LETTER TO THE EDITOR

A case of isolated splenic infarction associated with a foramen ovale and an interatrial septal aneurysm

Infarctus splénique isolé associé à un foramen ovale et à un anévrisme du septum interauriculaire

Introduction

Splenic infarction is a rare cause of abdominal pain. We describe an unusual case of splenic infarction in a patient with both a foramen ovale and an interatrial septal aneurysm. Although we did reach a consensus about the appropriate medical strategy, the patient underwent a percutaneous closure of the foramen ovale with insertion of a self-expandable septal occluder device. Antiplatelet agents were administered after surgery and there were no complications at the six-month follow-up.

Case report

On July 23, 2006, a 52-year-old man was admitted to the emergency ward because of epigastric pain associated with emesis. The patient had been fine until the day before, when he had a sudden onset of epigastric pain. Pain was constant and severe, and radiated to the left upper quadrant. There was no history of diarrhea, hematochezia, melena, contact with other illnesses or recent trauma. The patient’s past medical history included a sleep apnea syndrome for three years, renal colic, an appendectomy and a Hartman procedure for sigmoid diverticulitis. The patient had smoked for 15 years and had low alcohol consumption (20–40 g per week). He was not taking any medications. There was no family history of cerebral vascular disease or coronary artery disease. There had been no change in bowel habits. The pain worsened when the patient breathed deeply. On initial physical examination the patient presented mild painful distress, but no mass or hernia was detected. He weighed 128 kg and was 183 cm tall. His temperature was 36.9 °C, pulse rate 71 per minute, respiratory rate 20 per minute, and the blood pressure 160/90 mm Hg. Chest examination was clear and the cardiac examination did not show any tachycardia, murmure, gallop or rub. Abdominal examination showed an increase in pain during deep breathing in the epigastric and left upper quadrant, but there was no guarding in this area. Bowel sounds were normal. No abdominal aortic sounds were heard and all the pulsus were present. Rectal examination was normal. Laboratory evaluation demonstrated a white blood cell count of 11,200 with 8700 neutrophils, hemoglobin level was 14.8 g/dL and hematocrit was 47%. Serum levels of electrolytes, bilirubin, alkaline phosphatase, lipase, and creatinine were normal. Total protein level was normal and serum albumin was 45 g/L. An abdominal ultrasound showed a normal liver with sludge in the gallbladder and no abnormalities in the spleen, the kidneys or the pancreas. A computed tomographic scan of the abdomen and pelvis showed a focal wedge-shaped area of decreased density in the spleen with a thrombus of the splenic artery near its origin, consistent with a splenic infarct (Fig. 1). No atherosclerotic calcifications were seen in the abdominal aorta. Abdominal ultrasound doppler examination confirmed thrombus in the splenic artery with no other thrombi in the abdominal area.

The patient was then tested for the following: inherited thrombophilic factors including primary myeloproliferative disorders (myelogram), protein C, protein S, or antithrombin deficiency; lupus anticoagulant and antiphospholipid antibodies; paroxysmal nocturnal hemoglobinuria; activated protein C resistance or factor V Leiden mutation, prothrombin gene and JAK2 V617F mutations. All tests were normal. An electrocardiogram and a chest-X ray were also normal. Transesophageal echocardiography showed an interatrial septal aneurysm and a patent foramen ovale but no valvulopathy, no thrombus and normal cardiac function was. Magnetic resonance imaging of the brain was normal. The patient was promptly placed on anticoagulation therapy initially with fondaparinux sodique (Arixtra®) 10 mg/d subcutaneously, followed by fluidione (Previscan®). A marked decrease in abdominal pain was observed a few days later. On December 19, 2006, the patient underwent a percutaneous closure of the patent foramen ovale with insertion of a 35 mm self-expandable Amplatzer septal occluder device (AGA Medical Corp., Golden Valley, Minnesota). The device was inserted using a femoral approach, a transvenous sheath was moved forward and across the foramen into the left atrium, where a folded disk was expanded and pulled back, opposing the closed primum and secun-
symptoms or if a splenic abscess or hemorrhage occurs [3]. However, splenectomy can be performed for persistent pleuretic component. Treatment is generally non-surgical.

Symptoms of splenic infarct include fever, chills and abdominal pain radiating to the left shoulder occasionally with a hypodense area (arrow) in the spleen with persistent capsular enhancement. Tomodensitométrie abdominale (temps veineux portal) montrant un infarctus splénique avec une grande zone splénique hypodense (flèche) et un rehaussement capsulaire persistant.

dum septa. Then, a right-sided-disk was deployed and the two disk device was released. Aspirin (Kardegic®) 75 mg/d and clopidogrel (Plavix®) 75 mg/d were prescribed for three months and thereafter aspirin was continued alone following implantation. A clinical and echocardiographic follow-up was performed at one, three and six months and revealed no particular complications.

Discussion

We report a rare case of isolated splenic infarction revealed by abdominal pain and consecutive to a patent foramen ovale with an interatrial septal aneurysm. The causes of splenic infarction vary with age. Patients under 40 usually have an associated haematologic disorder such as sickle cell disease, while those over 40 often present an embolic event [1]. The conditions associated with splenic infarct have been classified as: non malignant or malignant haematologic disorders (haemoglobin SC disease, polycythemia vera, lymphoma...), infectious diseases (mononucleosis, AIDS, sepsis...), blunt abdominal trauma, cardiac disorders (valvular disease, endocarditis), vascular disorders such as aortic atherosclerosis, drugs (cocaine, erythropoietin...), and other miscellaneous causes (Gaucher disease, pancreatitis...). In older patients, two potential sources of a thrombotic event for splenic infarction have been identified: embolisation from the atherosclerotic aorta or thromboemboli from a patent foramen ovale [2]. Symptoms of splenic infarct include fever, chills and abdominal pain radiating to the left shoulder occasionally with a pleuretic component. Treatment is generally non-surgical. However, splenectomy can be performed for persistent symptoms or if a splenic abscess or hemorrhage occurs [3]. In half of the patients, anemia and a leucocytosis may occur [1]. Our patient only had abdominal pain and leucocytosis.

In our patient the cause of splenic infarct was a patent foramen ovale, a condition encountered in about 25% of unselected patients [4]. Patent foramen ovale is a vestige of the fetal circulation and occurs because the primum and secundum septa do not fuse postnatally. Persistence of the one-way flap valve overlaying the fossa ovalis causes right to left blood flow when right atrial pressure exceeds left atrial pressure. This latter condition is observed in obstructive sleep apnea syndrome during nocturnal apnea contributing to a more prevalent diagnosis of patent foramen ovale in patients with sleep apnea syndrome than in control patients [5]. The association of a patent foramen ovale with cryptogenic stroke has consistently been reported in patients under 55. Moreover, patients with both a patent foramen ovale and an interatrial septal aneurysm who have had a cryptogenic stroke have a higher risk of recurrent stroke (annual risk close to 4%) while taking aspirin than patients with no septal abnormality or either septal abnormality alone [6]. When an aerated saline is injected into a vein, the severity of the shunt can be semi-quantitatively graded according to the maximum number of bubbles seen in the left atrium after three cardiac cycles. In addition transesophageal echocardiography reliably excludes other potential sources of embolism. The typical symptoms of patent foramen ovale are migraine, headache and stroke but, as in our case, most of the patients do not have these symptoms. The typical consequences of patent foramen ovale are paradoxical embolism of the thrombus, air or fat with a subsequent cerebrovascular accident (ischemic brain lesions on MRI), refractory hypoxemia, and platypnea-orthodeoxia syndrome where patients develop arterial desaturation in the upright position due to orthostatic accentuation of a right to left shunt across the patent foramen ovale [7]. Paradoxical embolism occurs when venous thrombotic material crosses through the patent foramen ovale into the arterial circulation but a thrombus has rarely been documented in the venous system. Likewise, the role of coagulation disorders and prothrombotic genetic polymorphisms remains poorly defined.

Secondary prevention for patients with a patent foramen ovale or an interatrial septal aneurysm who have had a stroke is a matter of considerable debate. Some data suggest that the degree of right-to-left shunting, the presence of an atrial septal aneurysm, and a patent foramen ovale tunnel length may increase the risk of thromboembolic events and may represent the cohort of patients at the greatest benefit/risk ratio of receiving a closure device. A systematic review suggests that a substantial proportion of recurrent strokes may be prevented by implanting a closure device rather than conservative long-term medical therapy with antiplatelet agents or oral anticoagulants [8]. The overall one-year rate of recurrent neurological thromboembolisms with transcatheter intervention was 0 to 4.9% compared to 3.8 to 12% for medical management suggesting a lower risk for stroke with the interventional strategy although there are no randomized clinical trials comparing these two approaches [8]. Although the traditional approach involves open thoracotomy the perioperative risks and mortality are higher than with percutaneous closure devices. In our case, we decided to close the patent foramen ovale because the patient had several risk factors for recurrent stroke, in particular the association of both patent foramen ovale and an
interatrial septal aneurysm with a sleep apnea syndrome. To date, available data are still too scarce to make specific recommendations for using antiplatelet therapy or anti-vitamin K after percutaneous closure devices.

Our case illustrates the uncommon occurrence of an isolated splenic infarction consecutive to both a patent foramen ovale and an interatrial septal aneurysm, in a patient with a sleep apnea syndrome, a condition that is known to contribute to embolic events. Moreover, this case report emphasizes the need for a complete investigation in case of splenic infarction to elucidate the cause of this rare disease.

References


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