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# A fleshy papule on the eyelid

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## Abstract

Angiolymphoid hyperplasia with eosinophilia is an uncommon tumor affecting the head and neck region. It usually presents as solitary or multiple erythematous or brownish papules and nodules. It is considered a reactive angioproliferative disorder by some, whereas others believe it to be a neoplastic growth. Involvement of the eyelid is a rare occurrence. We report an instance of angiolymphoid hyperplasia with eosinophilia involving the eyelid in a 19-year-old woman with review of literature.

*Keywords: angiolymphoid hyperplasia with eosinophilia, plump endothelial cells, eyelid masses*

## Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon vasoproliferative disorder first described by Wells and Whimster in 1969 [1]. It usually presents as solitary or multiple erythematous to brownish papules or nodules of the head and neck region, predominantly involving the external ear and the periauricular skin. Involvement of other areas is uncommon. We report an unusual instance of this disorder presenting as an asymptomatic fleshy papule involving the eyelid in a 19-year-old woman.

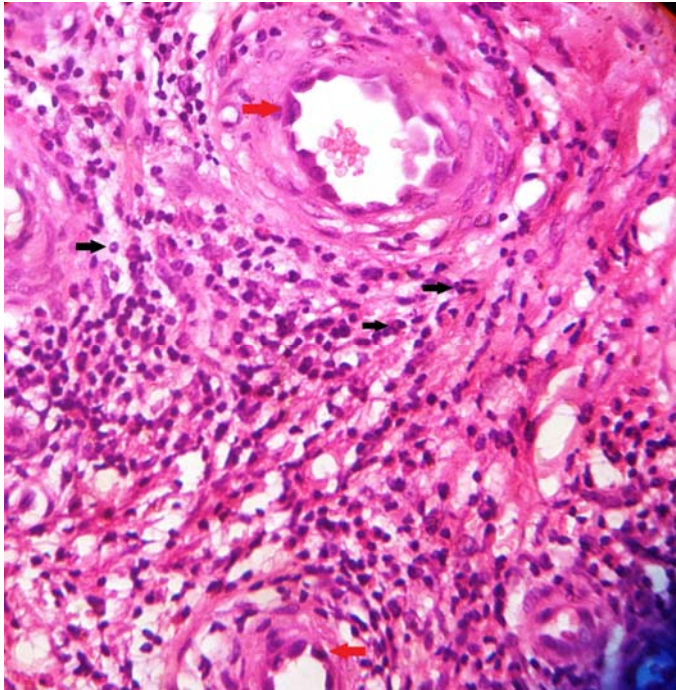
## Case Synopsis

A 19-year-old woman presented with a three-month history of an asymptomatic fleshy papule on the

right upper eyelid margin near the lateral canthus measuring about 3 mm×3mm (**Figure 1**). There was no trauma preceding the development of the papule and there was no bleeding spontaneously or on manipulation. The rest of the cutaneous and systemic examination was unremarkable. The lesion was completely excised under local anesthesia for histopathological analysis, which revealed a diffuse dermal infiltrate of lymphocytes and eosinophils that



**Figure 1.** A fleshy 3×3mm papule on the right upper eyelid margin near the outer canthus.



**Figure 2.** Photomicrograph showing diffuse infiltrate of lymphocytes and eosinophils (black arrows) with intervening capillary-like vessels that can be seen conspicuously lined by large 'plump' endothelial cells (red arrows). H&E, 40x.

were interspersed by multiple capillary-like vessels lined by plump endothelial cells (**Figure 2**). Based on the characteristic histopathological features a diagnosis of ALHE was established.

## Case Discussion

Angiolymphoid hyperplasia with eosinophilia is an uncommon vasoproliferative disorder first described by Wells and Whimster in 1969 [1]. It commonly affects middle aged Asians with a slight female preponderance. It has also been described by several other names as epithelioid hemangioma, histiocytoid hemangioma, papular angioplasia, inflammatory arteriovenous hemangioma, and pseudopyogenic granuloma [2, 3]. It usually presents as solitary or multiple erythematous to brownish papules or nodules of the head and neck region, mostly about the ear. Classically, ALHE presents as clusters of erythematous translucent papules and nodules involving the external ear, especially the pre- and retro-auricular skin. Involvement of the scalp and forehead (along the hairline) is also common. The trunk, extremities, and genitalia can be

infrequently affected [2, 3]. There is no agreement on whether this disorder is a reactive angioproliferative disorder or a neoplastic proliferation of vessels. Peripheral eosinophilia maybe seen in about 20% of the cases, but was not seen in our case. Histopathologically, ALHE is characterized by diffuse infiltrate of lymphocytes and eosinophils throughout the lesion. The former may be seen organized as lymphoid follicles at places as well. The other distinctive component is the proliferation of small and large capillary-like vessels lined by the conspicuous 'plump' or 'epithelioid' endothelial cells. Other characteristic features that may be seen include different types of proliferating vessels and endothelial cell aggregates without luminal differentiation, especially around the larger vessels [4, 5].

The clinical differential diagnosis in our case includes eccrine hydrocystoma that almost always occurs in the vicinity of the eyelid [6]. The classical lesion has a translucent cystic morphology. Pyogenic granuloma can involve the eyelid or conjunctiva, which could be sessile or pedunculated and usually has a bright red appearance. It is a friable vascular lesion and may be associated with bleeding on manipulation [7]. Kimura disease, which was previously believed to be a variant of AHLE owing to clinical and histological resemblance, has now been established as a separate entity, although there are a few rare instances of AHLE co-existing with Kimura disease [8]. Clinically, Kimura disease is characterized by a deep seated large swelling often associated with lymphadenopathy and without any surface skin changes. Histologically, multiple lymphoid follicles with germinal centers and dense eosinophilic infiltrate with eosinophilic abscesses characterize Kimura disease, which also exhibits minimal vascular proliferation and the absence of epithelioid endothelial cells [5]. There are a few reports of ALHE involving the orbit and the ocular adnexae, either unilaterally or bilaterally [5, 9, 10].

Definitive treatment is complete excision as pharmacologic measures (systemic or intralesional corticosteroids, topical imiquimod, oral isotretinoin, and methotrexate), electro-, radio- or cryosurgical procedures, and laser ablation are associated with

recurrences [5]. The excisional biopsy performed in our case was curative.

## Conclusion

Although a rare disorder involving the eye, it is

recommended to consider ALHE in the differential diagnosis of eyelid masses.

## Potential conflicts of interest

The authors declare no conflicts of interests

## References

1. Wells GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. *Br J Dermatol*. 1969; 81:1-14. [PMID: 5763634].
2. Calonje E. Soft-tissue tumours and tumour-like conditions. In: Burns T, Breathnach S, Cox N, Griffiths C, editors. *Rook's Textbook of Dermatology*, 8<sup>th</sup> ed. Oxford: Wiley-Blackwell; 2010. p. 56.1-56.62.
3. North PE, Kincannon J. Vascular neoplasms and neoplastic-like proliferations. In: Bologna JL, Jorizzo JL, Rapini RP editors. *Dermatology*, 2<sup>nd</sup> ed. London: Elsevier, 2008. p. 1771-1794.
4. Taylor SK, Glusac EJ, Meyerle JH (eds). Angiolymphoid hyperplasia with eosinophilia. 2016. <https://emedicine.medscape.com/article/1082603-overview#a6>. Accessed on March 26, 2018.
5. Mukherjee B, Kadaskar J, Priyadarshini O, Krishnakumar S, Biswas J. Angiolymphoid hyperplasia with eosinophilia of the orbit and adnexa. *Ocul Oncol Pathol*. 2015; 2: 40-7. [PMID: 27171790].
6. Singh AD, McCloskey L, Parsons MA, Slater DN. Eccrine hidrocystoma of the eyelid. *Eye (Lond)*. 2005; 19:77-9. [PMID: 15205675].
7. Ergün SS, Kocabora MS, Su O, Demirkesen C. Surgical treatment of giant pyogenic granuloma of the upper eyelid. *Ann Ophthalmol (Skokie)*. 2007; 39:264-6. [PMID: 18025641].
8. Marka A, Cowdrey MCE, Carter JB, Lansigan F, Yan S, LeBlanc RE. Angiolymphoid hyperplasia with eosinophilia and Kimura disease overlap, with evidence of diffuse visceral involvement. *J Cutan Pathol*. 2019; 46:138-142. [PMID: 30362278].
9. Miličić B, Velnar T, Pregelj R, Limbaeck-Stokin C. Multiple epithelioid hemangiomas with orbital involvement. *Case Rep Pathol*. 2015; 2015:629805. [PMID: 25802787].
10. Ueda S, Goto H, Usui Y, Nagai T, Nagao T. Angiolymphoid hyperplasia with eosinophilia occurring in bilateral eyelids. *BMC Ophthalmol*. 2013; 13:38. [PMID: 23914723].