Case Report

Paradoxical Brain Embolism Caused by Isolated Pulmonary Arteriovenous Fistula Successfully Treated with Recombinant Tissue Plasminogen Activator

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Pulmonary arteriovenous fistula (PAVF), a vessel malformation connecting the pulmonary circulation to the systemic circulation while bypassing the pulmonary capillaries, can cause paradoxical cerebral infarction. It is often associated with hereditary hemorrhagic telangiectasia (HHT), a genetic disease characterized by multiple dermal, mucosal, and visceral telangiectasia causing recurrent bleeding. Paradoxical cerebral embolism caused by PAVF without HHT is rare. Here, we report a patient with isolated PAVF who experienced an ischemic stroke caused by a paradoxical embolism from deep venous thrombosis; the patient was successfully treated with recombinant tissue plasminogen activator. She presented with a decrease in arterial oxygen saturation to 91%, and lung disease was suspected. A PAVF was subsequently found in the right S6 region using contrast computed tomography. Interventional radiologists successfully occluded the shunt using 6 microcoils. PAVF should be considered when determining the pathogenesis of cerebral ischemia in patients with hypoxia, which can be the only symptom of PAVF.

Key Words: Paradoxical embolism—pulmonary arteriovenous fistula—alteplase—coil embolization

Case Report

An 84-year old female with no family history or symptoms of hereditary hemorrhagic telangiectasia was hospitalized in a psychiatric department for the treatment of schizophrenia. One morning, she exhibited a consciousness disturbance, and a neurologic examination revealed motor aphasia, right tactile extinction, left conjugate gaze, and right hemiplegia. Her National Institutes of Health Stroke Scale (NIHSS) score was 22 points. She did not exhibit any physical abnormality except for a decrease in arterial oxygen saturation to as low as 91%. Laboratory findings showed an elevated D - dimer level (12.5 μg/mL) on a coagulation test and an elevated BNP level (22.9 pg/mL) on a blood chemistry examination. The levels of antithrombin-Ⅲ, protein C, and protein S were within the normal limits. Diffusion-weighted magnetic resonance images showed a hyperintense cortical lesion in the left middle cerebral artery territory (Fig. 1A). Magnetic resonance angiography showed a left middle cerebral artery occlusion in the M1 proximal portion (Fig. 1B). We started recombinant tissue plasminogen activator (rt-PA) treatment at 256 minutes after the onset of symptoms. Her paralysis improved 15 minutes after the start of treatment, although the aphasia and tactile extinction persisted. Her NIHSS score improved to 13 points and cerebral angiography at 364 minutes after the onset confirmed the recanalization of left M1 occlusion. A pulmonary arteriovenous fistula (PAVF) was found in the right lower lobe using contrast computed tomography, which was performed

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Received January 9, 2019; revision received March 12, 2019; accepted March 24, 2019.

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1052-3057/$ - see front matter

https://doi.org/10.1016/j.jstrokecerebrovasdis.2019.03.048

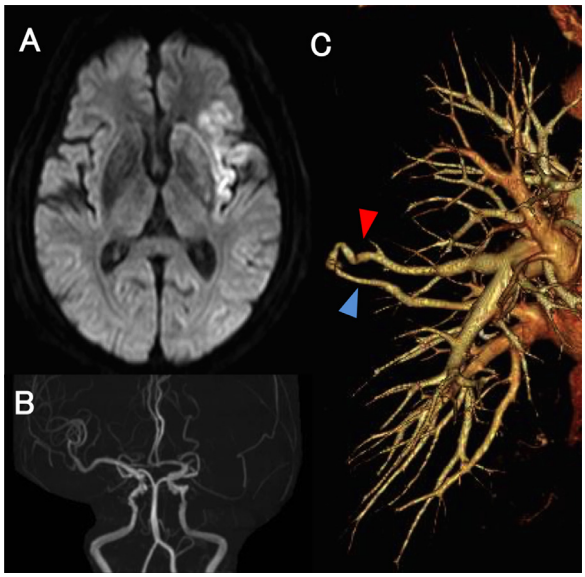

Figure 1. *Cranial magnetic resonance imaging (MRI) and pulmonary computed tomography (CT) angiography. (A) A cranial MRI examination performed at the onset of symptoms using diffusion-weighted imaging (DWI) showed a high signal intensity in the left middle cerebral artery area. The DWI Alberta Stroke Program Early CT Score (ASPECTS) was 6 out of 11 points. (B) Head MR angiography performed at the onset of symptoms revealed the complete occlusion of the left middle cerebral artery in the M1 proximal portion. (C) Pulmonary CT angiography showed PAVF in the right lower lobe S6 region. An inflow artery (red arrowhead) and a drainage vein (blue arrowhead) were visible. The diameters of the the minimum feeding artery, nidus, and drainage vein were about 4.3 mm, 3.8 mm, and 4.3 mm, respectively. (Color version of figure is available online.)*

because of the pulse oxygen desaturation (Fig. 1C). In addition, a contrast defect suggesting a thrombus was observed in a deep vein in the left lower leg. Intravascular coil embolization using 6 microcoils was successfully performed for the shunt 17 days after the onset of symptoms. After the intravascular treatment, we began administering 30 mg/day of edoxaban for the treatment of deep venous thrombosis. She was returned to the psychiatric ward 36 days after onset without the recurrence of cerebral infarction.

Discussion

Paradoxical cerebral embolism caused by PAVF without hereditary hemorrhagic telangiectasia is rare, accounting for 0.5% of all cerebral infarctions.[1] Treatment with rt-PA was effective in this case; to the best of our knowledge, only 1 previous report has described the use of rt-PA for paradoxical brain embolism in a patient with PAVF.[2] In that case as well, thrombolysis using rt-PA was effective and the patient was discharged without any neurological symptoms, although he did experience another episode of cerebral ischemia caused by Trousseau's syndrome 1 month later. One reason for the effectiveness of rt-PA was suggested to be that the thrombi associated with paroxysmal emboli caused by PVAF are smaller than those in cases with cardiogenic cerebral ischemia, since the thrombi must be able to cross the relatively small diameter of the nidus of the pulmonary vascular abnormality. PAVF should be considered when determining the pathogenesis of cerebral ischemia in patients with hypoxia, which can sometimes be the only symptom of PAVF.[3]

References

1. Kimura K, Minematsu K, Nakajima M. Isolated pulmonary arteriovenous fistula without Rendu-Osler-Weber disease as a cause of cryptogenic stroke. J Neurol Neurosurg Psychiatry 2004;75:311-313.
2. Sakamoto Y, Mitsumura H, Nakata M, et al. A case of Trousseau's syndrome and pulmonary arteriovenous fistula: a malignant combination for ischemic stroke. Neurol Sci 2015;36:1035-1036.
3. Pick A, Deschamps C, Stanson AW. Pulmonary arteriovenous fistula: presentation, diagnosis, and treatment. World J Surg 1999;23:1118-1122.