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Spontaneous splenic artery aneurysm rupture in a 38-year old female: a case report

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SUMMARY: **Spontaneous splenic artery aneurysm rupture in a 38-year old female: a case report.**

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Splenic artery aneurysm (SAA) is a rare and extremely difficult diagnosis. A rare case of a ruptured SAA in a 38-year old female, firstly treated with endovascular embolization and then with splenectomy, is presented. A 38-year old female presented to the emergency department with epigastric pain and fainting episodes. Direct catheter angiography revealed a ruptured SAA and distal, as well as proximal coil embolization was performed. Due to abdominal compartment syndrome the patient underwent open surgery with splenic artery ligation and splenectomy. Postoperative she showed signs of sepsis and was treated with i.v. fluids, steroids, packed red blood cells, platelets, fresh frozen plasma and antimicrobial treatment. Additionally, a multidrug resistant Acinetobacter baumanni was yielded from the urine culture. She had a satisfactory recovery. She is followed up a total of 5 years with no signs of overwhelming post-splenectomy infection syndrome. Direct catheter angiography is a very helpful option in diagnosis, as well as treatment, but a close monitoring after embolization is essential. Furthermore, post-splenectomy sepsis is a severe disease with high mortality rates that requires immediate appropriate treatment.

KEY WORDS: Splenic artery aneurysm - Coil embolization - Post-splenectomy sepsis - Overwhelming post-splenectomy infection.

Introduction

Splenic artery aneurysm (SAA) is a dilatation of more than 1cm of the splenic artery (1). It is a rare and extremely difficult diagnosis. Histopathologically, SAAs are classified into two types: true and pseudo-aneurysms. Pseudoaneurysms are expansions of the artery with focal disruption of the arterial wall, whereas true aneurysms are expansions of all wall layers (1, 2).

Despite being rare, SAAs are important due to their potentially life-threatening complications, such as spontaneous intraperitoneal rupture, rupture into the nearby hollow organs, and fistulization into the pancreatic duct. The pathogenic mechanisms of the SAA are not entirely clear. Trauma, portal hypertension, atherosclerosis, arterial degeneration, pregnancy, as well as the female gender are known risk factors (1, 3).

Spontaneous rupture is reported in 2-10% of cases and it is accompanied with up to 40% mortality rate. Nowadays, the endovascular embolization of the splenic artery is becoming more favored due to its acceptable technical success and low morbidity rates. However, open abdominal surgery remains the gold standard treatment (3, 4).

Post-splenectomy sepsis is a serious disease that can progress from a mild flu-like illness to fulminant sepsis in a short time period. Although relatively rare, it has a high mortality rate with delayed or inadequate treatment (5).

A rare case of a ruptured SAA in a 38-year old female, firstly treated with endovascular embolization and then with splenectomy, is presented. Postoperatively the patient showed also symptoms and signs of overwhelming post-splenectomy infection (OPSI).

Case report

A 38-year old female, with clear medical history, presented to the emergency department of a secondary ter-

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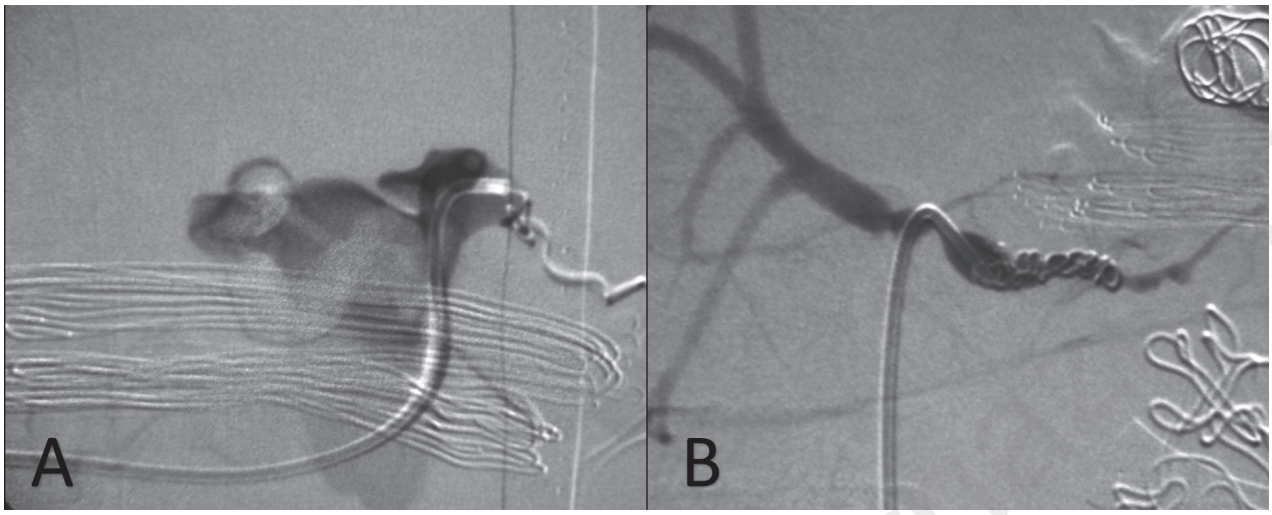

Figure 1 - Direct catheter angiography (femoral approach). A) Active bleeding from the splenic artery. B) Post-embolization.

tiary hospital about 100 km away from Athens, Greece due to sudden epigastric pain, weakness and two fainting episodes. The patient was confused and hemodynamically unstable [blood pressure (BP)= 70/20 mmHg and heart rate= 120 bpm], while her initial laboratory investigation showed hematocrit 29%, hemoglobin 10.4 g/dL and the ultrasound (U/S) free intraabdominal fluid. Additionally, the pregnant urine-test was negative. She received intravenous (i.v.) fluids and an emergency Computer Tomography scan (CT) was performed showing intraabdominal bleeding of unknown origin.

At that point she underwent emergency open abdominal surgery. The origin of the bleeding was not found. Therefore, the intubated patient was transferred to the "Sismanogleio" general hospital of Athens, Greece for further diagnostic and treatment.

At the "Sismanogleio" General Hospital, angiography using a femoral approach was performed and a ruptured SAA was found. She was treated with proximal and distal coil embolization. (Figure 1). Until that point the patient had been transfused with 20 blood units. She was transferred to the intensive care unit (ICU) for observation. After the embolization she was stabilized (BP=135/90 mmHg and heart rate=80 bpm). Due to the fact that she had developed abdominal compartment syndrome (intra-abdominal pressure =28 mmHg) she underwent the a few hours later one more time emergency surgery. During the open surgery about three litres of blood were removed from the abdomen. Furthermore, active micro-bleeding was revealed and therefore splenic artery ligation, at its origin the coeliac axis, aneurysmatectomy and splenectomy were performed.

The patient returned to the ICU. From the 2nd until the 9th postoperative day she was febrile (38° C), stable, lethargic and had cephalalgia. Her laboratory investigation showed white blood cell count between 30000- and 16000/mm^3 and c-reactive protein between 230 and 40mg/L. Additionally, blood, sputum and urine cultures were obtained. She was treated empirically for post-splenectomy sepsis with i.v. fluids, steroids, packed red blood cells, platelets, fresh frozen plasma and antimicrobial treatment (linezolid, ciprofloxacin and metronidazole). The obtained urine culture yielded a multidrug resistant (MDR) *Acinetobacter baumanni*, sensitive to colistin. The i.v. antimicrobial treatment was then switched to colistin.

The patient returned to the surgical ward on the 9th postoperative day. She was stable, alert and afebrile. She continued the i.v. antimicrobial treatment with colistin, ciprofloxacin and metronidazole during hospitalization. Furthermore, she received the 7-valent protein-conjugated pneumococcal vaccine, the *Hemophilus influenzae* type B vaccine, and the meningococcal vaccine. She had a satisfactory recovery and was discharged on the 19th postoperative day.

The patient received for two years post-splenectomy per os phenoxymethylpenicillin. She showed no drug-related side effects. She is followed up at the out-patient clinic for a total of 5 years, without any signs or symptoms of overwhelming post-splenectomy infection syndrome, while revaccination is planned for next year.

Discussion

The present case exhibits the difficulty in diagnosing a ruptured SAA, as well as the difficulties in handling a post-splenectomy patient. The majority (80%) of SSAs

are asymptomatic. Unspecific symptoms may be present in the remaining cases, such as abdominal pain in the epigastrium or the left upper quadrant, nausea, vomiting, anorexia hematemesis, hematochezia, melena and anemia (1, 3, 4).

Spontaneous rupture is presented to only 2-10% of SAAs and it is associated to high mortality rates (up to 40%). Regarding the histopathological type of the SAA, pseudo-aneurysms are caused usually by inflammatory conditions, or trauma and iatrogenic lesions and have a higher rupture-risk when compared to true aneurysms (1, 4, 6). Since the reported patient had no such medical history, it is believed that she suffered from a true SAA.

Radiographically it is also difficult to establish this diagnosis. The gold standard method is direct catheter angiography, which is both diagnostic and therapeutic. Other imaging techniques including ultrasound, CT and magnetic resonance imaging are also useful in diagnosis, with lower sensitivity, and follow-up monitoring (7-9).

The present patient had at the emergency department of the tertiary hospital a U/S and a CT without i.v. contrast, which established the intraabdominal bleeding but were unable to find its source. The final diagnosis was made through the direct catheter angiography, while at the same time proximal and distal coil embolization was performed.

A successful embolization ranges from 55 to 100%. Complications include: post-embolization syndrome, presenting with fever, pain and leukocytosis; splenic and intestinal infarcts; pancreatic or splenic abscess and migration of embolization material to non-targeted arteries (10, 11). The coil embolization in the presented patient was considered successful, since she was stabilized right after the procedure. However, during the open surgery, due to the developed abdominal compartment syndrome, active micro-bleeding was found.

Splenectomized patients are at risk for uncontrolled infections with poor outcomes (5). The predominant organisms responsible for overwhelming sepsis in splenectomized patients are pneumococci (50%), meningococci, and *Hemophilus influenza*. OPSI is a dangerous process that carries a high mortality rate (12, 13).

The spleen has 2 significant immunologic functions: to filter the blood of pathogenic bacteria and fungi and to shelter or sustain IgM opsonin-producing memory B cells. It has been customary to attempt immunization either 2 weeks before or 2 weeks after splenectomy to enhance immunogenicity (14). Vaccines available for the most common organisms include the 23-valent pneumococcal polysaccharide vaccine, a 7-valent protein-conjugated pneumococcal vaccine, the *Hemophilus influenzae* type B vaccine, and the meningococcal vaccine (12, 13). The presented patient received vaccination on the 12th postoperative day. Additionally, the Centers for Disease Control and Prevention in the US recommends revaccination for the prevention of overwhelming postsplenectomy infection every 6 years (15). According to these guidelines, revaccination is planned for the next year.

The patient presented with signs and symptoms of OPSI. However, no pathogen was yielded from the blood-culture. Although encapsulated organisms are the most virulent pathogens to splenectomized patients, according to the literature OPSI is pathogen-defined in only about 75% of cases. In the reported case only the urine-culture yielded a MDR *Acinetobacter baumanni*, a pathogen of hospital origin. Additionally, the mortality rate of OPSI is 50%-70% despite aggressive therapy that includes intravenous fluids, antimicrobial treatment, vasopressors, steroids, heparin, packed red blood cells, platelets, cryoprecipitates, and fresh frozen plasma (16). The patient was treated according to these guidelines at the ICU and showed improvement after 7 days of treatment. At that point she was able to return to the surgical department. It is also of note that although durations between splenectomy and onset of OPSI range from less than 1 week to more than 20 years, the appearance in the first post-operative days is considered extremely rare (17).

It is crucial that splenectomized patients be educated about the nature of this disease and seek medical evaluation early rather than to perform self-diagnosis and self-therapy (18). Finally, patient education represents a mandatory strategy for preventing OPSI. Studies have shown that from 11% to 50% of splenectomized patients remain unaware of their increased risk for serious infection or the appropriate health precautions that should be undertaken. Unfortunately, regarding guideline compliance in multiple, developed countries is only 60% to 75% (19).

Conclusions

Ruptured SAAs are extremely rare and very difficult to diagnose. The physician must keep a keen eye, especially for hemodynamically unstable patients with left upper quadrant or epigastric abdominal pain. Direct catheter angiography is a very helpful option in diagnosis, as well as treatment. A close monitoring after embolization is essential. The surgeon must be standby for rebleeding cases.

OPSI is a severe disease with high mortality rates that requires immediate appropriate treatment. Splenectomized patients are at high risk for infections. The proper education of patients, as well as physicians seems to be a crucial factor regarding the management and overall survival of these patients.

Authors' contributions

CK, EP, NN, NZ, AT for the literature search and analysis, and manuscript writing. EP and GV for the final manuscript revision. All authors have read and approved the final manuscript.

Disclosure of interest

The Authors report no conflict of interest.

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