

SHORT REPORT

Acute Leriche Syndrome due to Paradoxical Embolism

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Paradoxical embolism, describing the passage of venous or right-sided cardiac thrombus into the arterial or systemic circulation through patent foramen ovale, is an uncommon cause of acute arterial occlusion. Here, we report acute Leriche syndrome in a young woman attributable to paradoxical embolism. Ischaemia, patent foramen ovale, and venous thrombosis were the triad of evidence for paradoxical embolism.

Case Report

A 44-year-old French woman, on vacation in the Maldivian Islands, experienced right leg pain and widespread oedema of the right lower limb within 24 h of her arrival. She was rushed to the local emergency unit. There was no relevant history or other symptoms. She was a smoker (10 packs/year). Clinical examination revealed tachycardia (128/min), a pelvic mass and a sensory-motor deficit of the right lower limb. The pulses of the leg were not reported. The menstrual periods were regular. Laboratory investigations showed a microcytic anemia (Hb 6.1 g/dL, MCV 60 cu μm^3) and leukocytosis (23,000/mm³). Abdominal ultrasonography disclosed a solid pelvic mass. Over the next 24 h, both legs became painful and paralyzed and the patient was repatriated to a Department of Neurology in France. On admission, they noted that the lower extremities were cold and femoral pulses were absent. Consequently, the patient was transferred to a department of Vascular Surgery. A CT-scan showed an aortic embolic occlusion extending from the ostium of the inferior mesenteric artery to both external iliac arteries (Fig. 1A and B).

CT-scan confirmed the presence of a pelvic mass, probably from the uterus, without any sign of vascular compression or loco-regional invasion (Fig. 1B). Urgent embolectomy through bifemoral arteriotomies was performed associated with a right lower extremity fasciotomy. The patient received anticoagulation therapy with continuous unfractionated heparin infusion. One week later, a total hysterectomy (without oophorectomy) was performed. Uterine histopathology showed a submucosal leiomyoma. Trans-oesophageal echocardiography (TEE) did not show any thrombus in the heart cavity, but an atrial septal aneurysm (ASA) was identified (Fig. 2A). After injecting a bolus of agitated saline into an antecubital vein, microbubbles appeared in the left atrium within 3 cardiac cycles of their appearance in the right atrium suggesting a patent foramen ovale (PFO) (Fig. 2B). Upon further investigation, a venous duplex ultrasound scan revealed a free-floating thrombus in the right femoral and iliac veins. Despite the lack of thoracic pain or dyspnoea, a ventilation/perfusion lung scan was performed and showed a perfusional defect of 35–40% related to bilateral pulmonary embolism.¹ Lower extremity ischaemia, PFO and venous thrombosis suggested the diagnosis of paradoxical embolism (PDE). The patient was discharged home 3 weeks later on long-term oral anticoagulation (warfarin), with a walking exercise program for a right leg motor deficit. After warfarin was stopped (6 months later), a coagulation screen was normal.

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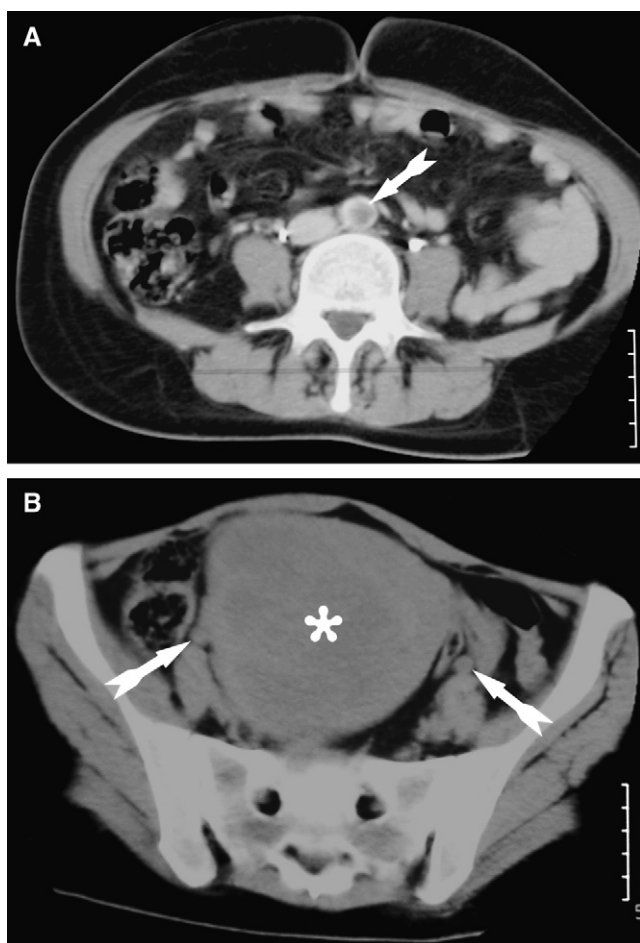


Fig. 1. (A) Distal aorta occlusion (arrow) confirmed by computed tomographic angiogram. (B) CT angiography provided visualization of a voluminous pelvic mass (asterisk) developed from the uterus with contact with vertebral column. Iliac arteries (arrows) are occluded but seem free of tumour invasion.

Finally, 1 year later the patient was asymptomatic. The patient refused percutaneous closure of the PFO.

Discussion

We present an unusual case of a Leriche syndrome in a pre-menopausal female smoker and without previous history of cardiovascular disease. On the basis of age, history, clinical and radiological findings an unconventional cause of emboli, such as PDE was suspected.

Cardiac sources as atrial fibrillation and myocardial infarction account for 80% of all peripheral arterial thromboembolic events.¹ Far less common are non cardiac sources like atherosclerotic disease involving large vessels, hypercoagulable states, non-cardiac tumors, white clot syndromes, cervical ribs, foreign bodies or PDE. Paradoxical embolism refers

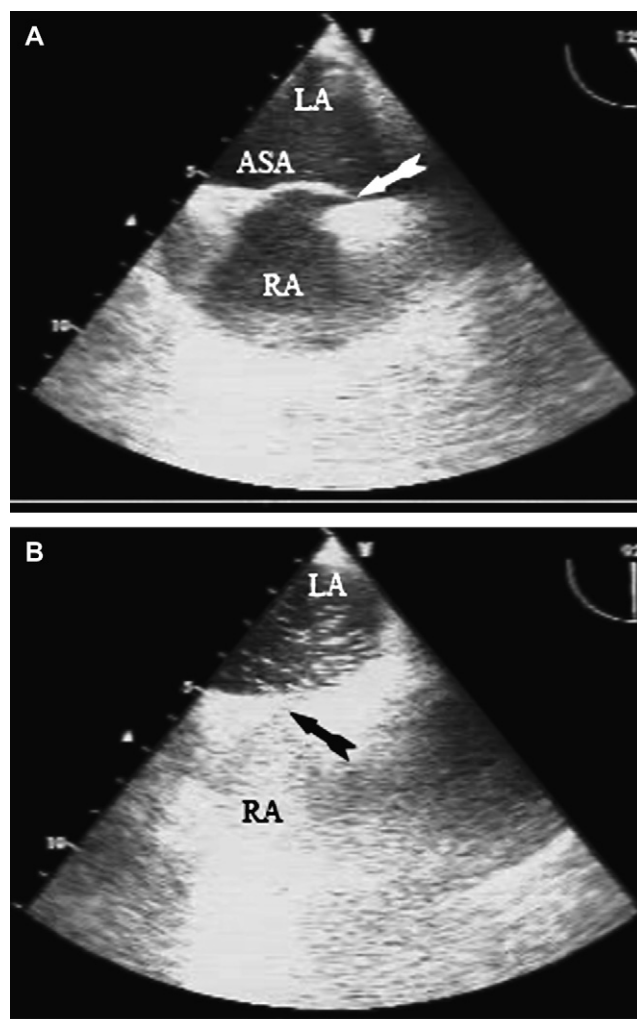


Fig. 2. (A) Atrial septal aneurysm and patent foramen ovale (arrow) was detected by oesophageal echocardiography. Exploration of heart cavities did not show any thrombus. (B) Passage of microbubbles from the right to left atrium across a patent foramen ovale (arrow) was demonstrated by oesophageal echocardiography. RA: right atrium; LA: left atrium; ASA: atrial septal aneurysm.

to the embolic entry of a venous thrombus into the systemic circulation through a right-to-left shunt. PDE accounts for less than 2% of systemic arterial emboli.² According to Johnson, the diagnosis of PDE should be considered in the presence of (i) a systemic arterial embolus does not arise from the left side of the heart, (ii) venous thrombosis and/or pulmonary embolus, and (iii) an intracardiac communication that permits a right-to-left shunt.³

To our knowledge, this is the only fifth case reporting an occlusion of the aortic bifurcation due to PDE.⁴⁻⁶ One study has suggested that upper and lower extremities arteries (55%) and cerebral arteries (37% to 50%) represent the most frequent sites of ischaemia due to PDE, with those of visceral and

coronary arteries being even rarer (6% to 9% and 7% to 9% respectively).⁷ In other studies, the most common site of arterial embolus due to PDE was not the cerebral circulation, but the peripheral arteries.^{4,6,8,9} Moreover, they noted a high frequency of emboli involving the mesenteric circulation.

In our case, deep venous thrombosis (DVT) appears to have been the source of paradoxical venous thromboemboli. In the absence of a procoagulant disorder, the recent long haul flight, dehydration and vein compression caused by the pelvic mass, likely contributed to the DVT in this patient. Atrial septal abnormalities like PFO and ASA have been also considered a nidus for local thrombus formation with subsequent embolization.^{10,11}

Although no randomized studies have investigated the long term efficiency of PFO closure devices on recurrence of embolic events, a recent systematic review of the literature has shown that a substantial proportion of recurrent thromboembolic events could be prevented by implantation of a PFO closure device compared with medical therapy (warfarin, or acetylsalicylic acid).¹² The subgroups most likely to profit from this procedure need to be defined.

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