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Case Report

Pinna Swelling an Angiolymphoid Hyperplasia with Eosinophilia: A Rare Case Report

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ABSTRACT

Pinna swelling that is angiolymphoid hyperplasia is a benign lesion that needs to be discussed. Most of the time it affects the face in that preauricular area involved, where it appears as a tiny erythematous lesion. Here we reported a case of a 24-years female patient who presented to us at the hospital with left ear pinna swelling for 7-years.

Keywords
Pinna; Swelling; Angiolymphoid; Hyperplasia; Eosinophilia.

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a benign, locally proliferating lesion, which usually affects middle-aged women. Which most of the time affects the preauricular area and scalp. Other common sites include oral mucosa, pharynx, and orbit lesions present as an erythematous and hyperpigmented lesions. ALHE is a rare benign tumor which is clinically manifested by the presence of dermal papules or nodules measuring about 2-3 cm, varying in color from light brown to pink. The lesions occur preferentially on the face, scalp, auricular region, and neck. There seems to be a higher incidence in females and lesions are more common in patients aged 20-50-years. Its pathogenesis remains unknown. Some authors believe that the damage would be due to a vascular tumor. Others claim that they could represent a reaction to vascular tissue injuries such as skin trauma, infections (human T-cell leukemia-lymphoma virus (HTLV) or herpes virus 8), or hormonal imbalance. Some recent studies tend to consider it as a vascular malformation secondary to a subcutaneous arteriovenous shunt, but the hypothesis most widely accepted is that it is a reactive vascular hyperplasia to various stimuli.

CASE REPORT

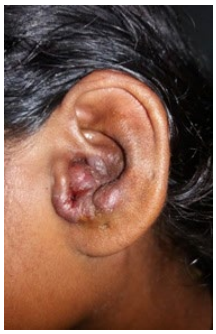
A twenty-four-year-old female patient reported to our ear, nose and throat (ENT) hospital out-patient department (OPD) with chief complaints of left pinna swelling since 7-years. and intermittent itching present over pinna swelling with getting relieved on medications and topical application of steroids. Initially, swelling was very small started as papule 7-years back gradually it gets increased to present size. The patient visited many general practitioners for the swelling but after giving primary treatment of medications and steroid ointment for applications swelling get regressed for some time but later on it recurs. Therefore, on detailed history no history of ear trauma, no history of ear discharge, and no history of any previous ear surgery on the left ear. No history of diabetes mellitus (DM), no history of hypertension (HT), and no history of any known communicable disease.

In the past, the patient had taken antihistamines, anti-inflammatory drugs, and steroids for a few weeks to relieve the swelling, yet it had not decreased in scale.

On palpation tenderness, the swelling was approximately 3 cm×3 cm in size, soft in consistency, and reddish in color, as

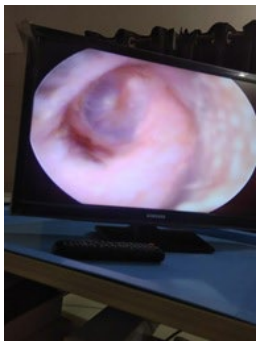
shown in Figure 1. The swelling spreads to the postural area and to the external auditory canal; there is no bleeding when touched, and the swelling is not fixed to the underlying structure. On palpations, pulsation is felt. There were no palpable lymph nodes in the cervical region, as well as in the preauricular and postauricular areas.

Figure 1. *Showing External Auditory Canal Swelling*



Endoscopic examination of the left ear revealed a swelling in the conchal region that extended a few millimetres into the external auditory canal, but the tympanic membrane of the left ear was normal, as shown in Figure 2.

Figure 2. *Endoscopic Picture of Left Tympanic Membrane*



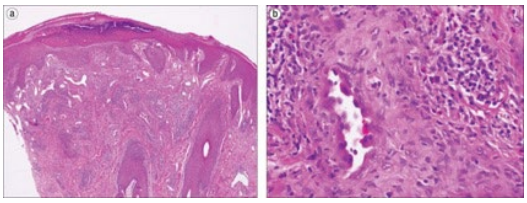
We recommended fine needle aspiration cytology (FNAC) of the swelling after a thorough examination. All routine blood investigations of the patients done for the surgical purpose required under general anesthesia revealed haemoglobin (HB) of 11 gm/dL and eosinophil count raised above normal range by 7%/cumm. 800 UI/mL IgE level raised Blood sugar 100 mg/dL, blood urea 20 mg/dL, Sr creatine 1.0 mg/dL, Hepatitis B Surface Antigen (HBsAg) test non-reactive, human immunodeficiency virus (HIV) test non-reactive, and blood group rhesus (RhB) positive. The reports of electrocardiogram (ECG), 2D echocardiography (2DECHO), and X-ray chest post-eroanterior (PA) view were all normal.

Computerized tomography angiography (CTA) of this swelling revealed a soft density lesion measuring 4 cm×3 cm in preauricular, infra-auricular, and extending to the external auditory canal, triggering luminal stenosis of the left ear's external auditory canal. CTA was performed to rule out any arteriovenous malformation and intracranial communication of this swelling. CTA is used to rule out any possible interaction between brain and swelling.

The arterial phase shows no obvious feeders within swelling while the venous phase shows the venous draining channels venules into the left external jugular vein.

Since the results of the fine needle aspiration cytology were inconclusive, we performed a punch biopsy of the swelling, which revealed Angiolymphoid hyperplasia with eosinophilia of the ear (Figure 3).

Figure 3. *A and B Showing Blood Vessels Endothelial Proliferation with Eosinophilia*



We advised the patient to have the swelling surgically removed after checking all of the blood tests and histopathology reports and explaining all of the complications and chances of the swelling returning after excision. Since the patient refused to undergo surgery, we recommended a different course of treatment i.e., pulsed dye laser therapy, which she accepted. She has taken it as 15 mm circular beam for every 2-months for 3 settings in 6-months where the swelling gets regressed in size but not fully. The patient was pleased with it and has been following-up with it for the last two years with good results.

DISCUSSION

Angiolymphoid hyperplasia with eosinophilia first time described by Wells et al.[1] The tumor is characterized by the proliferation of blood vessels lined by plump endothelial cells and admixed with a dense inflammatory infiltrate of lymphocytes, eosinophils, and mast cells. Weiss and Enzinger argued with the name of the diseases, for they wanted to differentiate the lesion from the malignant vascular tumor, epitheloid hemangioendothelioma. For this, they introduced the term epithelioid hemangioma (EH) in 1982.[2]

The argument that ALHE/EH may represent a monoclonal T-cell process which is supported insome cases.[3] In 2015, statistical analysis yielded no sex 908 patients.[4] Over fifty percent of the patients presented with a single lesion and the most common locations were the ear, and preauricular area, faces, and scalp. Considering age, statistics have shown a wide prevalence range (0.7-months to 91-years) and the mean age of presentation was 37.6-years.

According to the literature ALHE is a benign, locally proliferating lesion, which usually affects middle-aged women and tends to have affects the periauricular area and scalp. Other

common involved areas are oral mucous membranes, pharynx, and orbit[4] lesion present as anerythematous and hyperpigmented lesion.[2,3] The nodules are normally 2 to 3 cm in diameter, with rare cases of larger and deeper neoplasms.[2,3] It is unclear whether ALHE is a reactive or neoplastic disease.[4] In its active phase, it can be misdiagnosed as an angiosarcoma; however, eosinophilia is not a usual feature of malignant angiosarcoma.[5]

The main differential diagnosis of ALHE was put forward as Kimura's disease.[5] These two conditions are histologically described as lymphoid infiltration, vascular proliferation, and tissue eosinophilia. However, the clinical appearance of Kimura's disease is consistent with subcutaneous swelling and may not involve erythematous papules or nodules.[5,6]

Angiolymphoid hyperplasia with eosinophilia may also be confused with lymphomatoid papulosis, which is a form of primary cutaneous CD30+ T-cell lymph proliferative disorder.[7]

Few theories suggest ALHE may occur due to insect bite, injury, and administration of tetanus injection.[8]

No definitive treatment is reported for this condition. Complete excision can be curative, but recurrences are common. Moh's micrographic surgery with excision of abnormal vessels at the base of the lesion may be more effective in reducing recurrences.[3] Intraregional injections of corticosteroids, interferon α-2a, and cytotoxic agent's are effective.[2] Other methods of treatment used for such cases are cryotherapy, laser treatment with carbon dioxide, and pulse dye.[2]

The laser treatment is given for the Vasoformative component of the disease. There are almost many chances of recurrence in such diseases when existed with Kimura disease, we have to excise by surgery and followed by the full thickness skin graft.[9]

Though this lesion described in previous study as presentation to the head and neck and few cases of ear presentation, this case is different as its present at external auditory canal opening with diffuse base and spread to postauricualr region few millimeter deep to skin extending towards the tympanic membrane. Its clinically different with previous case reported findings.

CONCLUSION

This type of lesion is very uncommon in the presentation to the ear. What is distinctive to this case is the external appearance and site to the pinna and external ear and high-levels of immunoglobulin E (IgE) and eosinophilia. Taking into consideration the highly aggressive and cosmetically destructive nature of the disease, more progress should be targeted towards creating a standardized and effective therapeutic approach that could help physicians treat such a recurring disease.

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ETHICAL APPROVAL

This study approved by the institutional ethical committee

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

REFERENCES

1. Wells GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. *Br J Dermatol.* 1969; 81(1): 1-14. doi: 10.1111/j.1365-2133.1969.tb15914.x

2. Guo R, Gavino AC. Angiolymphoid hyperplasia with eosinophilia. *Arch Pathol Lab Med.* 2015; 139(5):683-686. doi: 10.5858/arpa.2013-0334-RS

3. Asadi AK. Angiolymphoid hyperplasia with eosinophilia. *Dermatol Online J.* 2002; 8(2): 10.

4. Olsen TG, Helwig EB. Angiolymphoid hyperplasia with eosinophilia.A clinicopathologic study of 116 patients. *J Am Acad Dermatol.* 1985; 12(5 Pt 1): 781-796. doi: 10.1016/s0190-9622(85)70098-9

5. Chong WS, Thomas A, Goh CL. 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.* 2006; 45(2): 139-145. doi: 10.1111/j.1365-4632.2004.02361.x

6. Reed RJ, Terazakis N. Subcutaneous angioblastic lymphoid hyperplasia with eosinophilia (Kimura's disease). *Cancer.* 1972; 29(2): 489-497. doi: 10.1002/1097-0142(197202)29:2<489::aid-cncr2820290239>3.0.co;2-x

7. Diamantidis MD, Myrou AD. Perils and pitfalls regarding differential diagnosis and treatment of primary cutaneous anaplastic large-cell lymphoma. *Scientific World Journal.* 2011; 11: 1048-1055. doi: 10.1100/tsw.2011.80

8. Akosa AB, Ali MH, Khoo CT, Evans DM. Angiolymphoid hyperplasia with eosinophilia associated with tetanus toxoid vaccination. *Histopathology.* 1990; 16(6): 589-593. doi: 10.1111/j.1365-2559.1990.tb01164.x

9. Jun R, Liu XK, Zeng K. Successful treatment of angiolymphoid hyperplasia with eosinophilia and Kimura's disease in the same patient with surgery. *Dermatol Ther.* 2014; 27(1): 36-38. doi: 10.1111/dth.12040