Nontraumatic pseudoaneurysm of the proximal ulnar artery with eosinophilia

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Proximal ulnar artery aneurysms, including pseudoaneurysms, have not been described in the English literature. We report a nontraumatic pseudoaneurysm of the proximal ulnar artery with eosinophilia in a 54-year-old man. Radial, coronary, and hepatic artery aneurysms associated with eosinophilia in idiopathic hypereosinophilic syndrome or allergic granulomatous angiitis (Churg Strauss syndrome) have been reported. Although it is unclear in the present case whether eosinophilia was associated with the pseudoaneurysm of the ulnar artery, eosinophil infiltration into the aneurysmal wall may have influenced vascular injury as the cause of the pseudoaneurysm formation. (J Vasc Surg 2005;42:1233–5.)

Upper-extremity arterial aneurysms are uncommon lesions and are most commonly false aneurysms. Aneurysms of the distal ulnar artery complicating hypothenar hammer syndrome have been frequently reported. To our knowledge, however, aneurysms of the proximal ulnar artery, including pseudoaneurysms, have not been described in the literature. We report a case of a pseudoaneurysm of the proximal ulnar artery with eosinophilia and discuss the association of the aneurysm and eosinophilia.

CASE REPORT

A 54-year-old man had an approximately 3-cm × 3-cm pulsatile mass, without pain and tenderness, in the right forearm near the antecubital fossa. The right radial and ulnar arteries were well palpable, and the pulse status was normal in all the other extremities. He had had no thromboembolic event of the right hand, no history of forearm trauma or brachial artery puncture, and no history of smoking or drug use. There was no family history atopy or vasculitis.

Magnetic resonance arteriography showed a saccular and loculated aneurysm of the proximal ulnar artery immediately distal to the brachial artery (Fig 1). On the blood laboratory test, remarkable eosinophilia (16,300/mm3) and an increased serum creatinine level (1.79 mg/dL) were identified. Aspiration of the bone marrow showed nonspecific findings for eosinophilia. A biopsy specimen of the kidney revealed no vasculitis. Auscultation revealed no heart murmur, and transthoracic echocardiography showed no valvular heart disease.

The patient had sustained rhinitis for a year before the operation, and carbocysteine and clarithromycin had been administered. The etiology of his rhinitis might have an allergic factor, because head radiography showed no sign of sinusitis, and his serum immunoglobulin E (IgE) level was increased to 6,220 (<250) IU/mL. C-reactive protein was within the normal level (0.11 mg/dL), and test results for antinuclear antibody, perinuclear-antineutrophil cytoplasmic antibody, serine proteinase 3-antineutrophil cytoplasmic antibody, and cryoglobulin were all negative.

The patient was diagnosed with ulnar artery pseudoaneurysm. To prevent distal emboli and rupture, the patient was taken to

Fig 1. Magnetic resonance arteriography showed a saccular and loculated aneurysm of the proximal ulnar artery just branched from the brachial artery.
surgery where the aneurysm was found to arise from the ulnar artery, to be adherent to surrounding tissues, and to involve the radial artery (Fig 2). We performed partial resection of the aneurysm and ligation of the ulnar artery both proximal and distal to the aneurysm, without ischemic complications of the hand. The aneurysm was opened, and all the branches of the aneurysm were completely sutured and closed from inside.

Microscopic appearance of the resected aneurysmal wall revealed no proper structures of the arterial wall. The tissue was infiltrated with numerous eosinophils (Fig. 3). The eosinophilia gradually resolved after the operation, and the eosinophils vastly decreased to 320 /mm$^3$ at the 7-months follow-up.

**DISCUSSION**

Radial, coronary, and hepatic artery aneurysms associated with eosinophilia have been reported. Khaira et al$^3$ reported angiolymphoid hyperplasia with tissue eosinophilia presenting as a radial artery aneurysm in a 28-year-old man with a mild eosinophilia (1,000/$\mu$L).

Okinaka et al$^4$ presented coexistence of a giant aneurysm of the sinus of Valsalva and multiple coronary artery aneurysms associated with an idiopathic hypert eosinophilic syndrome (HES) in a 53-year-old woman. The patient’s sustained eosinophilia (9,000/$\mu$L), elevated eosinophilic cationic protein concentration, and pathologic findings of eosinophil infiltration of the aortic wall suggested the association of eosinophilia-induced vascular injury as the cause of these aneurysms.

Nakamura et al$^5$ described a ruptured hepatic artery aneurysm in a 46-year-old woman with allergic granulomatous angiitis (AGA) (Churg Strauss syndrome), a disorder characterized by hypereosinophilia (6,700/$\mu$L in the case by Nakamura et al) and systemic vasculitis that may occur in individuals with asthma and allergic rhinitis. Although eosinophilic tissue infiltration is emphasized as a characteristic finding in AGA, it was not observed anywhere in the resected specimens, including the wall of the ruptured hepatic artery in the case by Nakamura et al. They stated that the absence of eosinophilic tissue infiltration could be explained by the effect of prednisolone administration.

Drogue et al$^6$ reported an aneurysm of the right coronary artery in a 54-year-old woman. The patient was diagnosed with Churg Strauss syndrome after the successive occurrences over a 6-year period of asthma, eosinophilia that peaked at 18,600/$\mu$L, and mononeuritis multiplex.

As just mentioned, arterial aneurysm or dissection can be associated with HES or AGA. Three defining features of HES are (1) sustained blood eosinophilia of >1,500/$\mu$L present for >6 months, (2) absence of other apparent etiologies for eosinophilia, including parasitic infections and allergic diseases, and (3) signs and symptoms of organ involvement. Our patient had allergic rhinitis as an etiology for eosinophilia.

The diagnosis of AGA (Churg Strauss syndrome) is established clinically. The presence of asthma, rhinitis, or sinusitis associated with peripheral eosinophilia and symptoms suggesting a vasculitis supported a diagnosis, but a tissue biopsy specimen should be obtained to document the pathologic features of the disease. In this patient,
despite the presence of rhinitis and mononeuropathy, the biopsy specimen of the dysfunctional kidney revealed no vasculitis.

Eosinophils possess phagocytic and oxidative functions similar to neutrophils, and the release from eosinophilic granules of cationic proteins, such as eosinophil peroxidases, lysophospholipases, eosinophil-derived neurotoxin, eosinophil granule major basic protein, and eosinophilic cationic protein, has been shown to produce tissue damage. Eosinophils have the potential to secrete cytokines, including interleukin (IL)-1 and IL-5, and may act as antigen-presenting cells in association with major histocompatibility complex class II molecules. Eosinophils release chemoattractants for other granulocytes as well as release leukotriene C4, which increases vascular permeability.

The presence of large numbers of eosinophils and quantities of IgE in the vessels and tissue of patients with Churg Strauss syndrome, along with toxic proteins such as eosinophil-derived neurotoxin and major basic protein, suggests a direct role in the pathogenesis of the vasculitis. Potential mechanisms of eosinophil-related tissue damage have been appreciated, including tissue infiltration with eosinophils (space occupying), damage relating to an eosinophil function, and damage related to eosinophil products.

It is unclear in the present case whether eosinophilia was associated with the pseudoaneurysm of the ulnar artery because, unfortunately, the arterial wall was not identified in the pathology specimen. Thus, there was no direct evidence that the eosinophils had infiltrated into the wall of the artery, which at least lends credence to the theory that this was caused by the eosinophilia. Eosinophil infiltration into the pseudoaneurysmal wall, however, may have influenced vascular injury as the cause of the pseudoaneurysm formation. When we encounter a true aneurysm of the artery, which is seldom involved by arteriosclerosis or a nontraumatic pseudoaneurysm, the search for a systemic disorder, including eosinophilia as in the present case, as the etiology of aneurysm formation is recommended.

REFERENCES


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