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Short communication

# Rare lesion, unusual location, uncommon presentation: a case of angiolymphoid hyperplasia with eosinophilia

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**Abstract**

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare vascular proliferation of unknown pathogenesis that may be related to trauma. Although it affects mainly the head and neck, the zygomatic area is rarely involved. We report a case that affected the zygomatic region of a 46-year-old black man. The lesion had been present for about a year and the patient reported that it appeared after a facial injury during a soccer match. Clinical and tomographic investigations suggested a benign tumour, and the lesion was excised through an intraoral approach. Histopathological examination showed the unexpected diagnosis of ALHE. This tumour was interesting because of its rarity, and also because of its unusual site within the head and neck region. The diagnosis of ALHE is hardly ever considered in the differential diagnosis of zygomatic nodules.


*Keywords:* Oral and maxillofacial pathology; Oral surgery; Vascular tumours; Zygomatic region

## Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare vascular lesion of unknown aetiology that is characterised by blood vessels that are lined by prominent, large, "epithelioid" endothelial cells, and an inflammatory infiltrate that is rich in eosinophils and organised into lymphoid follicles.[1,2] Most affected patients are in their third to fifth decades of life, and it has a predilection for white women.[3] Here we report a case of ALHE that presented as a subcutaneous nodule in a black man.

## Case report

A 46-year-old black man presented with a subcutaneous nodule of one year's duration in the zygomatic area. The lesion had apparently appeared after an injury that occurred while he was playing a game of soccer. Clinical and tomographic examination showed a well-circumscribed subcutaneous nodule that measured 4.0 cm in its greatest dimension, which suggested a benign tumour (Fig. 1). Because of its site, and considering that he had aesthetic concerns, the lesion was excised through an intraoral approach. The biopsy specimen was oval, yellow-brown in colour, fibrous, and had a smooth surface. Microscopic analysis showed a proliferation of blood vessels that were lined by plump endothelial cells, sometimes showing epithelioid morphology, and mixed with an intense mononuclear inflammatory infiltrate and many eosinophils (Figs. 2 and 3). Immunostaining for smooth mus-

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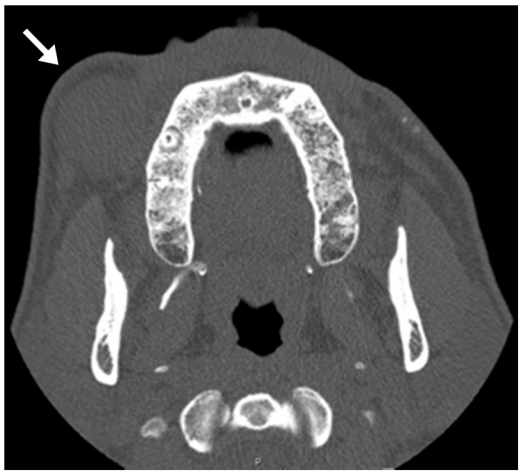

Fig. 1. Tomographic image showing no infiltration into the surrounding tissue (arrow).

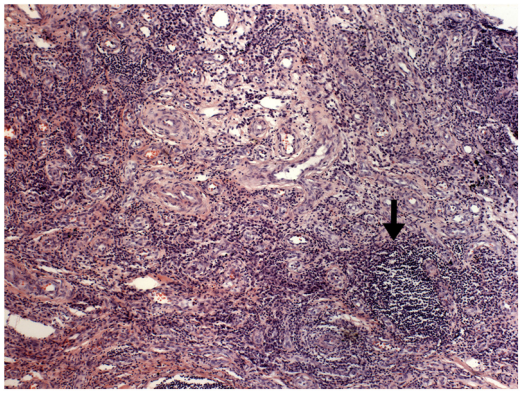

Fig. 2. Low-power view showing an intense mononuclear inflammatory infiltrate. Note the formation of the lymphoid follicle (arrow) (haematoxylin and eosin, original magnification ×25).

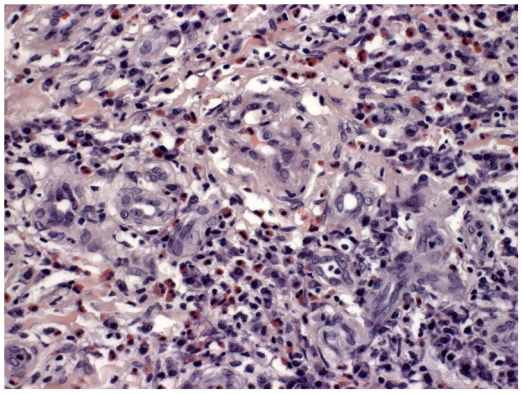

Fig. 3. A higher view that highlights the proliferation of epithelioid vessels mixed with a large number of eosinophils (haematoxylin and eosin, original magnification ×100).

cle actin and CD34 highlighted that the lesion was vascular. The final diagnosis was angiolymphoid hyperplasia with eosinophilia.

## Discussion

ALHE, also known as epithelioid haemangioma, is a vascular proliferation that was first described in 1969.[1] Its pathogenesis is uncertain; some authors consider it to be a benign vascular neoplasm, while others think that it is probably a secondary vascular proliferation that may be related to injury.[4] In the present case the possibility that the lesion was reactive is more likely, as there was a history of local trauma.

The head and neck is certainly the expected site for ALHE, but the skin of the zygomatic area is rarely involved.[2,5] We found only one case that was similar to the present one in site, size, and clinical and tomographic findings.[2] The main difference between that study and the case reported here is the surgical access, which in our case was intraoral.

Kimura's disease, which is usually considered in the differential diagnosis of ALHE, has a similar histological picture, but it is usually defined by three main characteristics: subcutaneous masses, eosinophilia in the peripheral blood, and high serum concentrations of IgE.[3] In the present case, all the patient's blood tests were within the normal ranges, including the eosinophil count.

The present case was the first seen in our oral pathology service, which has dealt with over 95,000 biopsy specimens. It is an interesting case because the diagnosis of ALHE is unlikely to be considered in the differential diagnosis of zygomatic nodules. The intraoral surgical approach showed itself to be a good option, particularly in that it avoided undesirable scars on the face.

## Ethics statement/confirmation of patients' permission

Neither is required.

## Conflict of interest

We have no conflicts of interest.

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