but it is stated that these lesions occur in response to various stimuli, such as abnormal growth of blood vessels, injury, or pregnancy (1).

When the damaged tissue is examined under a microscope (histology), a well-defined area with abundant new blood vessels is seen. These vessels have large, swollen endothelial cells with a large amount of eosinophilic cytoplasm (the inner part of the cell). In addition, there is a mixture of white blood cells, including lymphocytes and eosinophils around these vessels, and the amount of these cells can be differ. Recent research has reported cases of ALHE in unusual locations such as the tongue, penis (2,3), eyelids, or even the eye socket.

Some studies have reported successful treatments used in individual cases, including pulsed dye laser therapy, intralesional bleomycin injection, imiquimod cream, and interferon alfa-2b (4).

Interestingly, some recent studies suggest that ALHE may in some cases be caused by abnormal cell growth (clonality) (5,6). This hypothesis challenges the previous description that ALHE is purely a reactive condition. Studies have reported a 33% recurrence rate with surgery (7). We reported a case in which there was no recurrence after 15 months of surgery despite multiple lesions.

Case Report

A 42-year-old woman presented with about six red lesions of 0.5 to 1 cm in diameter in the area around the ear and ear canal two years ago. One lesion started at the beginning of the ear canal and extended approximately 3 mm into the canal.

Also there were two lesions on the root of the helix, one lesion on the tragus, and two lesions on the concha. She also noted a yellow discharge from her left ear. These lesions were not sensitive to touch and were firmly attached to the underlying tissue. They had a smooth surface and felt solid to the touch (Figure 1).

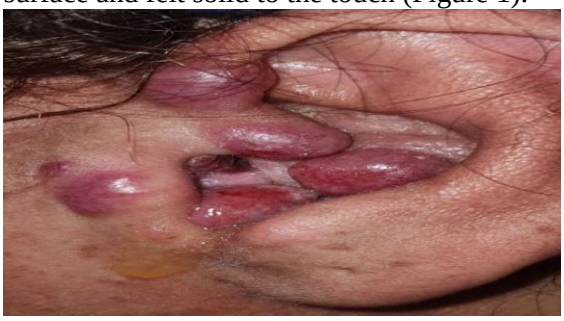

Fig. 1. A close-up image of the patient's left ear, showing reddish-pink lesions in the auricle and external canal.

The tympanic membrane was normal. The audiogram was also normal. There was no lymphadenopathy in the neck or near the ear. Routine blood tests (CBC,diff) were normal. Biopsy was taken from the lesions, which indicated an infection of the hair follicles (folliculitis). These lesions were treated with steroid cream and triamcinolone injection, but without success. While the itching improved, the lesions did not get smaller after about a month of treatment. Since the medical treatment was not effective, the lesions were completely removed during surgery under general anesthesia. Some of the areas were repaired originally, and the defects of the concha region and ear canal were reconstructed by a full-thickness skin graft (Figure 2). The patient had no problems after the operation and was discharged after two days. During the follow-up 15 months after the operation, there was no recurrence. In microscopic examination (histology), the skin tissue was observed with increased blood vessels and a mixture of immune cells, including lymphocytes, plasma cells, and eosinophils. The cells lining the blood vessels (endothelial cells) had large and clear nuclei and pinkish cytoplasm that had an uneven appearance (Figure 3).

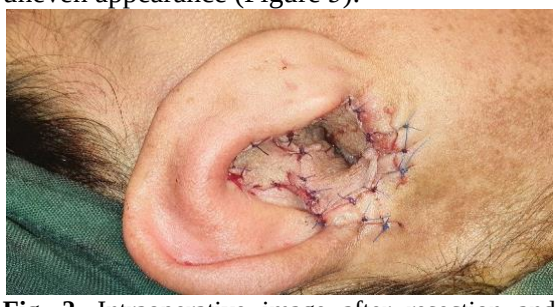

Fig. 2. Intraoperative image after resection and reconstruction with full-thickness skin graft.

Based on these findings, the pathologist diagnosed angiolymphoid hyperplasia with eosinophilia (ALHE). After 15 months of follow-up, the patient showed no signs of recurrence (Figure 4).

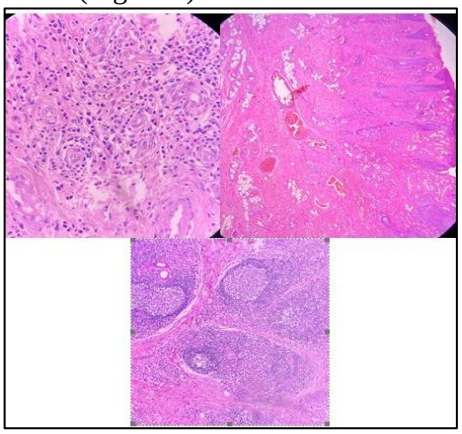

Fig. 3. A microscopic image of the patient's tissue, showing the characteristic features of angiolymphoid hyperplasia with eosinophilia (ALHE). The cells lining the blood vessels (endothelial cells) are plump and have large, clear nuclei. They are surrounded by a mix of immune cells, including lymphocytes (which form clusters called germinal centers) and eosinophils (which appear as pinkish dots).

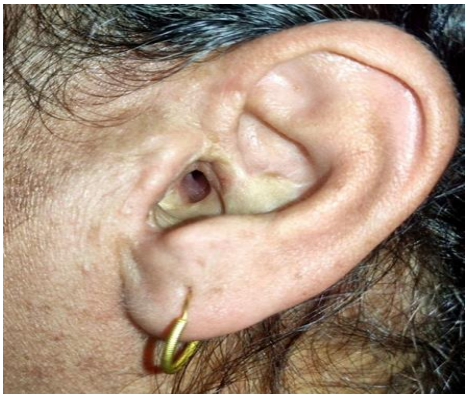

Fig. 4. Reconstruction of the defects after 15 months of surgery with no new lesions and no recurrence.

Discussion

ALHE is a rare disease of unknown etiology that usually presents as multiple nodules. Clinicians must distinguish it from other similar conditions. The main differential diagnosis is Kimura's disease (8). This disease is more common in Asian people and appears as deep lumps or bumps under the skin that often affect the main salivary glands and nearby lymph nodes. People with Kimura disease often have high levels of eosinophils and immunoglobulin E (IgE) in their blood. Other conditions that clinicians should consider include Kaposi's sarcoma, angiosarcoma, epithelioid hemangioendothelioma, cutaneous epithelioid angiomatous nodule (9,10), and cutaneous metastases from cancer.

Studies show that ALHE affects women slightly more often, as seen in our case report. It also typically occurs in young and middle-aged adults (2,11). The exact cause of ALHE is still unknown, and physicians debate whether it is a reactive process or a true tumor of the blood vessels. Some researchers have found a virus called human herpesvirus 8 in ALHE bumps, but this finding has not been consistent across studies (12-14). Others have suggested a possible link with the lymphatic system based on one case, but this requires further investigation (15). Recently, some studies have identified specific immune cells (clonal T cell populations) in ALHE (5,6). This suggests that some types of ALHE may originate from these immune cells and can be considered benign or even low-grade cancer of the lymphatic system (malignant lymphoproliferative disorder) (16). Also, some factors such as pregnancy, trauma, or abnormal blood vessel development may trigger or worsen ALHE (17); we could not identify any of these in our patient. HIV and HCV infections are rarely associated with ALHE, but there are two cases reported in the medical literature (18).

In the microscopic examination (histology) of the patient's tissue, the findings were consistent with what is usually seen in ALHE. In three reported cases, the bumps were located under the skin (subcutaneous). Special tests (immunohistochemistry) showed that most of the immune cells were T cells, but some B cells formed clusters (lymphatic aggregates) (19). While ALHE is usually a benign, slow-growing lump, it often recurs after surgery. In our case, the individual ridges were not contiguous (uncoherent) and were less similar to typical ALHE. These bumps responded better to surgery and were less likely to recur, possibly because it was easier for the doctor to remove them completely. In some rare cases, ALHE bumps can disappear on their own without treatment (20). There are also recent reports of successful treatment using steroids injected directly into the bumps (intralesional corticosteroids) (21). However, in our study, this treatment was effective for only half of the

lesions and resulted in their shrinking. Radiation therapy has also been successful in treating ALHE in some difficult areas, such as the hand (22). We could not use laser therapy in our study because this treatment was not available in our center, and the patient did not want to be referred for laser treatment (16).

Conclusion

Angiolymphoid hyperplasia with eosinophilia (ALHE) should be considered as a potential diagnosis when a patient presents with isolated, well-defined bumps that look like blood vessels and may have a broken surface (ulcer) or crust. While these lesions are typically considered harmless reactions, they can persist for a long time, often do not respond well to treatments, often recur, and in some cases show signs of abnormal growth. Despite ongoing research, questions regarding the etiology and pathogenesis of ALHE remain unanswered. Because ALHE is relatively rare, larger studies involving multiple hospitals are needed to improve our understanding of its causes, how it develops, and the best course of treatment.

Declaration

This study was approved by the ethical committee of Guilan University of Medical Sciences (GUMS) with registration number IR.GUMS.REC. 1404.082.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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