A Small Pulmonary Arteriovenous Malformation as a Cause of Recurrent Brain Embolism

Kenichi Todo, Hiroshi Moriwaki, Masahiro Higashi, Kohji Kimura and Hiroaki Naritomi

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Summary: We report a case of recurrent paradoxical brain embolism mediated through a small pulmonary arteriovenous malformation (PAVM) with a 1.8-mm-diameter feeding artery. In this case, the further recurrent stroke was prevented successfully by PAVM embolization. Although embolization therapy is currently recommended only for PAVMs with feeding arteries greater than 3 mm in diameter, the therapy may be needed also in the smaller PAVMs.

Pulmonary arteriovenous malformation (PAVM) is well recognized as a cause of paradoxical brain embolism. Brain infarction associated with PAVMs most likely occurs in patients with feeding arteries of more than 3-mm diameter and not in those of smaller size (1–5). We report the case of a PAVM patient with a 1.8-mm-diameter feeding artery who had recurrent paradoxical brain embolism and was successfully treated with embolotherapy.

Case Report

A 67-year-old right-handed woman was admitted in February 2002 after suddenly developing dizziness, nausea, and speech disturbance. She was admitted to our hospital 7 hours after the onset of symptoms. At the time of admission, her blood pressure was 140/80 mm Hg. She had a regular heart rate of 60 beats per minute and a respiratory rate of 16 breaths per minute. Neither pathologic breath sounds nor heart murmurs were auscultated. No edema was present in her extremities. No cutaneous vascular malformations were observed. She had no episodes of hemoptysis or dyspnea. In neurologic examinations, she had dysarthria and left limb ataxia. Laboratory findings showed normal blood cell counts, normal liver and renal functions, and normal serum cholesterol and blood glucose levels. Plasma protein C, protein S, and antithrombin III levels were within normal ranges. Arterial blood gas analysis showed mild hypoxemia, with PaO2 84.6 mm Hg and PaCO2 43.2 mm Hg in room air. Chest radiographic and electrocardiographic findings showed no stenotic lesion in the extra- or intracranial vertebral arteries or basilar artery. With suspicion of embolic brain infarction, anticoagulation therapy with 10,000 U/day heparin was started 18 hours after the onset of symptoms, although the embolic source could not be identified.

On day 3, the patient also developed right limb ataxia. Activated partial thromboplastin time was 28.0 seconds on admission and was prolonged to 43.1 second on day 3. The second CT scan showed only a hypoattenuated area in the left cerebellar hemisphere. Diffusion-weighted imaging on day 5 confirmed the development of new ischemic lesion in the right cerebellar hemisphere (Fig 1A). Twelve hours after the onset of stroke, intraarterial digital subtraction cerebral angiography revealed no stenotic lesion in the extra- or intracranial vertebral arteries or basilar artery. With suspicion of embolic brain infarction, anticoagulation therapy with 10,000 U/day heparin was started.

Discussion

PAVMs are commonly associated in hereditary hemorrhagic telangiectasia (HHT): approximately
15–35% of HHT patients have PAVMs, and 50–85% of PAVMs patients have HHT (1). The present patient, however, had no clinical signs suggestive of HHT. PAVMs increase the risk of paradoxical embolism because of continuous right-to-left shunt. Brain infarction is one of the most important complications that may occur in PAVM patients (1, 2). Stroke is often the first manifestation of PAVM and even of HHT (7).

Moussouttas et al (1) documented that brain infarction most likely occurs in PAVM patients with feeding arteries more than 3 mm in diameter and not in those with smaller size. Accordingly, they recommended embolization therapy for all PAVMs with feeding artery diameters exceeding 3 mm. To the best of our knowledge, the occurrence of brain infarction in PAVMs with less than 3 mm in feeding artery diameter has not been previously reported. The present patient had a small-sized PAVM, the diameter of which was 1.8 mm. In the present patient, the other embolic sources—including atrial fibrillation or flutter, bacterial endocarditis, valvular pathology, myocardial infarction, dilated cardiomyopathy, akinetic left ventricular segment, intracardiac thrombus, atrial myxoma, spontaneous echo contrast of the left atrium, or complex atheroma of the aortic arch—were ruled out, and other causes of stroke—such as atherosclerotic arterial changes, nonatherosclerotic arteriopathies, coagulopathies, hematologic or systemic disorders, or migrainous infarction—were found to be negative (8, 9). The patient had deep vein thrombosis, which may cause arterial embolism under the presence of right-to-left shunt. Thus, paradoxical embolism is only one possible mechanism that can explain recurrent strokes in this patient. After the embolization therapy, this patient had no more recurrent stroke, and C-TCD never detected HITS, indicating successful obstruction of right-to-left shunt (10).

A recent study reported that the stroke recurrence rate in patients with patent foramen ovale (PFO) was unaltered regardless of whether the diameter was more than 2 mm or less (11). Paradoxically, PAVMs
and PFO share the similar mechanisms in the development of brain infarction through the right-to-left shunt. The shunt flow goes through PAVMs continuously, whereas it can pass through PFO only intermittently at the time of right atrial pressure increase. In this context, PAVM is regarded to be more risky than PFO as a route of paradoxical embolism. The size of right-to-left shunt may not be a critical factor for stroke occurrence. Embolization therapy appears to be useful for preventing recurrent attacks in stroke patients with a small PAVM in whom other causes of cerebral infarction are definitely ruled out. A randomized controlled study may be necessary to elucidate the requirement of embolization therapy in small-sized PAVMs.

References