Infarction of cerebellum vermis attributed to a right-to-left shunt by pulmonary arteriovenous malformation: a case report.

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Research Article

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Abstract

Objectives: We reported this case to remind neurologists that pulmonary right-to-left shunt is one of the causes of paradoxical embolism.

Methods: A 47-year-old woman was diagnosed with infarction of cerebellum vermis by MRI. Atheromatous, immunological, infectious and hypercoagulable abnormality was negated. A doubtful pulmonary arteriovenous malformation (PAVM) was accidentally found in the process of CT angiography of subclavian arteries.

Results: Then the pulmonary angiography confirmed the existence of PAVM in her left pulmonary lobe. Meanwhile, the bubble contrast study with transcranial Doppler found bubble signals passing her middle cerebral artery.

Discussion: Other than intracardiac, but pulmonary right-to-left shunt can induce embolic stroke.

Introduction

The pulmonary arteriovenous malformation (PAVM) is a thin-walled aneurismal sac, which connected the pulmonary artery and vein directly. Through this sac, the blood flows from the pulmonary artery to the vein without any screening and oxygenation. Potential complications include haemoptysis, cyanosis, dyspnea, stroke, and cerebral abscess. Patients with single or small PAVM may be asymptomatic, or have some of these symptoms only after intense physical activity. As reported, approximately 33% of patients with PAVM will have a history of stroke, 18% of transient ischaemic attacks. We report here a case of cerebellum vermis infarction attributed to a right-to-left shunt by pulmonary arteriovenous malformation.

Observation

A 47-year-old Chinese woman visited the neurology department complaining of continued dizziness and nausea for 22 hours after a hard work, which became aggravated when she rolled over axially. She presented with horizontal nystagmus when turning her eyes to the right. The brain magnetic resonance imaging demonstrated an isolated linear infarction lesion on the cerebellum vermis (Fig. 1). Besides the age over 45 years, no other cardiovascular risk factors were identified during hospitalization. No significant atheromatous lesions were found in intracranial arteries. The examination of thrombophilia and immunologic abnormalities was also negative. Yet, the woman had a history of cyanosis after intense physical activity or emotions. And she often felt short of breath when lying on the left side. Transthoracic echocardiography showed only a small amount of mitral regurgitation. Suggestion of transesophageal echocardiography was rejected. The no-contrast chest computed tomography (CT) revealed a pulmonary nodule of 2.3cm in diameter (Fig. 2). Meanwhile, an intrapulmonary right-to-left shunt related to the nodule was accidentally found in the process of CT subclavian angiography. We also found bubble signals passing her middle cerebral artery with transcranial Doppler. We performed a
specific pulmonary angiography, which confirmed a PAVM in the left superior pulmonary lobe finally (Fig. 3). This PAVM was feeded by two pulmonary arteries and drainaged by a pulmonary vein. The feeding arteries measured 7.46 mm and 7.21 mm, respectively. The PAVM was successfully resected one month after stroke. Further investigation revealed that the woman flowed nosebleed occasionally. But no telangiectasia was found on her nasal mucosa or tongue. On ambient air, her oxygen saturation was 97%.

**Discussion**

We reported a case of embolic stroke caused by an intrapulmonary right-to-left shunt. She suffered a cerebellum vermis stroke after a hard work on a hot day. It was suspected that dehydration and chronic hypoxaemia provided conditions for thrombosis[3]. Concurrently, her left vertebral artery originated from the aorta arch. Thus, posterior cerebral circulation became more vulnerable.

PAVM is always complicated with hereditary hemorrhagic telangiectasia (HHT), which is an autosomal dominant vascular disorder attributed to ENG or ACVRL1 gene mutation[4]. Patients of HHT can be observed with mucocutaneous telangiectasia and recurrent epistaxis in clinical. As reported, approximately 90% of patients with PAVM have HHT, and about 50% of patients with HHT have PAVM[2]. PAVM also can be idiopathic. In this case, no exact evidence was found for diagnosing HHT.

One cause of the embolic stroke of undetermined sources (ESUS) is PAVM, which should be kept in mind when stroke patients present with cyanosis, hypoxemia and telangiectasia. The no-contrast chest CT is necessary for young patients suspected of ESUS. The bubble contrast study with transcranial Doppler is conducive to find a right-to-left shunt[5]. Transesophageal echocardiography helps identifying the shunt as being intracardiac or extracardiac[5]. Pulmonary angiography is the standard of care to diagnose PAVM. In this case, PAVM was occasionally recognized in the examination of CT extracranial angiography. Patient’s suggestive history and discovery of pulmonary nodule on no-contrast CT provide clues for its diagnosis.

The PAVM is an uncommon but treatable cause of embolic stroke. Embolization is the preferred treatment. It is less invasive compared to surgical resection. Well, they can both prevent recurrence of embolic stroke due to the pulmonary shunt. In this case, the woman was treated by surgical resection. As reported, embolization is technically difficult for PAVM smaller than 3mm[6]. Of note is that, the incidence of PAVM recanalization is approximately 10% 5 to 7 years after embolization[7], and neither embolization nor resection could eradicate PAVM among patients with HHT, as new PAVM may be formed over years[2]. Thus, annual follow-up chest CT is necessary.

**Declarations**

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Authors' contributions: All authors contributed to the study conception. Material preparation, image collection were performed by Weitao Zhang. The first draft of the manuscript was written by Guojuan Chen. Fenjie Kan and Yibin Cao commented on previous versions of the manuscript. All authors read and approved the final manuscript.

References


Figures
Figure 1

Diffusion-weighted magnetic resonance imaging demonstrated an isolated linear infarction lesion on the cerebellum vermis
Figure 2

No-contrast chest CT revealed a pulmonary nodule in the left superior pulmonary lobe
Figure 3

Pulmonary angiography confirmed the existence of PAVM

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