Paradoxical embolism is a risk factor for cryptogenic stroke in young adults, and can be primarily attributed to the presence of patent foramen ovale (PFO). Although contrast echocardiography may facilitate diagnosis, it would be difficult to distinguish a PFO from a pulmonary arteriovenous fistula (PAVF), which could also result in paradoxical embolism. We report a 46-year-old woman with recurrent ischemic stroke, who was scheduled for PFO closure because of a right-to-left shunt detected by contrast echocardiography. The diagnosis turned out to be a PAVF confirmed by angiography. Thereafter, coil embolization of this PAVF was performed successfully.

Key Words: Embolic stroke • Patent foramen ovale • Pulmonary arteriovenous fistula • Right-to-left shunt

INTRODUCTION

Patent foramen ovale (PFO) with right-to-left shunting is a well-known cause of cryptogenic stroke in relatively young patients. It has been demonstrated that contrast echocardiography may be helpful in the diagnosis of PFO. However, there are still some uncertainties about the exact etiology of right-to-left shunt even in the presence of positive contrast echocardiographic result. We report a young woman with recurrent ischemic stroke who was scheduled for PFO closure because of a right-to-left shunt detected by contrast echocardiography. The diagnosis turned out to be a pulmonary arteriovenous fistula (PAVF) after extensive imaging studies using transesophageal echocardiography and angiography.

CASE REPORT

A 46-year-old woman experienced two episodes of ischemic stroke within 3 years. She had no history of diabetes mellitus, hypertension, or coronary artery disease. Multifoci involvement revealed by brain magnetic resonance imaging suggested that the cause of acute infarcts was embolic in origin. Transthoracic echocardiography failed to demonstrate definite intracardiac shunting; however, PFO was suspected based on the appearance of contrast echoes within the left atrium by contrast echocardiography. The patient was referred to our hospital for transcatheter closure of the presumed PFO.

On admission, the patient was in good clinical condition except for persistent loss of right-sided vision. The oxygen saturation measured by pulse oximeter was 96%. On physical examination, her heart beats were regular, and the cardiac and chest auscultation revealed unremarkable findings. There was no muscle weakness or finger clubbing. Right-sided cardiac catheterization was performed under general anesthesia, and endotracheal intubation was achieved in preparation for planned PFO closure. However, no intratrial communication was found, either by repeated probing using a multipurpose catheter or by transesophageal echocar-
After the injection of agitated saline, bubbles were noted in the left atrium after 3 cardiac cycles, but there was no passage of contrast echoes through the location of foramen ovale. Therefore, a right-to-left shunt of extracardiac origin was suspected. We then performed a selective right pulmonary arterial angiography, which revealed an isolated PAVF in the right lower pulmonary artery, with a direct connection to the left atrium (Figure 1A). The narrowest portion of this PAVF was around 3 mm in diameter. This PAVF could not be demonstrated on chest x-ray (Figure 2A). Coil embolization (Cook Inc. Embolization Coil, MWCE 38-8-5) of the PAVF was performed successfully (the loop diameter of this coil was 5 mm; Figure 1B and 2B). The patient was discharged the next day. She did not have clinical manifestations suggestive of hereditary hemorhagic telangiectasia expected for pulmonary AVF. Subsequently, there was no recurrence of stroke noted at the 2 year follow-up.

**DISCUSSION**

Cardioembolic stroke is responsible for about 20% of all ischemic stroke cases. The thrombi from the left heart and arterial system can directly cause an embolic stroke. Another possible cause of embolic stroke is paradoxical embolism which results from thrombi entering the systemic arterial circulation from the venous system. This pathologic event is possible only when thrombosis of the venous system as well as a right-to-left shunt exist at the same time. The most common cause of right-to-left shunting in adult patients is PFO, which could be detected by contrast echocardiography with the presence of bubbles within the left atrium. However, our present case suggested that PAVF is another structural abnormality which could lead to paradoxical embolism. Previous studies have demonstrated that 0.5% of patients with cryptogenic stroke had PAVFs. In patients with positive contrast echocardiographic results but no evidence of interatrial communication, PAVF should be immediately considered. The timing of left heart contrast entry during saline contrast echocardiography has been proposed to distinguish a PFO from a PAVF. In general, the “three-beat rule” is used to distinguish the intracardiac from extracardiac shunts. Our experience with this patient supported the diagnostic implication of this rule in cases with positive contrast echocardiographic results.

Pulmonary arteriovenous fistula is a rare vascular malformation. The overall incidence is estimated to be approximately 1–3 per 100,000 population. It is estimated that about 70–90% of individuals with PAVFs have hereditary hemorrhagic telangiectasia (HHT), and about 50% of individuals with HHT have PAVFs. Non-HHT-related PAVFs, as found in this patient, are mostly sporadic, or secondary to hepato-pulmonary syndrome, caval pulmonary shunts, or trauma. Pulmonary arteriovenous fistulas tend to increase in size over time, and rarely regress spontaneously. Stroke and brain abscess are common complications of PAVFs due to paradoxical embolism. In one study of 93 consecutive patients with PAVFs, the incidence of symptomatic stroke and brain abscess was 18% and 5%, respectively.

Percutaneous embolization therapy has been the most commonly used treatment of PAVFs since the 1980s. Current devices used for arteriovenous fistula...
embolization include coils, detachable balloons, and Amplatzer vascular plugs or duct occluders.\textsuperscript{7,9,10} The choice of embolization devices to be employed is not just based on operator preference, but also depends on the size, location, and accessibility of the PAVF.\textsuperscript{9}

In summary, PAVF should be considered as a possible etiology of cryptogenic stroke in patients with evidence of right-to-left shunt, but manifesting a lack of interatrial communication. As long as diligent clinicians retain a high index of suspicion, this could routinely lead to correct diagnosis and prompt treatment.

REFERENCES