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Multiple Scalp Aneurysms Caused by Atypical Temporal Arteritis

—Case Report—

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Abstract

A 55-year-old male suffered sudden onset of dysarthria and mild left hemiparesis due to a right intracerebral small hemorrhage. On admission, six subcutaneous elastic hard lumps were found on the scalp with painless and regular pulsation. The lumps were located along the course of the bilateral superficial temporal arteries (5 locations) and the occipital artery. The patient did not have symptoms of headache or blurred vision associated with temporal arteritis. The largest lump was removed for cosmetic reasons and definitive diagnosis. Histological examination demonstrated many infiltrating inflammatory cells along the entire vascular wall but without giant cells or fibrinoid necrosis. These multiple scalp aneurysms were probably caused by atypical temporal arteritis.

Key words: arteritis, multiple scalp aneurysms, superficial temporal artery

Introduction

Scalp subcutaneous nodules may be caused by various diseases, such as epidermoid cyst, angioma, lipoma, neurofibroma, periarteritis nodosa, aneurysm due to trauma or angiitis, and angiolymphoid hyperplasia with eosinophilia.[7] Periarteritis nodosa, aneurysm, and angiolymphoid hyperplasia with eosinophilia are related to inflammatory alteration of the vascular structures.[1-8] Aneurysmal change of the superficial temporal artery (STA) is often caused by post-traumatic vascular initial dissection, and sometimes by arteritis. Temporal arteritis, which is characterized by giant cells and fibrinoid necrosis, is a well-known disease causing headache and visual acuity disturbance.[1,3,4,6,8] The definitive diagnosis is based on the typical clinical symptoms, laboratory data, and pathological findings.[3] We treated a patient with multiple scalp aneurysms due to atypical temporal arteritis without the typical histological characteristics.

Case Report

A 55-year-old male presented with abrupt onset of dysarthria and weakness of the left upper extremity. He was immediately taken to our hospital by ambulance. Neurological examination found dysarthria and left hemiparesis. Computed tomography showed a small high-density lesion at the right internal capsule. He was admitted for treatment of intracerebral hemorrhage. His previous medical history revealed only hypertension, and his blood pressure was 160/90 mmHg on admission. Physical examination revealed six painless and elastic hard lumps pulsating on the bilateral scalp. Five of the lumps were located along the course of the STA: two in front of the ears, two on a peripheral portion of the right frontal branch, and one on the left parietal branch. The other lump was on the left occipital artery (OA). The patient reported that these sites had felt itchy about 4 years ago, but had not shown any sensation since then. However, the lesions gradually enlarged to 3 by 4 cm in diameter. The patient had had another lump on the left forehead, but this was

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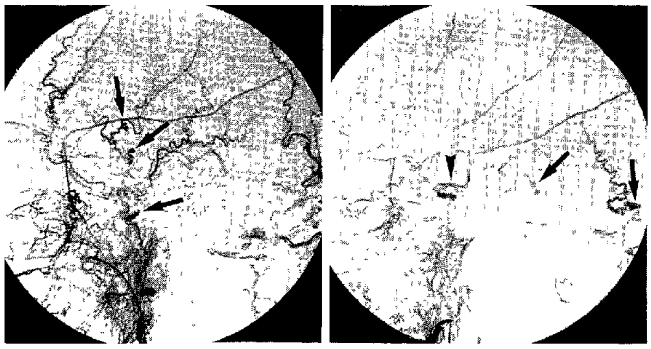

Fig. 1 ***left*: Right external carotid angiogram (lateral view) showing tortuosity of the superficial temporal artery and the occipital artery (OA), and three lesions associated with pooling of contrast medium (*arrows*). *right*: Left external carotid angiogram (lateral view) showing tortuosity of the OA and three lesions associated with pooling of contrast medium (*arrows*). The largest lesion, which was later removed, was slowly and tortuously filled (*arrowhead*).**

removed by a surgeon 2 years earlier for cosmetic reasons. He had never experienced headache, ophthalmic pain, blurred vision, or insect bite. There was no history of prior trauma, systemic illness, or allergy.

Laboratory examination revealed a white blood cell count of 7000/mm^3, with 65.0% neutrophils, 23.7% lymphocytes, 7.5% monocytes, and 3.4% eosinocytes. The platelet count was 20.4 $\times$ 10^4/μl. The erythrocyte sedimentation rate was 6 mm per hour. Auto-immune studies, i.e., an lupus erythematosus test (anti-nuclear antibody), anti-Sm antibody, and anti-deoxyribonucleic acid antibody, were all negative.

Bilateral external carotid angiography showed remarkable tortuousity of the STAs and OAs, and several slow-filling pools of contrast medium coincided with the locations of the lumps (Fig. 1). Internal carotid angiography detected no vascular abnormality. One month after the onset of the intracerebral hemorrhage, the largest lesion, involving a serpentine dilatation of the left STA at the front of the left ear, was removed for cosmetic reasons and definitive diagnosis (Figs. 2 and 3). The lesion had dilated along a serpentine pathway over the zygoma and was elastic hard and dark reddish in color. Histological examination demonstrated that the intima was fibrous and severely thickened with hyalinization, and the media and the adventitia had undergone fibrous changes (Fig. 4). The lumen was narrowed and filled with thrombus. Lymphocytes,

Fig. 2 Photograph showing the largest lump in front of the left ear.

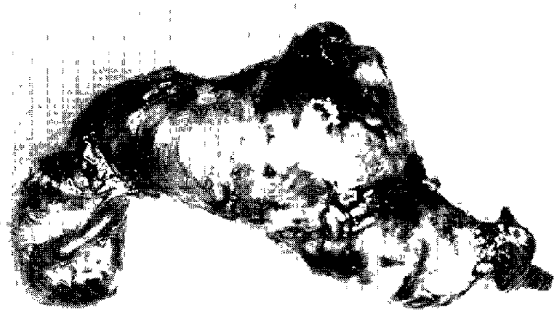

Fig. 3 Photograph showing the serpentine dilatation removed from the superficial temporal artery.

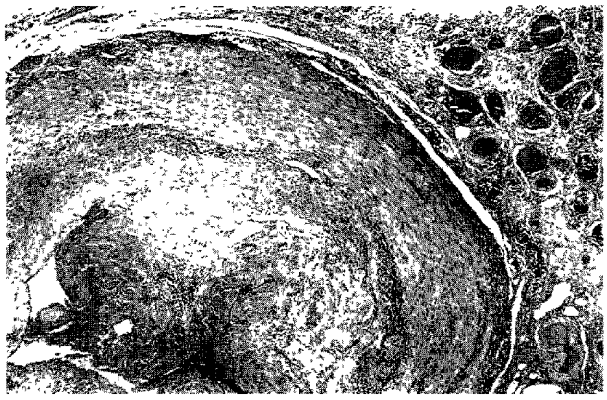

Fig. 4 Photomicrograph revealing that the intima is fibrous and severely thickened with hyalinization, and fibrous changes in the media and the adventitia. HE stain, $\times$4.

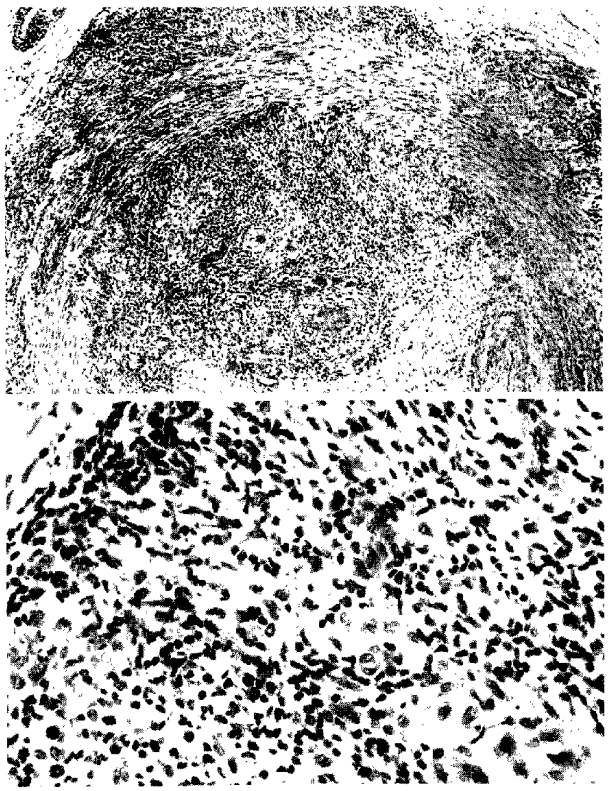

Fig. 5 Photomicrographs demonstrating that lymphocytes, eosinophils, and neutrophils have extensively infiltrated into the whole wall. HE stain, *upper*: ×4, *lower*: ×10.

eosinophils, and neutrophils had extensively infiltrated into the intima and the media, as if moving toward the internal elastic lamina (Fig. 5). No giant cells or fibrinoid necrosis were detected. These findings suggested nonspecific temporal arteritis.

The postoperative clinical course was uneventful. His left hemiparesis gradually improved. He demonstrated no inflammatory signs and was discharged from our hospital. There has not been any change in the residual aneurysms.

Discussion

The histological findings suggested that the multiple scalp aneurysms in this patient caused by nonspecific arteritis in the bilateral STAs. The histological features of classical temporal arteritis generally involve granulomatous arteritis with prominent Langerhans giant cells, predominantly at the media of the artery with smooth muscle necrosis; nonspecific inflammation reaction, with lymphocytes, neutrophils, and eosinophils concentrated at the arterial wall; and intimal fibrosis with occlusion of the vessel lumen.[3] Histological examination in our patient revealed no giant cells or fibrinoid necrosis, but the granulomatous and fibrotic changes of the intima and severe infiltration of inflammatory cells observed along the whole wall were similar to the findings of temporal arteritis. The major clinical symptoms in classical temporal arteritis are headache, polymyalgia rheumatica, jaw claudication, diaphoresis, anorexia, malaise, extremity claudication, and blurred vision.[3] Our patient only reported previous itchy feeling. His erythrocyte sedimentation rate was certainly not elevated. Therefore, the pathogenesis seemed to be different from that of typical temporal arteritis.

Accumulation of eosinophils along the entire vascular wall is more often related to hypersensitive angiitis and might provoke lymphoid granulomatous inflammation with eosinophilic infiltration inducing the prominence.[2,7] However, our patient had no increase in the eosinophil count in the blood and he had not experienced asthma or allergic diseases. In addition, these lesions were limited to the bilateral STAs and OAs. The eosinophilic accumulation might be involved in temporal arteritis. Such atypical temporal arteritis may cause multiple aneurysms. Non-bilateral multiple scalp aneurysms are rare, and it is not known why only bilateral temporal arteries are affected.

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