Omitted Pulmonary Arteriovenous Fistula Complicated With Brain Abscess

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Abstract

A 52-year-old female, diagnosed with a bilateral lower lobe pulmonary arteriovenous fistula (PA VF) at the age of 14 due to dyspnea, underwent a lobectomy of the right lower lung with a severe lesion, and symptom relief without treatment of the minor lesion in the other lung. She had active chest tightness since 42 years old and headaches 10 days before hospitalized, was diagnosed with PA VF and brain abscess, and cured by a successful PA VF embolization and a surgical resection of brain abscess. In the same patient, both methods of treatment for PA VF were all satisfactory, and each has advantages and indications.

Keywords: Pulmonary arteriovenous fistula; Brain abscess; Transcatheter embolization

Introduction

Pulmonary arteriovenous fistula (PAVF) is a congenital or acquired pulmonary vascular malformation (PVM) with a thin-wall and dilated structure. PAVF brings about anatomic right-to-left shunts between pulmonary arteries and veins without capillary beds for gas exchange and filtration. The patients with serious shunt experience chest tightness, dyspnea, palpitation, thoracalgia, hemoptysis and other symptoms. PAVF predisposes to complication of hemorrhage, paradoxical embolization leading to stroke and brain abscess [1, 2].

Case Report

A 52-year-old woman presented to neurology department with a chief complaint of headache for 10 days, and had an anamnesis of surgical therapy for right lower lobe PAVF before 38 years. She was hospitalized with active chest tightness before 10 years and diagnosed with left lower lobe PAVF. The lesion was significantly more severe than the original. Family history included mother dying of PAVF complicated with severe empysema, and daughter suffering from PAVF in left lower lobe. Outpatient computed tomography (CT) displayed occupying lesion in the occipital lobe of the right brain and peripheral edema (Fig. 1a), multiple nodules in the left lower lung and vascular malformations with high probability (Fig. 1b, c). On admission, body temperature was 36.2°C, cyanotic lips, foreheads visible vascular nevus and limbs tendon hyperreflexia. Blood examination showed a white blood cell count of 9,030 cells/μL, neutrophil ratio of 79%, hemoglobin of 151 g/L, hematocrit of 47.9% and hypersensitive C-reactive protein of 14.3 mg/L. Arterial blood gas (ABG) reported resting oxygen partial pressure of 58.0 mm Hg and oxygen saturation of 83%. Rachiocentesis results showed intracranial pressure was 130 mm H2O, cerebrospinal fluid was colorless transparent (protein, 1.862 mg/dL; chloride, 117.5 mmol/L; glucose, 2.50 mmol/L), Paneth test was weakly positive, red blood cell counts was 10 cells/μL and leucocyte count was 335 cells/μL. Pulmonary computed tomography angiography (CTA) presented left lower lobe PVM and PAVF, and the lesion diameter was about 10.5 mm (Fig. 2a). Head MRI reported that brain abscess occurred in the right occipital lobes, and there were many of old cerebral infarctions (Fig. 2b).

As antibiotic therapy, 3.0 g/day of ceftazidine and 1.6 g/day of norvancomycin hydrochloride had been administered intravenously. The oxygen saturation was increased to 94% from 83% by supplementary oxygen. Headache symptom in the patient improved significantly, and interventional embolization took place under local anesthesia after 7 days (Fig. 3). Spring coils were implanted to the aneurysm neck, and angiography showed that the fistula was closed completely. Chest tightness disappeared immediately after occlusion. Post-procedure ABG indicated that resting oxygen partial pressure increased to 89.0 mm Hg and oxygen saturation increased to 99%. Excision of cerebral abscess was performed 5 days later, and a cystic lesion and brown turbid pus were shown. Pathological findings suggested chronic granulomatous disease and acute inflammation. Twelve days after the operation, the patient was followed up for MRI: the area of the right occipital lobe lesion was blurred, and the white matter could be seen with edema band. She was discharged on the 22nd day after operation.

Discussion

PAVF was first reported by Churton in 1897 [3]. Due to the
limitations of imaging, diagnosis of such diseases was more
difficult, and was considered to be a rare disease before 1950s.
Hereditary hemorrhagic telangiectasia (HHT) is the most
common cause of PAVFs in patients, an autosomal dominant
disease, also known as Randu-Osler-Weber disease [4, 5]. A
diagnosis mainstay of HHT is the Curacao criteria, which rep-
resent an international consensus initially published in 2000.
A diagnosis of definite HHT is recognized if at least three of
the following are present: 1) epistaxis (spontaneous, recur-
rent); 2) telangiectasia (multiple at characteristic sites, i.e.,
lips, oral cavity, fingers, nose); 3) visceral lesions, including,
but not limited to, GI telangiectasia (with or without bleeding),
pulmonary, hepatic, cerebral, and spinal arteriovenous malfor-
mations; and 4) family history of a first-degree relative with
HHT according to these criteria [6]. In addition, mutations in
three genes (ENG, ACVLI/ALK1 and SMAD4) with varying
clinical phenotypes have been identified. The genes can also be
used to diagnose HHT [3]. At present, the main physical exam
in clinical diagnosis of PAVF is CTA, pulmonary artery an-
giography, contrast echocardiography and MRI [7]. The most
effective treatment is surgical excision and interventional em-
bolization, and the latter as a gold standard in the treatment of
PAVF is prevalent.

No matter whether there is a symptom or not, emboliza-
tion should be performed for PAVF with a diameter greater than
3 mm, which can effectively reduce the risk of complications
such as paradoxical embolism, namely “3 mm rule” [8]. Surgical
treatment as a supplementary approach is mainly used in
patients with interventional treatment failure or who need lung
transplantation [3]. It should be noted that PAVF often increased
in size and number during pregnancy, and a small proportion of
women experienced life-threatening complications [1, 9].

Figure 1. (a) Head CT showing an irregular low density in the right occipital lobe. (b) Chest CT scan illustrating multiple nodules
with varying sizes and uniform density in the left lower lobe. (c) Chest roentgenogram shows PAVFs occurred in the left lower
lobe and the residual lobes had a good compensation in right lung after operation.

Figure 2. (a) Three-dimensional CT imaging displaying pulmonary arteriovenous malformation, tortuosity and dilation in left lower
lobe (arrow). (b) Cranial MRI showing a cystic lesion with peripheral edema in the right occipital lobe, brain abscess formation.
The patient was diagnosed with bilateral lower lobe PAVF at the age of 14, and received lobectomy, and the mild lesion in the other lung was without treatment. With the progression of the lesion, the symptom reappeared before 10 years, complicated with multiple cerebral infarction and brain abscess, and complete treatment with interventional embolization. In one case, two different treatment methods were used successively, and good therapeutic effect was achieved. Lobectomy is a radical treatment. The residual lobes have a good compensation after operation in Figure 1c, but the disadvantage is trauma. Advantages of transcutaneous catheter embolization are minimally invasive and rapid recovery. The defect is that the lesion may continue to grow without excision, and there are potential hazards of occlude abscission, bleeding, stroke, and other risks.

The case gives us a revelation that PAVF should be diagnosed and treated early to avoid infarct, hemorrhage, brain abscess and other complications. Young patients with the single lesion, especially the women who have a pregnancy plan should be suggested to apply a radical surgical treatment. Geriatric patients and those with multiple lesions should receive embolization treatment aim to improve symptoms with minimally invasive methods.

Disclosure

No potential conflicts of interest relevant to this article were reported. Ethical approval was given by the medical ethics review committee of hospital.

References