Learning from errors

Traumatic pulmonary arteriovenous fistula may be misdiagnosed with residual shunt after patent foramen ovale closure

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Summary

We report the case of a 48-year-old woman with a history of transient ischaemic attacks (TIA). The patient was diagnosed with a large patent foramen ovale (PFO), for which she underwent percutaneous PFO closure. Two years postprocedurally, the patient suffered frequent TIA and assessment showed the displacement of the device. The device was surgically removed, and the septum was repaired with a pericardial patch. Afterward, however, the patient experienced repeat TIA. It was discovered that the patient had a pulmonary arteriovenous fistula (PAVF), and a diagnosis of a trauma was made. The PAVF was later coil embolised. This case illustrates the importance of evaluating PFO closure patients for the associated causes of right-to-left shunting.

BACKGROUND

Transcatheter closure of patent foramen ovale (PFO) has rapidly evolved as the preferred management strategy for the prevention of recurrent cerebrovascular events in patients with stroke and paradoxical embolus. It is advisable that physicians be aware of such possible misdiagnosis and be prepared to better manage these kind of patients.

CASE PRESENTATION

In January 2005, a 48-year-old woman was referred to our hospital for further assessment of some frequent transient ischaemic attacks (TIA), which had left no residual sequel.

Transesophageal echocardiography revealed a prominent Eustachian valve, lipomatous hypertrophy of the atrial septum and a PFO with an atrial septal aneurysm as well as a permanent right-to-left shunt (not a Valsalva manoeuvre-induced right-to-left shunt).

The patient was subjected to the transcatheter closure of the PFO using a 25 mm amplatzer occluder in another centre. The antiplatelet regimen consisted of 81 mg of aspirin and 75 mg of clopidogrel daily for the first month, and 325 mg of aspirin thereafter.

Two years after the procedure, the patient presented with a new ischaemic stroke in the right middle cerebral artery territory, confirmed by brain diffusion-weighted sequence MRI. Physical examination was otherwise unremarkable. Cervical and transcranial ultrasonography, 12-lead ECG, 24-h ECG monitoring, and chest radiography were normal. Routine blood tests and thrombophilia study, including tests for protein C, protein S, antithrombin III, factor V Leiden, factor II, hyperhomocysteinaemia and antiphospholipid antibodies, did not show any abnormality. Contrast transesophageal echocardiography revealed pronounced appearance of contrast bubbles along the anterior edges of the device. Echocardiography showed the mild displacement of the device into the right atrium and a residual shunt (video 1).

The device was surgically removed, and the septum was repaired via a pericardial patch. Interestingly, the amplatzer occluder device, which was removed after 2 years, showed no signs of mechanical failure, corrosion or clinically relevant immunological response at the device-tissue interface.

Postoperative transesophageal echocardiography demonstrated a good result with no residual shunt. The patient had good recovery. After 6 months, however, she returned with recurrent TIA. Contrast transesophageal echocardiography revealed the massive appearance of contrast bubbles from an unknown origin. The septum was intact, but there was a massive early (in the third cardiac cycle) passage of microbubbles to the left side of the heart without evidence of pulmonary artery hypertension or ventricular cavity enlargement.

With the probability of a pulmonary arteriovenous fistula (PAVF), the patient was scheduled for multisliced CT, which confirmed the presence of a large AV fistula in the right lower lobe with a small metallic particle (figure 1). The

Video 1 Echocardiography showed the mild displacement of the device into the right atrium and a residual shunt.

Figure 1 (A) and (B) CT scan, large arteriovenous fistula in the right lower lobe with a small metallic particle.

diagnosis was a laterolateral fistula between the laterobasal segmental pulmonary artery (6 mm in diameter) and an aneurysmal sac draining towards an enlarged pulmonary vein (8 mm in diameter) and the distal pulmonary artery (5 mm in diameter). Clinical examination did not show a continuous murmur in the left posterior thorax.

The patient's medical history revealed that 24 years earlier, during the war, she had suffered a penetrating chest trauma with missile debris. Multisliced CT demonstrated shrapnel in the site of the fistula in the right lung: this could be the cause of the AV fistula. The draining artery was occluded first with a plug, 10 mm in diameter and the feeding artery was subsequently occluded with a plug, 12 mm in diameter. The patient recovered uneventfully, and continues to do well at present.

The early shunting in this case was related to the proximal location of the fistula and to the massive shunting.

OUTCOME AND FOLLOW-UP

The patient appears to be in good physical condition at long-term follow-up, with no other related problems.

DISCUSSION

The following points should be underscored in the case presented herein.

The presence of lipomatous hypertrophy and an aneurysmal atrial septum can also be associated with persistent early shunting. An excessively thick septum will limit the ability of the closure device to straddle the interatrial septum and will predispose it to inappropriate seating and residual shunting.¹ The use of a usual device for our patient, in combination with inappropriate positioning of the device, was first mistakenly deemed to have complicated the case. The second presentation of the patient with recurrent TIA, however, raised the suspicion of a residual shunt of the device, and the presence of bubbles in the left atrium confirmed these suspicions. Other problems of note were the early appearance of the bubbles in the left atrium and the rarity of a traumatic PAVE.

The medical literature contains numerous accounts of PAVFs; be that as it may, only a small percentage of these references have dealt with acquired PAVFs.² Aetiologies of acquired fistulas encompass metastatic carcinoma of the thyroid, pulmonary schistosomiasis, actinomycosis and long-standing hepatic cirrhosis, Fanconi syndrome, mitral stenosis and, very rarely, penetrating chest traumas.^{3–5} Penetrating chest traumas are an unusual cause, with only a few previous cases having been reported in the existing literature.^{6–11} The rarity of AV fistulas following the trauma of the pulmonary vasculature is probably due to relatively small pressure differences between pulmonary arterial and venous circulations, in contrast to the high gradient between the arteries and veins of the systemic circulation. Consequently, the canalisation of a haematoma is less likely to occur in the pulmonary vasculature. Generally, the diagnosis of both congenital and acquired PAVFs has been made using a combination of data from the patient's history, physical examination, laboratory testing, chest radiographs, CT scans, contrast echocardiography and pulmonary angiography.¹

Our case also demonstrates a remarkable feature with respect to the timing of the appearance of the contrast in the left atrium. The timing of the appearance of the agitated saline in the left atrium helps us differentiate an intrapulmonary from an intracardiac shunt: whereas in an intrapulmonary connection, it takes three to five cardiac cycles after right atrial opacification before the contrast appears in the left atrium, the contrast was visualised in the left atrium within one to two cardiac cycles in our patient, which is more suggestive of an intracardiac shunt such as an atrial septal defect. We think that the early appearance of the bubbles in the left atrium was the result of an extraordinarily large fistula and low distance to the heart.

Learning points

- To avoid such mismanagements, it is advisable that due need be paid to patent foramen ovale closure in the complex interatrial septum anatomy.
- Furthermore, associated anomalies should always be taken into account in all medical fields before embarking upon any intervention.
- And last but not least, a meticulous and thorough extraction of the patient's medical history is integral to accurate diagnosis and management.

Competing interests None.

Patient consent Obtained.

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