Short Report

Angiolymphoid Hyperplasia with Eosinophilia Presenting as an Ulnar Artery Pseudoaneurysm

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ABSTRACT

Introduction: Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon proliferative benign lesion, which most commonly affects the skin of the head and neck. Noncutaneous localization of this pathology is unusual, and it is rare in the extremities.

Report: We herein report a case of ALHE presenting as an ulnar artery pseudoaneurysm. This case revealed eosinophilia, however, after the operation, the count of eosinophils had decreased to within the normal range.

Discussion: ALHE should be considered in the differential diagnosis of a pulsatile mass in the extremities.

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Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is an unusual vascular proliferation with stromal inflammation occurring predominantly in the dermis and subcutaneous tissue of the head and neck. There has been considerable controversy about whether ALHE and Kimura’s disease (KD) are related. ALHE affecting large muscular arteries is extremely rare, and a review of the literature revealed only a few reports of ALHE affecting arteries in the extremities. We herein report a case of ALHE presenting as an ulnar artery pseudoaneurysm.

Report

A 53-year-old female presented with a painless mass in her left forearm. On physical examination, she had a 3 cm × 3 cm mobile, pulsatile mass in the left forearm. The left radial and ulnar arteries were palpable, and the pulse status was normal in all the other extremities. She had a history of left brachial artery puncture for an angiogram 15 years prior. She had a past medical history of uveitis that had been treated with steroids for 1 year and was successfully cured. The laboratory findings revealed mild eosinophilia (700/μl). We couldn’t find no cause of eosinophilia.

Computed tomography showed a mass in her left forearm. Arteriography demonstrated a 3 cm diameter pseudoaneurysm of the left proximal ulnar artery just branched from the brachial artery (Fig. 1). The patient was diagnosed with an ulcerated pseudoaneurysm. To prevent distal emboli and rupture, the patient was taken to surgery. During the operation, the mass was totally resected, with proximal and distal ligation of the ulnar artery, without ischemic complications of the left hand. The histology of the resected aneurysmal wall revealed no proper structures of the arterial wall. The tissue was infiltrated with numerous eosinophils (Fig. 2a and b), and we diagnosed this case as ALHE. The patient did well postoperatively, and the eosinophilia gradually resolved after the operation. After 3 months, the count of eosinophils had decreased to within the normal range.

Discussion

ALHE was first described by Wells and Whimster in 1969. The term was later included under the umbrella classification of histiocytoid hemangiomas described by Rosai in 1979. It has been debated whether ALHE is a reactive or neoplastic lesion, as is reflected by the various names given to the condition, including epithelioid hemangioma, pseudopyogenic granuloma, and inflammatory angiomatous nodules.

Typically, ALHE presents in young to middle-aged females, with no predilection for race. The clinical appearance is usually one of bright red papules or nodules in the dermis or
subcutaneous tissue of the head and neck region.\(^1\) ALHE distinguishes itself from other hemangiomas by its histologically unique appearance. The most typical findings are a histiocyte-like endothelium with cuboidal to dome-shaped endothelial cells lining a florid vascular proliferation. Additionally, there may be heavy inflammatory infiltration of lymphocytes and eosinophils.\(^2\) Occasionally, ALHE leads to peripheral blood eosinophilia.\(^3\) In our case, the endothelioid cells demonstrated the typical appearance of large, round, vesicular nuclei within a dome-shaped cell. Additionally, an intense vascular proliferation surrounded by an abundance of lymphocytes and eosinophils was observed. The microscopic findings also showed a lymphoid follicle. Although the presence of a lymphoid follicle does not rule out a diagnosis of ALHE, their presence can make it difficult to differentially diagnose ALHE from KD.

Clinically, KD is associated with regional lymphadenopathy accompanied by peripheral blood eosinophilia and elevated serum IgE.\(^2\) Histologically, KD differs from ALHE in that the former lacks the dome-shaped endothelioid cells, and is characterized by the presence of multiple lymphoid follicles combined with massive eosinophilic infiltration and eosinophilic abscesses.\(^2\)

The pathogenesis of ALHE has yet to be elucidated. It has been considered to be a true vascular neoplasm of unknown etiology, while others believe that the proliferation is reactive to arterial or venous traumatic insults.\(^2\) Morton et al.\(^4\) suggest that trauma may be an initiating factor or may alter the disease process. In the present patient, there was a history of the left brachial artery puncture, which may have influenced the occurrence of ALHE in this case.

Interestingly, the eosinophilia in our case resolved after the resection of the diagnosed ulnar artery pseudoaneurysm. Sekino et al.\(^5\) also reported a pseudoaneurysm of the ulnar artery with eosinophilia, which resolved after the resection. In their report, there was no evidence that eosinophils had infiltrated into the arterial wall, however, in our case, the eosinophils had infiltrated the aneurysmal wall. This supports the theory that the eosinophilia in our case was caused by ALHE. The aneurysm infiltrated with eosinophils continued the eosinophilia, and after the resection of the origin of eosinophilia, the eosinophilia spontaneously resolved.

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**Conflict of Interest**
None.

**References**