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Pulmonary arteriovenous malformations complicated by splenic infarction and abscess

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Abstract

Pulmonary arteriovenous malformation (PAVM) is an abnormal blood vessel connecting a pulmonary artery and a vein, and is accompanied by paradoxical embolism to other organs due to a right-to-left shunt. We report the case of a 66-year-old woman with PAVM complicated by splenic infarction and abscess. Although the PAVM had been detected on a chest image 2 years previously, and she had been advised to have further investigations, she decided not to follow this further at the time. She then visited our hospital complaining of worsening dyspnoea on exertion. Detailed examinations revealed splenic infarction and abscessation due to PAVM. PAVM embolization was performed after antibiotic treatment. It is very rare for PAVM to be complicated by splenic infarction and abscess. Regardless of its size, embolization of a PAVM as soon as possible can reduce not only the risk of central nervous system complications, but also the risk of splenic infarction and abscess.

Introduction

Pulmonary arteriovenous malformation (PAVM) is a structurally abnormal blood vessel, resulting in direct capillary-free communication between the pulmonary and systemic circulations, and therefore an anatomic right-to-left shunt [1]. PAVM pre-disposes to a variety of complications such as ischaemic stroke and cerebral abscess. However, it is extremely rare to encounter splenic infarction [2]. Herein, we present a case of PAVM complicated by splenic infarction and abscess.

Case Report

A 66-year-old female was admitted to a nearby hospital with dyspnoea and an abnormal shadow on chest computed tomography (CT) in September 2014. Contrast-enhanced CT of the chest showed a PAVM, approximately 5 cm in size, in the left lower lung lobe. She was referred to our hospital for observation and treatment. However, because her symptoms were controlled, she was not admitted, and no follow-up for the PAVM was conducted.

In June 2016, due to worsening symptoms, she was admitted to our hospital. Her mMRC score was 2. She had no other relevant medical history. Close relatives had experienced intracranial haemorrhage and two descendants had presented with recurrent nosebleeds. However, no one was diagnosed with hereditary haemorrhagic telangiectasia (HHT). At admission, her weight was 66 kg and height was 155 cm. Her vital signs were: Glasgow coma scale, 15 (E4V5M6); blood pressure, 98/77 mmHg; pulse rate, 112 beats/min; pulse oximetry, 83% in room air; respiratory rate, 22/min; and body temperature, 38.0°C. Her quick sepsis-related organ failure assessment (qSOFA) score was 2 (respiratory rate ≥ 22/min and systolic blood pressure ≤ 100 mmHg). Chest auscultation revealed a vascular murmur in the left lung field. She had no cutaneous telangiectasia. Blood tests revealed inflammatory findings associated with infection, disseminated intravascular coagulation (DIC), and liver dysfunction. No significant findings were obtained in various culture tests. Thoracic radiographs demonstrated the presence of a PAVM in the left lower lobe (Fig. 1A). Enhanced CT of the chest and

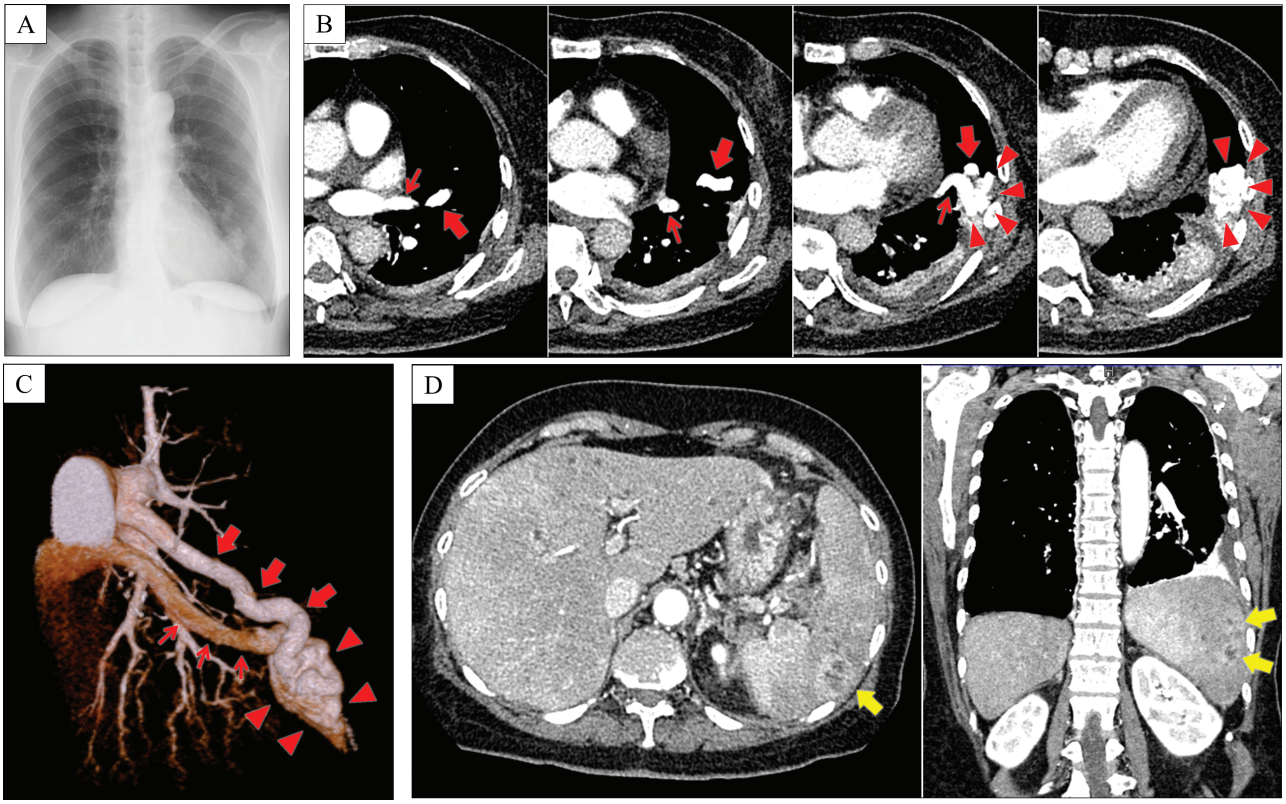

Figure 1. (A) Thoracic radiography showed an abnormal shadow suspected to be pulmonary arteriovenous malformation (PAVM) and associated artery and vein in the left lower lobe. (B, C) Enhanced computed tomography (CT) of the chest and three-dimensional CT showed PAVM sac (triangle), feeding artery (arrow), and draining vein (thin arrow). (D) Abdominal CT showed a heterogeneous low density area in the spleen (yellow arrow); this lesion was determined to represent splenic infarction and abscess.

three-dimensional reconstructed images revealed that the PAVM had one feeding artery and one draining vein with diameters of 10 and 12 mm, respectively. The sac was 16 mm in diameter (Fig. 1B, C). Abdominal CT and ultrasonography revealed a low-density area and hypoechoic lesion in the spleen, which was consistent with splenic infarction and abscessation (Fig. 1D). Needle aspirations of the splenic abscess were not performed because she had DIC. No findings suggestive of infective endocarditis and congenital heart disease were found on performing echocardiography. Lung perfusion scintigraphy gave a shunt ratio of 13.9%. Furthermore, brain magnetic resonance imaging revealed multiple cerebral infarctions and a cerebral aneurysm, whereas upper gastrointestinal endoscopy detected gastric varices. Although HHT was suspected, her clinical features did not satisfy its diagnostic criteria [3].

Anticoagulant and antimicrobial therapies [doripenem 1 g intravenous (i.v.) every 8 h for 25 days, following oral amoxicillin/clavulanate 250 mg every 6 h and metronidazole 250 mg every 6 h for 3 days and then amoxicillin/clavulanate 250 mg every 6 h and clindamycin 300 mg every 8 h for 32 days] were initiated. Approximately 30 days later, the splenic abscess and the patient's general condition had improved. Embolization of the PAVM was then performed (Fig. 2). No complications occurred during or after embolization.

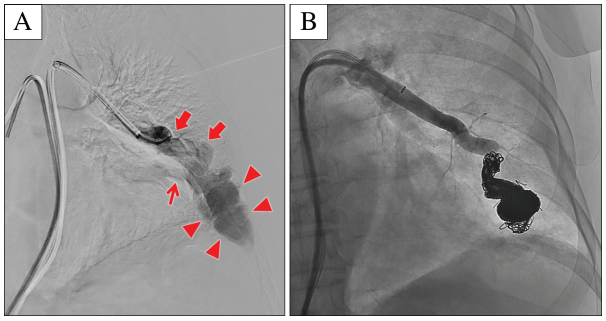

Figure 2. (A) Pulmonary arteriography showed pulmonary arteriovenous malformation (PAVM) sac (triangle), feeding artery (arrow), and draining vein (thin arrow). (B) After embolization with coils, pulmonary arteriography showed enhancement of the feeding artery but no enhancement of the PAVM sac or draining vein.

Discussion

The present case shows two notable clinical findings. To begin, splenic infarction and abscess can complicate PAVM. Second, because untreated PAVM can cause serious complications, appropriate therapeutic intervention is vital.

Splenic infarction and abscess is a rare complication in PAVM. Complications usually involve neurological emboli [1]. In untreated PAVM, 37% of untreated patients suffer transient cerebral ischaemic attack, 18% have stroke, and 9% develop brain abscesses [4]. Embolic symptoms involving other organs are rarely reported, and splenic infarction and abscess has been reported from Japan only once [2]. In that report, PAVM was considered the cause of splenic infarction and abscess because infective endocarditis and other congenital heart disease were excluded. As PAVM has a right-to-left shunt, bacteria and thrombus, normally filtered out by the lung remain, causing abscesses and infarction of other organs. Here as well as in the previous report, the splenic issues were considered to be caused by an embolism via PAVM. In general, splenic abscess is mainly caused by *Staphylococcus aureus*, *Klebsiella* spp., and anaerobic bacteria [5]. In this case, we first chose doripenem in view of septicaemia and intraperitoneal abscess. After that, amoxicillin/clavulanate and metronidazole were selected because anaerobic bacteria were suspected. Because anorexia, which was thought to be caused by metronidazole, appeared, it was changed to clindamycin. Although causative bacteria were not identified in culture tests, symptoms and imaging findings improved.

Early intervention for PAVM is important to prevent severe complications. The preferred treatment for PAVM is transcatheter coil embolization, and the indication for treatment is a feeding artery of 3 mm or more, the "3-mm rule" [1]. However, Shovlin et al. reported that embolization should not be limited by the "3-mm rule" [6]. They also reported that the diameter of PAVM blood vessels and cerebral embolism were not associated, and embolization of all angiographically visible PAVM prevented any central nervous system complications [6]. Complications of PAVM in other organs, such as splenic infarction and abscess, may be caused by the same mechanism of paradoxical embolism as those of the central nervous system. The risk of these complications may similarly be lowered by embolization. The present case had a large PAVM with a high degree of shunting, and severe complications including splenic infarction and abscess; perisplenic inflammation, septicaemia, and DIC occurred due to not embolizing the PAVM sooner. These complications could have been prevented, if appropriate therapeutic interventions had been performed at diagnosis.

In conclusion, splenic infarction and abscess can occur as complications of PAVM. Furthermore, early intervention will lower the risk of complications, including splenic embolism.

Disclosure Statements

No conflict of interest declared.
Appropriate written informed consent was obtained for publication of this case report and the accompanying images.

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