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LETTER TO EDITOR PEER REVIEWED | OPEN ACCESS

A case of transient bilateral medial longitudinal fasciculus syndrome, accompanied with the coil embolization to the basilar superior cerebellum artery aneurysm

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To the Editors,

Here I report a rare case of transient bilateral medial longitudinal fasciculus (MLF) syndrome associated with the endovascular aneurysm embolization. A 57-year-old man had an unruptured aneurysm at the bifurcation of the basilar superior cerebellar artery. Percutaneous endovascular embolization of the aneurysm with coils was performed two years ago. Due to recanalization of the aneurysm neck (Figure 1A), additional endovascular embolization was performed via right femoral artery approach. A microcatheter (Excelsior SL-10; Stryker, Kalamazoo, MI, USA) was engaged in the aneurysm through the left basilar artery under the guidance of a microguidewire (Transcend EX Platinum; Boston Scientific, Marlboro, MA, USA). Coil (Target Helical Φ3 mm × 4 cm, Φ2 mm × 3 cm, Φ2 mm × 2 cm, Φ2 mm × 2 cm, Φ1.5 mm × 2 cm; Stryker) embolization was completed without stent or balloon support (Figure 1B). After surgery, the patient had double vision with bilateral ocular adduction defects and abduction of the left eye. Magnetic resonance imaging (MRI) of the head showed bilateral hyperintense lesions around the midbrain aqueduct on diffusion-weighted imaging (DWI) (Figure 1C–F). He was diagnosed with bilateral MLF syndrome [1]. For treatment of ischemic complication, argatroban [2, 3] and edaravone [4, 5] were infused intravenously. Eye movement disorders disappeared within one day. No recurrence of ocular hypotension was observed.

During the procedure, the microguidewire and microcatheter never migrated into the perforator of the basilar artery. Delivery of the microsystem around the aneurysm was also smooth, and no ledge or jumping of the microsystem was observed. The transient ischemic attack was thought to be caused by the temporary occlusion of the orifices of the para-median perforators by the microatheroma spread over them. Their incomplete occlusion may have contributed to the early reperfusion

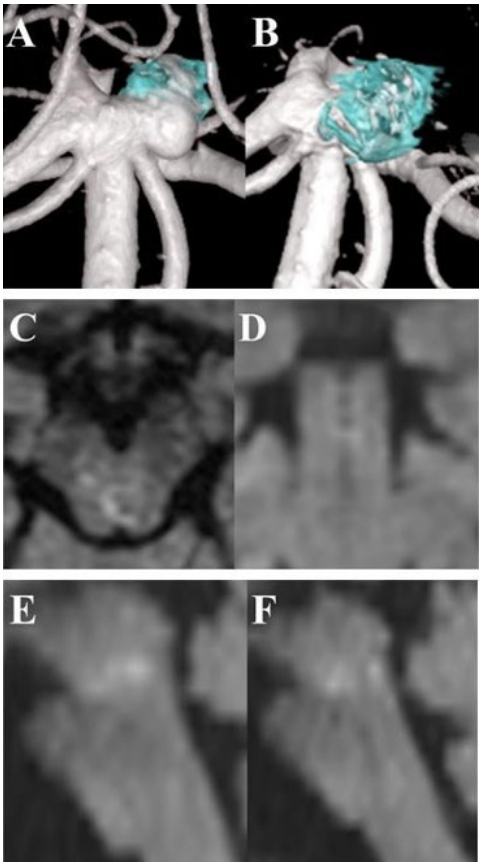

Figure 1: Three-dimensional left vertebral artery angiography (lt. VAG) of the right basilar superior cerebellar artery (rt.BA-SCA) aneurysm before (A) and after (B) endovascular intervention. The posterior view shows the recanalized aneurysmal neck between two years after coil embolization (A) and the packed aneurysmal neck with coils (B). The head magnetic resonance imaging (MRI) of the volume diffusion-weighted image (DWI) after coil embolization. Bilateral ischemic change is observed in the dorsal area of the midbrain by axial view (C). The ischemic lesion is distributed in the para-median area in the coronal view (D). Sagittal view of right (E) and left (F) show the DWI hyper spots, near the aqueduct.

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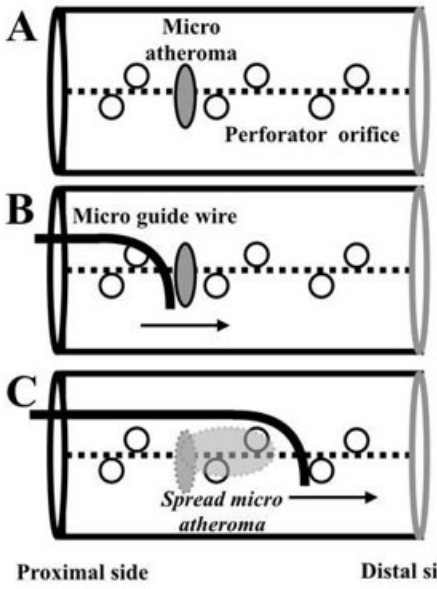

Figure 2: (A) Diagram of the basilar artery (BA). The dotted line shows the middle part of the BA. Small circles represent the orifices of the paramedian perforators. Gray circle represents the microatheroma on the wall of the BA. Blood flow is from left (proximal) to right (distal) side on the scheme. (B) Advanced microguidewire (MGW) direction (arrow) and portion (black J-shaped feature) are drawn. (C) Ischemic mechanism through the expanding microatheroma. The MGW slides on the wall of the BA with the spread of the microatheroma, which covers the perforator orifices as a thin film. Light gray area, expanded from gray circle (means the microatheroma), represents atheroma spreading along the luminal wall. The expanded light gray area (microatheroma) overlaps the small circle (BA branch orifice), which represents the speculated state of BA branch occlusion by thin atheroma.

of the midbrain without permanent deficits. It was known that microatheromas on the vessel wall sometimes detach and migrate distally due to the mechanical stress of the microcatheter. Different from the distal migration, the present case showed the ischemic change on the way to the aneurysm access. This unusual ischemic phenomenon might be caused by the spreading microatheroma forming a thin film over the perforator orifice (Figure 2). To avoid perforation or laceration to the vessel wall, the tip of the microguidewire was usually shaped to J-curve. During the microcatheter delivery, the J-shaped microguidewire tip proceeds to the vessel with sliding its wall. As the tip of the microguidewire moved, the microatheroma might spread along the wall like butter on bread. I would like to propose this phenomenon and call it “spread butter effect.”

In the field of endovascular intervention, distal thrombosis has been well documented with discussion of the mechanism and the prevention strategies. The main causes are embolus dispersal associated with interventional devices induced dislodgement of atheromatous thrombus. However, the ischemic complication is rarely observed in the access root perfusion area. Intracerebral stenting in the middle cerebral artery (MCA) or basilar artery (BA) sometimes leads to the ischemic deficit in the perforator region by the snowplow effect [6, 7]. This is caused by the displacement of microatheroma over the orifice of the MCA perforators during the stent expansion [8, 9]. In contrast, no intravascular stent or balloon was used in the present case. Microcatheter and microguidewire went through the basilar trunk to deliver the system. From the ischemic lesion area and the surgical manipulation method, this transient MLF syndrome was suggested to be the result of the microatheroma spread over the orifices of the para-median perforators by the advancing microguidewire. Figure 2 shows the hypothesized mechanism of the ischemic event. Other possibilities of ischemic changes were considered. The possibility of direct occlusion of the perforator origin by the microcatheter during the coil embolization was excluded. However, the intervention time was not long enough to induce an ischemic stroke, which was excluded. Micro-plaque rupture on the way from the vertebral artery to the basilar artery during the intervention was not excluded because no severe atheromatous changes were found before the procedure. Spasm of the perforator origin caused by the scratching stress of the microcatheter was speculated as the cause of the transit ischemic event. It is difficult to visualize or confirm the morphologic change of perforators due to spasm. This hypothesis is still under discussion.

The main cause of bilateral MLF is multiple sclerosis [1, 10]. Tumor, infectious disease including encephalitis and stroke may cause bilateral MLF [1, 10]. Occlusion of basilar artery branch by atheroma is known to cause brain stem syndrome including MLF [11, 12]. However, transient ischemic changes are rare manner in bilateral MLF. In the present case, the transient ischemic defect originated from the para-median perforator of the basilar artery to the mid brain. Because of the manipulated aneurysm existed to the distal basilar artery, microguidewire might scratch the upper basilar intima frequently during the engagement of microsystem into the aneurysm. This mechanical sweep could spread microatheroma across the perforator orifices.

In the present case, the mechanism of ischemia has never been proved exactly. The proposed transient ischemic mode is still under discussion. The dynamic change of microperfusion through brain stem perforator is difficult to analyze. In addition, it is difficult to detect the minute length of microatheroma shift or spread on the arterial intima in a clinical manner. Basic research would be needed to estimate the microatheroma dynamics by microguidewire stress. Further accumulation of clinical knowledge and experience of similar stroke should be needed to establish the “spread butter effect” hypothesis. It is necessary to continue the careful neurological adverse event control and radiological image argumentation on the catheter intervention cases to verify this hypothesis.

Keywords: Basilar artery aneurysm, Catheter intervention, MLF syndrome, Transient bilateral ischemia

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Author Contributions
Akira Tempaku – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Data Availability
All relevant data are within the paper and its Supporting Information files.

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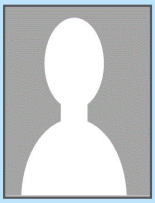

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