

Arterial Graft to Treat Ruptured Distal Middle Cerebral Artery Aneurysms in a Patient With Mucosa-Associated Lymphoid Tissue Lymphoma

—Case Report—

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Abstract

A 60-year-old man was admitted to our hospital with sudden onset of motor aphasia, Gerstmann syndrome, and incomplete right hemiparesis one week after administration of chemotherapy for mucosa-associated lymphoid tissue (MALT) lymphoma. Computed tomography showed an intracerebral hematoma in the left subcortical area. Cerebral angiography revealed 2 aneurysms on the distal middle cerebral artery (MCA) that increased in size in the course of 2 weeks. The aneurysms were excised and a bypass placed using a superficial temporal artery (STA) graft. Histological study showed no bacteria, infiltration of inflammatory cells, or lymphoma cells in the aneurysm wall. The chemotherapy against MALT lymphoma was highly effective, so we presumed that the lymphoma cells had disappeared. The source of the distal MCA aneurysms was thought to be oncotic. Distal MCA aneurysms caused by MALT lymphoma are extremely rare. Surgical reconstruction using the STA may be effective in patients with ruptured distal MCA aneurysms if the wall is intact.

Key words: distal middle cerebral artery aneurysm, reconstruction, intracerebral hematoma, superficial temporal artery, mucosa-associated lymphoid tissue lymphoma

Introduction

Distal cerebral aneurysms tend to arise in the anterior circulation and the etiology includes mitotic, oncotic, and traumatic causes. Consequently, the natural history and treatment of these aneurysms differ. The incidence of central nervous system lymphoma is relatively low, and the reported rate of secondary brain lymphoma is 6.1%.⁷⁾ The most common neuropathological findings in patients with metastasis are infiltration of the leptomeninges, nerve roots, perivascular space, or brain parenchyma, which can result in both focal and generalized symptoms.^{6,10)}

We report an extremely rare case of ruptured aneurysms on the distal middle cerebral artery (MCA) that may be attributable to mucosa-associated lymphoid tissue (MALT) lymphoma. The patient was successfully treated with vasoconstructive surgery using a superficial temporal artery (STA) graft.

Case Report

A 60-year-old man had been diagnosed with MALT lymphoma (stage IV) of the lung 6 years previously.

Chemotherapy resulted in complete remission. Three months before visiting our internal medicine department, he noticed weight loss and fever. He again received chemotherapy under a diagnosis of recurrent MALT lymphoma of the lung. One week later, he suffered sudden onset of motor aphasia, Gerstmann syndrome, and incomplete right hemiparesis, and was referred to our department.

Computed tomography showed a subcortical hematoma in the left parieto-temporal region (Fig. 1A). Magnetic resonance (MR) imaging demonstrated the hematoma and 2 thrombosed aneurysms adjacent to the hematoma (Fig. 1B). Left internal carotid angiography revealed 2 distal saccular aneurysms of the posterior parietal artery and angular artery (Fig. 2A, B). As the patient had myelosuppression due to chemotherapy, we placed him under observation. Two weeks later he had recovered from myelosuppression, and angiography at this time showed that the aneurysm on the left posterior parietal artery had disappeared, but the aneurysm on the angular artery had tripled in size (Fig. 2C, D).

He underwent aneurysmectomy and vessel reconstruction using an STA graft on the next day. After incision of the dura, the arachnoid was found to be thick and whitish, and the cortex adjacent to the aneurysm on the angular ar-

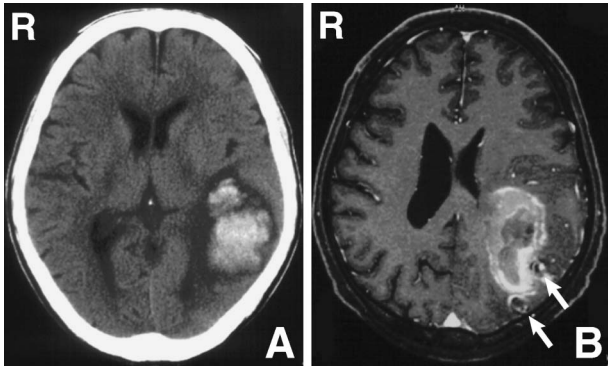


Fig. 1 A: Computed tomography scan demonstrating a subcortical hematoma in the left parieto-temporal lobe. B: T₁-weighted magnetic resonance image with gadolinium showing the hematoma and 2 adjacent thrombosed aneurysms (arrows).

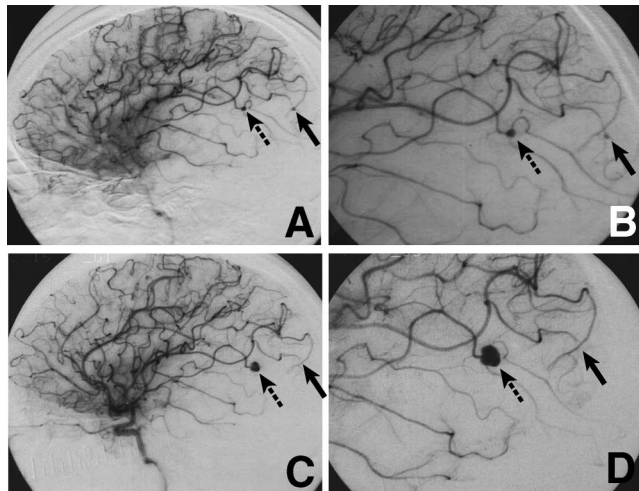


Fig. 2 A, B: Initial left cerebral angiograms, lateral view (A) and the higher magnification (B), revealing 2 saccular aneurysms in the posterior parietal artery (solid arrows) and angular artery (dotted arrows). C, D: Repeat left cerebral angiograms obtained 2 weeks after the first, lateral view (C) and the higher magnification (D), showing the aneurysm on the angular artery 3 times larger than at the first angiographic study (dotted arrows). The aneurysm on the posterior parietal artery was thrombosed (solid arrows).

tery was reddish (Fig. 3A). The proximal and 2 distal parent arteries of the aneurysm were identified (Fig. 3B). After establishing an end-to-end bypass from the proximal to larger distal parent artery (1-mm diameter) using an STA graft, the aneurysm was excised and a side-to-end bypass placed from the angular artery to its smaller (0.5-mm diameter) distal parent artery using an STA graft. Blood flow Doppler study confirmed good flow through the end-to-end graft (Fig. 3C).

Postoperative clinical course was uneventful and his neurological deficit improved. Postoperative angiography revealed excellent patency of the larger distal parent artery graft, but no blood flow through the smaller graft was

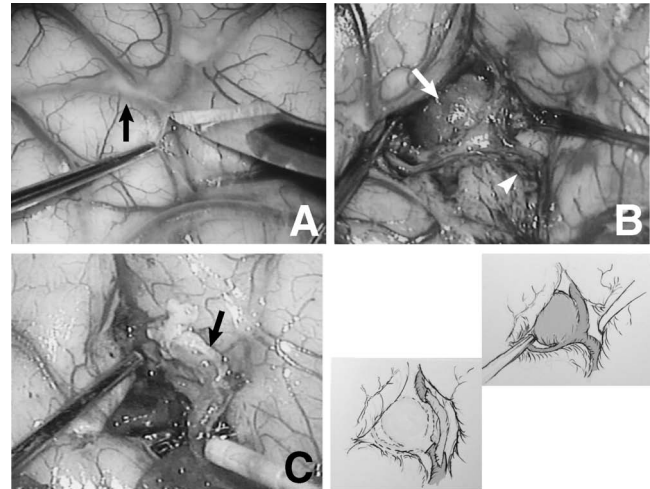


Fig. 3 Intraoperative photographs showing muddy arachnoid (arrow) and reddish cortex adjacent to the aneurysm on the angular artery (A), the angular artery aneurysm (arrow) and its parent artery (arrowhead) detected after incision of the muddy arachnoid (B), and arterial graft using the superficial temporal artery (arrow) forming an end-to-end bypass from the proximal parent artery to the largest distal parent artery (C).

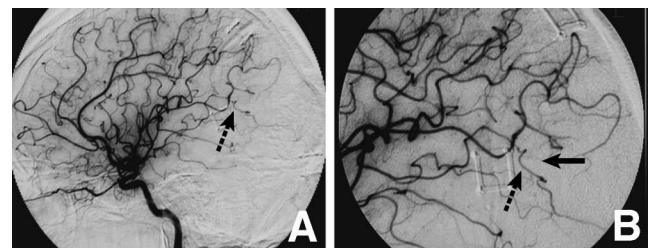


Fig. 4 Left cerebral angiograms obtained 10 days after the operation, lateral view (A) and the higher magnification (B), showing excellent patency of the reconstructed angular artery (dotted arrows) and absence of blood flow in the reconstructed posterior parietal artery (solid arrow).

recognized (Fig. 4). He was moved to the internal medicine department in our hospital where he underwent 4 courses of chemotherapy for MALT lymphoma of the lung. Histological examination of the thin arachnoid showed collections of necrotic and denatured tumor cell-like scars (Fig. 5A), with structural retention of the parent artery, but the aneurysm wall was not infiltrated by inflammatory cells (Fig. 5B). The adventitia and thin intima of the artery contained local collections of lymphocytes (Fig. 5B), but immunohistochemistry revealed no expression of CD79a or CD20, both of which are B-cell markers. Various stains excluded the presence of bacteria.

Discussion

Distal aneurysms secondary to oncotic embolization invading and destroying the arterial wall are rare, and most notably involve choriocarcinoma.⁴⁾ In our case, the

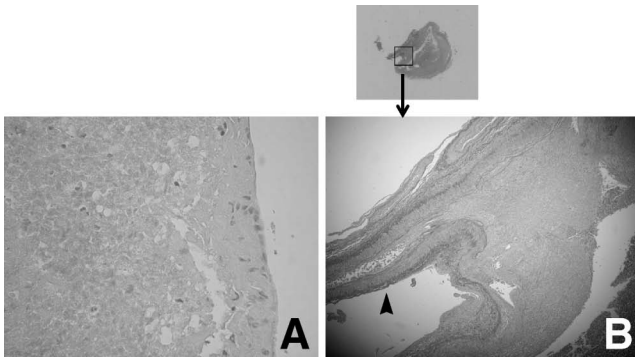


Fig. 5 Photomicrographs showing collections of necrotic and denatured tumor cell-like scars in the thin arachnoid (A: hematoxylin and eosin stain, $\times 200$); and no infiltration of inflammatory cells into the aneurysm wall, local collection of lymphocytes in the adventitia and thin intima of the artery, and structural retention of the parent artery (arrowhead) (B: elastica-van Gieson stain, $\times 50$).

arachnoid contained collections of necrotic and denatured tumor cell-like scars, but no malignant cells in the aneurysm wall or the arachnoid. Although the lymphocytes in the adventitia and intima of the artery did not express CD79 or CD20 immunoreactivity, we thought that the aneurysms were oncotic secondary to metastasis from the MALT lymphoma. Although we could not exclude other factors, we speculate that the aneurysms harbored no malignant cells because of the effective chemotherapy against the tumor.

Another patient with intravascular large B-cell lymphoma presented with a ruptured distal MCA aneurysm filled with tumor cells, but secondary brain involvement by MALT lymphoma is extremely rare.^{1,2} How secondary MALT lymphoma induced the distal aneurysm is unclear. Metastasis from lymphoma appears to spread through four routes: direct infiltration from an adjacent primary site, spread to the bone marrow followed by direct extension through perforating vessels into the dura and leptomeninges, hematogenous dissemination from a remote site, or spread via lymphatic channels that directly connect any 2 lymph node chains.^{3,5,6} Although we did not examine the cerebrospinal fluid of our patient, the collections of necrotic and denatured tumor cell-like scars in the arachnoid, and the absence of tumor cells in the aneurysm wall suggest that these aneurysms developed via intrathecal dissemination.

Distal MCA aneurysms have been treated by surgical excision, interventional embolization, clipping, ligation, and reconstruction. We planned excision and reconstruction of the arteries using an STA graft. Although veins are more commonly used as grafts for cerebral vessel reconstruction, the STA can be used if its caliber is sufficiently large.^{8,9} The patency of the STA graft we used for reconstruction after excising the oncotic aneurysms was good, the structure of the parent artery close to the aneurysm was retained, and the distal vascular walls were completely normal. In this case, second angiography showed that the aneurysm on the angular artery had tripled in size. The

cause of the expansion is not clear but MR imaging should be helpful to distinguish between the increase in aneurysm diameter and the expansion of the lumen in the aneurysm resulting from thrombus reduction.

In conclusion, a distal MCA aneurysm that developed after the delivery of chemotherapy in a patient with MALT lymphoma was successfully treated by vasoconstructive surgery using the STA. The treatment of intracranial distal MCA aneurysms that may be attributable to disseminated malignant tumor must consider both aneurysm excision and retention of the blood flow in the parent artery to preserve its normal structure and to ensure patency of the graft. Moreover, such aneurysms should be treated as soon as possible, because distal aneurysms of oncotic origin may enlarge in the course of weeks even if the chemotherapy for the tumor is effective.

Conflicts of Interest

None.

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