CASE REPORT



Thrombus rooting in the pulmonary arteriovenous fistula in a patient with cryptogenic stroke, a case report



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Abstract

Background Cryptogenic stroke constitutes approximately 40% of ischemic strokes in young adults, imposing a significant socioeconomic burden. However, the source of embolus has been specifically investigated in a few number of studies. In this report, We document the first case of a cryptogenic stroke associated with a pulmonary arteriovenous fistula (PAVF), providing evidence of mural thrombus formation within the PAVF vessel.

Case presentation A 52-year-old woman present with sudden-onset blurriness, right-sided numbness and paresthesia, speech difficulties, and salivation. It was confirmed as embolic stroke with magnetic resonance imaging. No evidence of cerebral arteriosclerosis or stenosis was identified, and cardiogenic embolism was ruled out. Further investigation was conducted to determine the cause of the anomalous embolism. A right-to-left shunt was detected using trans-cranial Doppler and transthoracic echocardiogram. Pulmonary arteriography revealed a simple PAVF. Optical coherence tomography (OCT) was used to examine the interior structure of the fistula vessel, revealing the presence of mural thrombus and a rough endangium.

Conclusions This case provides the initial evidence regarding the location of thrombus formation assessed and raises the awareness of a potential emboligenic mechanism in PAVF.

Keywords Cryptogenic stroke, Pulmonary arteriovenous fistula, Thrombus, Optical coherence tomography

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Background

Cryptogenic strokes are neither locular nor associated with identifiable source of embolism such as atrial fibrillation or atherosclerotic stenosis and account for approximately 40% of strokes in young adults [1]. As the underlying mechanisms is not clear, cryptogenic strokes pose challenges in identifying appropriate measures for secondary prevention. An abnormal right-to-left shunt is widely believed to be a potential threat of cryptogenic stroke. Pulmonary arteriovenous fistulas (PAVFs) are abnormal vascular communications directly connecting a pulmonary artery and a pulmonary vein, thereby functioning as a permanent right-to-left shunt and increasing the risk of paradoxical embolism [2]. Patients with PAVFs



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may develop neurological embolic complications, with a probability ranging from 9 to 41% [3]. However, the onset of thrombosis is rarely observed in clinical practice. Identifying the source of embolism is crucial in patients with PAVFs. Although deep venous thrombosis is widely accepted in context of paradoxical embolism, it has been suspected that thrombus formation can also occur within the fistula vessels. In this study, we reported an unusual case of embolization with ischemic stroke and investigated the source of thrombus with optical coherence tomography (OCT).

Case report

A 52-year-old female presented to the outpatient clinic with a chief complaint of repeated blurry vision and salivation, accompanied by dizziness. Five months ago, a sudden blurriness occurred when the patient was working part-time. Subsequently, the patient had right-sided numbness and paresthesia, speech difficulties, and salivation. The symptoms largely resolved within 30 min of lying down. She had no history of smoking or thrombotic vascular diseases. The patient underwent magnetic resonance imaging (MRI) that showed a cerebral infarcted lesion in the left parietal lobe and then was diagnosed with stroke. Magnetic resonance angiography ruled out prominent cerebral artery stenosis. Antithrombotic therapy was prescribed for the cerebral infarction. However, ischemic attacks recurred several times in the following months.

In physical exam the patient had normal vital signs. Auscultation of the chest revealed normal breath sounds without any crackles. Cardiac sounds were normal and no murmurs was detected. Electrocardiogram and Holter monitoring showed no abnormalities, including atrial fibrillation. Fundus stereo photography revealed no evidence of retinal artery embolism or bleeding. Color Page 2 of 4

duplex ultrasonography did not detect any severe atherosclerotic plaque in the major arteries or deep vein thrombosis in the lower limbs.

Based on the medical history of the patient, we tested the patient for vascular anomalies. Transcranial Doppler detected micro-embolic signals, indicating a right-to-left shunt (Fig. 1a). Transthoracic echocardiogram revealed positive bubbles in left atrium and ventricular even without the Valsalva maneuver (Fig. 1b). We performed atrial septum exploration in Catheterization Room, but the guidewire failed to pass through the atrial septum, indicating the absence of patent foramen ovale. Pulmonary arteriography revealed a simple PAVF in the right inferior pulmonary region (Fig. 2a) as the source of the rightto-left shunt. With the help of a 6 F coronary guiding catheter, an OCT was sent along with a 0.014 inch coronary guide wire to visualize the PAVF. To our surprise, multiple mural thrombi were observed adhering to the endangium surface (Fig. 2b), and a localized defect was detected on the endangium surface (Fig. 2c). Catheter embolization was performed, successfully occluding the angiographically visible abnormal vessel with an Abbott vascular plug II (Fig. 2d). Aspirin was prescribed as antithrombotic therapy. No bleeding events were observed during the six-month follow-up.

After 6-month follow-up, the patient demonstrated good recovery without new cerebral infarction or transient ischemic attacks. As with the patient's request, genetic testing for hereditary hemorrhagic telangiectasia was not performed.

Discussion

In this study, we presented a case that highlights the significance of vascular fistulas as a potential emboligenic provenance in patients with PAVF-associated strokes, as demonstrated by OCT.





Fig. 1 a Micro-embolic signals in trans-cranial Doppler examination. b Positive bubbles in transthoracic echocardiogram



Fig. 2 a Pulmonary arteriography showed an isolated pulmonary arteriovenous fistula. b Mural thrombi detected by OCT(arrows). c Endangic surface discontinuity under OCT detection(arrow). d Pulmonary arteriography of catheter embolization

PAVFs are largely responsible for neurological complications, including abscesses and ischemic damages [4], particularly in fistulas with a diameter of at least 2 mm [5]. In this case, the neurological symptoms were caused by an embolic stroke related to the PAVF, a rare condition supported by findings from retrospective studies [6]. It is commonly accepted that the passage of venous emboli through the fistula into the cerebral artery is the most likely etiology of cerebrovascular accident [3]. In this case, we use OCT to examine the interior structures of PAVFs and identified the potential source of thrombus. OCT revealed the presence of mural thrombi in fistula vessels, verifying that the thrombosis originated from these vessels. Slow hemorheology and the endangic roughness might be the significant cause of thrombosis. OCT has been used in the therapy of many cardiovascular diseases, particularly for optimizing percutaneous coronary intervention [7]. In recently years, OCT began to be used in cases of congenital heart disease. Ren et al. reported a 31-year-old male with confirmed PFO, in which several thrombi were detected in situ detected by OCT [8]. To our best knowledge, this is the first study to utlize OCT to unveil the interior structures of PAVFs.

Surgical resection and catheter embolization are the definitive treatments for PAVFs. Surgical resection may range from wedge resection to pneumonectomy [9]. Embolization is less invasive option with a high successful rate [10]. The complications of embolization include

pleuritic chest pain, pulmonary infection, air embolism and paradoxical embolism. There is no prospective study comparing the efficacy and safety of these appraoches. Surgery is usually preferred in cases where embolization is unsuccessful or not possible. In this case, we provided a safe transcatheter embolization option.

In summary, we report a case of PAVFs embolization in a patient with cryptogenic stroke and revealed in situ thrombosis in PAVFs. This has given us great insights into the fact that PVAFs should be considered as an important mechanism of ischemic stroke. Nevertheless, further studies are needed to confirm the necessity and efficacy of PAVFs screening in patients with cryptogenic stroke.

Abbreviations

MRIMagnetic resonance imagingOCTOptical coherence tomographyPAVFsPulmonary arteriovenous fistulasPFOPatent foreman ovale

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We thank the patient for giving consent to share patient information.

Author contributions

M.S. wrote the main manuscript. M S. and H.D. were involved in the management of the patient. W.L. was involved in the manuscript conception and design. X.M. was involved in the manuscript review and editing. All authors reviewed the manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

The study was approved by the ethics committee of Qilu Hospital Shandong University. Written informed consent was obtained from the patient's relatives to publish this case report and any accompanying images. Written informed consent was obtained from the patient to publish this Case report and any accompanying images.

Competing interests

The authors declare no competing interests.

Dual publication

The results/data/figures in this manuscript have not been published elsewhere, nor are they under consideration by another publisher.

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