

# World Journal of *Clinical Cases*

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- 1888 Endoscopic transluminal drainage and necrosectomy for infected necrotizing pancreatitis: Progress and challenges  
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- 1903 Functional role of frontal electroencephalogram alpha asymmetry in the resting state in patients with depression: A review  
*Xie YH, Zhang YM, Fan FF, Song XY, Liu L*
- 1918 COVID-19 related liver injuries in pregnancy  
*Sekulovski M, Bogdanova-Petrova S, Peshevska-Sekulovska M, Velikova T, Georgiev T*
- 1930 Examined lymph node count for gastric cancer patients after curative surgery  
*Zeng Y, Chen LC, Ye ZS, Deng JY*
- 1939 Laparoscopic common bile duct exploration to treat choledocholithiasis in situs inversus patients: A technical review  
*Chiu BY, Chuang SH, Chuang SC, Kuo KK*
- 1951 Airway ultrasound for patients anticipated to have a difficult airway: Perspective for personalized medicine  
*Nakazawa H, Uzawa K, Tokumine J, Lefor AK, Motoyasu A, Yorozu T*

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- 1963 Clinicopathological features and expression of regulatory mechanism of the Wnt signaling pathway in colorectal sessile serrated adenomas/polyps with different syndrome types  
*Qiao D, Liu XY, Zheng L, Zhang YL, Que RY, Ge BJ, Cao HY, Dai YC*

**Randomized Controlled Trial**

- 1974 Effects of individual shock wave therapy *vs* celecoxib on hip pain caused by femoral head necrosis  
*Zhu JY, Yan J, Xiao J, Jia HG, Liang HJ, Xing GY*

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- 1985 Very low calorie ketogenic diet and common rheumatic disorders: A case report  
*Rondanelli M, Patelli Z, Gasparri C, Mansueto F, Ferraris C, Nichetti M, Alalwan TA, Sajoux I, Maugeri R, Perna S*
- 1992 Delayed versus immediate intervention of ruptured brain arteriovenous malformations: A case report  
*Bintang AK, Bahar A, Akbar M, Soraya GV, Gunawan A, Hammado N, Rachman ME, Ulhaq ZS*

- 2002** Children with infectious pneumonia caused by *Ralstonia insidiosa*: A case report  
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- 2009** Transient ischemic attack induced by pulmonary arteriovenous fistula in a child: A case report  
*Zheng J, Wu QY, Zeng X, Zhang DF*
- 2015** Motor cortex transcranial magnetic stimulation to reduce intractable postherpetic neuralgia with poor response to other therapies: Report of two cases  
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## Transient ischemic attack induced by pulmonary arteriovenous fistula in a child: A case report

Jun Zheng, Qi-Yue Wu, Xia Zeng, Du-Fei Zhang

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### Abstract

#### BACKGROUND

Cerebral ischemic stroke is attributed to paradoxical cerebral embolism. Pulmonary arteriovenous fistula (PAVF) is a rare potential cause of cerebral ischemic stroke, and cerebral ischemic stroke induced by PAVF in children is rare.

#### CASE SUMMARY

We report a case of right PAVF that presented as a transient ischemic attack (TIA) in a 13-year-old boy. The patient underwent embolization therapy and remained clinically stable for 2 years after treatment.

#### CONCLUSION

TIA induced by PAVF in children is rare, lacks typical clinical manifestations, and should not be ignored.

**Key Words:** Pulmonary arteriovenous fistula; Transient ischemic attack; Paradoxical cerebral embolism; Children; Case report

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**Core Tip:** Pulmonary arteriovenous fistula (PAVF) is a rare potential cause of cerebral ischemic stroke. Children with PAVF have atypical clinical presentations, and even present with cerebral ischemic stroke or transient ischemic attack as the only clinical finding. If a PAVF is suspected, we recommend that appropriate examinations should be performed for early detection, and then active treatment and follow-up should be offered.

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## INTRODUCTION

Cases of cerebral ischemic stroke induced by pulmonary arteriovenous fistula (PAVF) are mostly reported in adults, and are largely attributed to paradoxical cerebral embolism (PCE)[1-5]. Due to congenital or acquired arteriovenous communications, PCE can be caused by emboli transferring from a vein to an artery, or transferring from the right heart system to the left heart system. PCE is most commonly induced *via* a patent foramen ovale (PFO)[6], and PAVF is a rare potential cause. Cerebral ischemic stroke induced by PAVF in children is rare. We report the case of a 13-year-old boy with a transient ischemic attack (TIA) induced *via* PAVF. We aim to bring attention to this rare cause of PCE with the goal of decreasing the rate of missed diagnosis.

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## CASE PRESENTATION

### **Chief complaints**

A 13-year-old boy was admitted to our hospital on September 2, 2020 due to intermittent dizziness for 2 d.

### **History of present illness**

The patient presented with intermittent dizziness and bilateral limb weakness for 2 d. During the attack, he fainted and then recovered spontaneously approximately 5 min later. He had no dyspnea, nausea, vomiting, tinnitus, hearing loss, or convulsions.

### **History of past illness**

The patient had a no previous medical history.

### **Personal and family history**

The patient had no relevant personal or family history.

### **Physical examination**

Physical examination showed no neurological or cardiorespiratory abnormalities. His vital signs were stable.

### **Laboratory examinations**

The results of laboratory examinations were normal.

### **Imaging examinations**

Subsequent chest x-ray showed a mass shadow in the right upper lung (Figure 1). Cardiac color ultrasound showed no obvious abnormalities and no PFO was seen. Cardiovascular computed tomography (CT) angiography (CTA) showed that the right upper pulmonary artery (diameter 7.4 mm) had expanded. Its branching vessels were thickened and twisted into an abnormal vascular nest with direct reflux into the right upper pulmonary posterior vein, and the artery finally merged into the right upper pulmonary vein (Figure 2). Brain magnetic resonance imaging and cerebral artery magnetic resonance angiography showed no obvious abnormalities (Figure 3). Contrast-enhanced transcranial Doppler (c-TCDD) ultrasound was remarkable for a large right-to-left shunt (RLS) (Figure 4).

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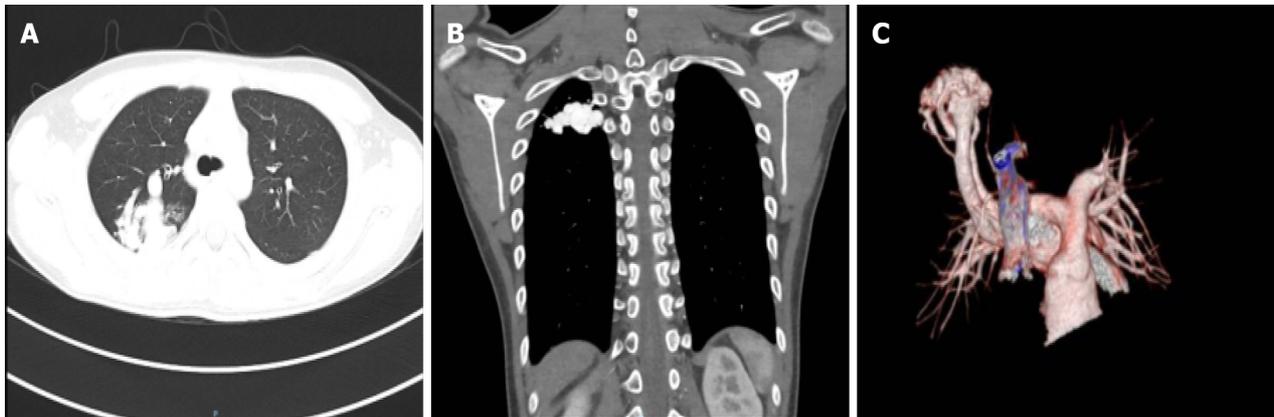
## FINAL DIAGNOSIS

The patient was diagnosed with TIA and PAVF. TIA induced *via* PAVF was considered.



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Figure 1 Chest x-ray. A mass shadow in the right upper lung (red arrow).



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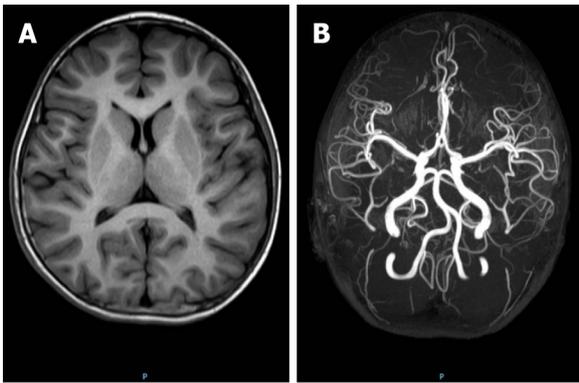
Figure 2 Cardiovascular computed tomography angiography. A and B: Representative images showing the abnormal vascular nest in the right upper lung; C: Expansion of the right upper pulmonary artery with thickened and twisted branching vessels to form an abnormal vascular nest with direct reflux into the right upper pulmonary posterior vein. The artery finally merged into the right upper pulmonary vein.

## TREATMENT

The patient underwent embolization therapy of the PAVF under general anesthesia on the day 3 of hospitalization. Intraoperative angiography showed that the PAVF originated from the right upper pulmonary branch artery (Figure 5A and B), which was no longer detected after embolization with a vascular plug (Figure 5C and D).

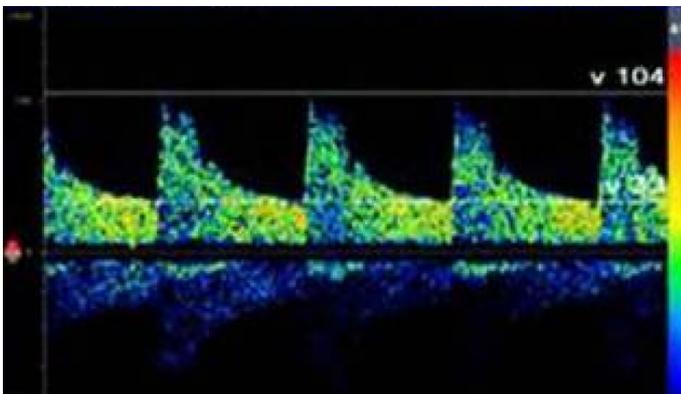
## OUTCOME AND FOLLOW-UP

At the 10 mo postoperative follow-up, a chest x-ray showed that the position of the vascular plug was stable (Figure 6A), and a CT scan showed that the PAVF had markedly shrunk (Figure 6B). At the 1 year and 2 years postoperative follow-up points, the patient remained clinically stable without symptoms of dizziness, limb weakness, or fainting.



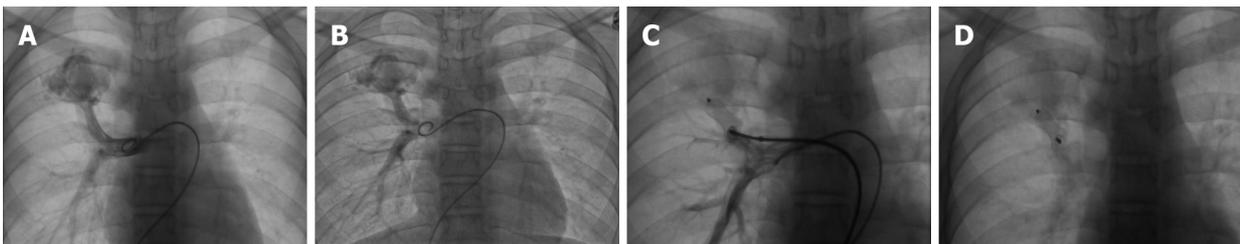
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**Figure 3 Brain magnetic resonance imaging and cerebral magnetic resonance angiography.** A: Magnetic resonance imaging showed no obvious abnormalities; B: Magnetic resonance angiography showed no obvious abnormalities.



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**Figure 4 Contrast-enhanced transcranial Doppler ultrasound.** Significant embolus signals appeared in the middle cerebral artery within 10 s after Valsalva maneuver.

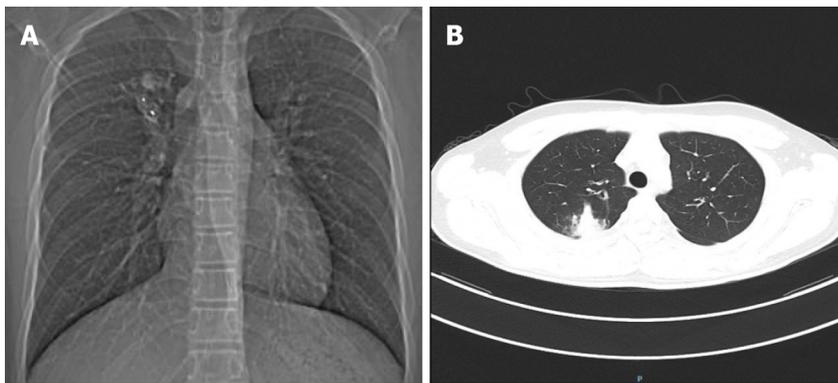


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**Figure 5 Angiography and embolization treatment.** A and B: Representative angiography images showing a pulmonary arteriovenous fistula (PAVF) originating from the right upper pulmonary branch artery; C and D: Representative images showing the PAVF was no longer detected after embolization with a vascular plug.

## DISCUSSION

The presence of a RLS, which can result in paradoxical brain embolism, is an important etiology of ischemic stroke especially in young adults. PFO and PAVF are the most important causes of RLS. TCD ultrasound is currently reported to be a noninvasive and useful method for detecting RLS. The gold standard for detecting RLS is contrast-enhanced transesophageal echocardiography. It is recommended that these examinations be carried out immediately after onset of stroke[7]. A previous report showed that the prevalence of PAVF is 0.026% [8], and TIA is a clinical manifestation of PAVF in up to 20% of cases[1]. At present, it is believed that the risk factors for cerebral ischemic stroke in patients with PAVF are as follows: (1) Feeding artery diameter > 3 mm; and (2) existence of multiple PAV malformations[9]. Whether patients with PAVF present with clinical manifestations depends on the level of the RLS. There



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**Figure 6 Postoperative follow-up imaging.** A: Chest x-ray showed that the position of the vascular plug was stable; B: Computed tomography showed that the pulmonary arteriovenous fistula markedly shrank after embolization therapy.

are intrapulmonary and extrapulmonary manifestations in patients with PAVF. The intrapulmonary manifestations include dyspnea after activity, dizziness, easy fatigability, and others[10-12]. Physical examination may reveal cyanosis, clubbing of fingers or toes, or continuous chest murmur. Some patients may only present with abnormal arterial blood gas analysis. The extrapulmonary manifestations include migraine, TIA, ischemic stroke, brain abscess, epilepsy, and others[10-12].

Espejo-Herrero *et al*[1] reported a patient with TIA presenting with a short-term (30 min) right limb paralysis, with subsequent pulmonary arteriography showing a PAVF. Pulmonary digital subtraction angiography (DSA) is the gold standard for the diagnosis of PAVF, which can determine fistula size, feeding artery, draining vein, and other conditions[10]. However, DSA is an invasive examination. As a noninvasive procedure, CTA can not only show the lesions (even mild lesions) and the blood vessels involved in PAVF, but can also accurately judge peripheral and complex PAVF anatomy, which is more helpful in diagnosis[10]. At present, c-TCD is widely used for RLS screening, which can observe the dynamics of emboli entering the intracranial arteries and the changes of cerebral blood flow in real time [13]. In our case, c-TCD ultrasound revealed a large RLS, which was in line with the diagnosis of PAVF.

Most PAVFs will gradually enlarge and rarely shrink spontaneously, which may cause serious complications. The mortality rate in untreated patients with PAVF is as high as 50%, which can be reduced to 3% after treatment. It is currently recommended that active treatment should be pursued in patients with symptomatic or asymptomatic PAVF if the lesion diameter exceeds 3 mm. PAVF treatments mainly include surgery and interventional embolization, both of which can improve the symptoms of hypoxia and prevent the occurrence of central nervous system complications[10,11]. Todo *et al*[14] reported a patient with recurrent ischemic stroke caused by PAVF with a feeding artery diameter of 1.80 mm, and embolic events were successfully prevented after fistula embolization. Our patient was treated with interventional embolization of PAVF in our hospital. The patient had no recurrent seizure or TIA symptoms for 2 years, further suggesting that recurrent TIA symptoms are indeed associated with PAVF.

## CONCLUSION

Although PAVF is a rare cause of abnormal embolism, it should not be ignored. Most patients with PAVF have atypical clinical presentations, and can even present with cerebral ischemic stroke or TIA as the only clinical finding. Therefore, in patients with cryptogenic stroke, especially children, appropriate examinations should be performed to determine whether a PAVF may be present. If a PAVF is discovered, active treatment and follow-up should be pursued.

## FOOTNOTES

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