Outcomes of Coil Embolization for Pulmonary Arteriovenous Fistula as Evaluated Using a Novel Soft Ultrasound Probe Pasted to the Neck

Hidetaka Mitsumura¹, Ayumi Arai², Kenichiro Sakai¹, Yuka Terasawa¹ and Jun Kubota³

Abstract:
An 81-year-old woman presented to our emergency room by ambulance with gait disturbance and pain in her left neck. Magnetic resonance imaging (MRI) showed acute left-sided dorsolateral medullary infarction and an occluded left vertebral artery. The temporal bone echo window was insufficient, but our pastable soft ultrasound probe attached to the cervix (PSUP) detected many microembolic signals caused by the contrast agent (cMES) in the common carotid artery. Chest CT revealed right pulmonary arteriovenous fistula (PAVF) and she underwent coil embolization for PAVF considering the possibility of paradoxical embolism via PAVF. After embolization, the cMES disappeared on PSUP. Therefore, PSUP was useful for diagnosing and confirming the interventional procedural success for performing PAVF.

Key words: pulmonary arteriovenous fistula, contrast microembolic signal, transcranial Doppler, novel probe attached to the neck

(Intern Med Advance Publication) (DOI: 10.2169/internalmedicine.3788-19)

Introduction
Transcranial Doppler (TCD) ultrasonography with contrast agent can help to diagnose a right-to-left shunt (RLS), such as that caused by a patent foramen ovale (PFO), atrial septal defects, and pulmonary arteriovenous fistula (PVAF) (1). However, an RLS cannot be evaluated in elderly patients without a temporal bone window. Therefore, we developed a pastable soft ultrasound probe that easily attaches to the neck (PSUP; Figure 1A), to diagnose RLS because ultrasound can measure the blood flow in the carotid artery in most patients with stroke (2). The square, thin PSUP is modified for attachment to the neck and has the same properties as those of TCD (center frequency 2.0 MHz, pulse repetition frequency 5,000 Hz, sample volume 7 mm) (HDK-MB001, Hashimoto Electronic Industry Co. Ltd., Matsusaka, Japan) (Figure A-C) (2). PSUP attachment to the neck to examine the carotid artery is both stable and easy. Thus, when we would like to diagnose RLS using less invasive equipment, the PSUP has a major advantage in comparison with TCD and transesophageal echocardiography (TEE) (2).

We herein describe a patient with ischemic stroke caused by PAVF who underwent coil embolization using PSUP.

Case Report
An 81-year-old woman was admitted to the emergency room with gait disturbance and pain on the left side of the neck. She was taking medication for hypertension and dyslipidemia. She was started on treatment for peripheral vertigo, but her symptoms did not improve after two hours. Her blood pressure was 193/98 mmHg, ECG revealed sinus rhythm, and she was dyslipidemic. She had dysarthria, left Horner sign, left curtain sign, a loss of pain and temperature sensation on the left side of her face and the right side of her body, left limb ataxia, and a National Institute of Health Stroke Scale (NIHSS) score of 5. Magnetic resonance imaging (MRI) assessment revealed acute left sided dorsolateral
medullary infarction, occlusion of the left vertebral artery (VA) (Figure D and E), and an old cerebral infarction in the left posterior lobe suggesting embolism (Figure F). The bilateral middle cerebral arteries were not detected by TCD due to an insufficient echo window, therefore, we applied PSUP with a contrast agent at the right common carotid artery to detect RLS. Many microembolic signals caused by the contrast agent (cMES) were evident without performing the Valsalva maneuver (Figure G). Enhanced chest CT showed right isolated PAVF without deep venous thrombosis and pulmonary embolism (Figure H). Follow-up MRI at 14 days after onset showed the spontaneous recanalization of the left VA (Figure I). In-hospital electrocardiographic monitoring did not show atrial fibrillation. Moreover, no left atrial thrombus, aortic arch plaque or valve vegetation were observed in TEE. A probable paradoxical brain embolism was finally diagnosed, and the patient underwent coil embolization for PAVF at 19 days after onset. The cMES were undetectable on images obtained using the PSUP after embolization (Figure J) and the patient was transferred to a re-

Figure. Probe arrangement and imaging findings. A: PSUP with box-type attachment is smaller than the standard linear probe. B: Ultrasound system including PSUP and exclusive cart is as compact as transcranial Doppler. C: PSUP is easily fixed to the neck using surgical tape. D: A diffusion-weighted image shows acute cerebral infarction in the left dorsal lateral medulla (circle). E: Left vertebral artery occlusion on MRA (arrow). F: An old cerebral infarction of the left posterior lobe on FLAIR MR image (circle). G: Right CCA Doppler wave form on ultrasound using PSUP shows many cMES (arrow). H: Right isolated PAVF in enhanced chest CT (circle). I: Follow-up MR angiography shows spontaneous recanalization of the left vertebral artery (arrow). J: After coil embolization for PAVF, PSUP monitoring shows absence of cMES in the right CCA.
hability center at 30 days after onset.

**Discussion**

TEE with contrast agent and TCD is the standard procedure for evaluating PAVF, and TCD with contrast agent can confirm the presence or absence of PAVF after embolization (3). To our knowledge, this is the first report to describe cryptogenic stroke with PAVF evaluated at the carotid artery using PSUP. The advantages of PSUP are that the procedure is less invasive, simpler to perform than TEE, and it can be carried out in patients without a patent temporal bone window (2). Furthermore, the PSUP should be useful for monitoring before and after percutaneous PFO closure, for which future authorization in Japan expected in the near future. The disadvantage of single-channel PSUP is that it can evaluate the carotid artery only on one side. The device will require upgrading for bilateral monitoring.

Our patient underwent coil embolization to prevent recurrent ischemic stroke. According to the overseas guidelines, embolization is recommended for all patients with radiographically-visible PAVF (4). Notably, the incidence of ischemic stroke after embolization has significantly decreased (4). We selected catheter embolization to prevent ischemic stroke and brain abscess in our patient because she had already experienced two ischemic strokes, spontaneous recanalization indicating embolism, and no evidence of an embolic source other than that provided by PAVF (5).

In conclusion, PSUP is now expected to play an important role in diagnosing RLS and for confirming the appropriate intervention for PAVF.

**Author’s disclosure of potential Conflicts of Interest (COI).**
Yasuyuki Iguchi: Research funding, Takeda Pharmaceutical Company Limited, Otsuka Pharmaceutical Company Limited, Medtronic Japan Company Limited and Daiichi Sankyo Company Limited.

**References**

1. Chimowitz MI, Nemec JJ, Marwick TH, Lorig RJ, Furlan AJ, Salcedo EE. Transcranial Doppler ultrasound identifies patients with right-to-left cardiac or pulmonary shunts. Neurology 41: 1902-1904, 1991.
2. Mitsumura H, Arai A, Sato T, et al. A novel probe attached to the neck can accurately detect a large patent foramen ovale. J Neurol Sci 392: 122-125, 2018.
3. Kimura K, Minematsu K, Wada K, et al. Transcranial Doppler of a paradoxical brain embolism associated with a pulmonary arteriovenous fistula. AJNR American journal of neuroradiology 20: 1881-1884, 1999.
4. Shovlin CL, Condliffe R, Donaldson JW, Kiely DG, Wort SJ. British Thoracic Society Clinical Statement on Pulmonary Arteriovenous Malformations. Thorax 72: 1154-1163, 2017.
5. Swanson KL, Prakash UB, Stanson AW. Pulmonary arteriovenous fistulas: Mayo Clinic experience, 1982-1997. Mayo Clin Proc 74: 671-680, 1999.

The Internal Medicine is an Open Access journal distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (https://creativecommons.org/licenses/by-nc-nd/4.0/).