# Splenic arteriovenous fistula – late complications of splenectomy

W. Woźniak<sup>1</sup>, R.K. Mlosek<sup>2</sup>, T. Miłek<sup>1</sup>, P. Myrcha<sup>1</sup>, P. Ciostek<sup>1</sup>

(1) 1st Department and Clinic of General and Vascular Surgery, (2) Department of Diagnostic Imaging, 2st Faculty of Medicine, Medical University of Warsaw, Poland.

#### Abstract

This is a case report of a 30-year-old female patient with a splenic arteriovenous fistula of rare, atypical clinical course. The patient was admitted to the hospital due to strong abdominal pains and fever. 13 years earlier, the patient underwent splenectomy due to post-traumatic rupture of the spleen. On imaging examinations prior to surgery (ultrasonography, CT), a splenic arteriovenous fistula was diagnosed. The patient was operated on due to increasing abdominal pain. The fistula was closed by splenic artery and splenic vein banding during the course of laparotomy. This case report will be discussed based on literature review. (Acta gastroenterol. belg., 2011, 74, 465-467).

**Key words**: portal hypertension, splenic arteriovenous fistula, splenectomy, surgery, complications.

## Introduction

The spleen is a relatively commonly injured organ in abdominal traumas, partly because of its location in the peritoneal cavity and its parenchymatous structure. As is often the case, life-saving splenectomy needs to be performed promptly to stop massive bleeding. Spleen excision may lead to the development of thrombocytosis, and increases the risk of thromboembolic complications (portal vein thrombosis, mesenteric venous thrombosis) and overwhelming postsplenectomy sepsis. Remote post-splenectomy complications also include rare cases of splenic arteriovenous fistulae.

Splenic arteriovenous fistula is considered to be one of the rare causes of prehepatic portal hypertension and was first described by Carl Weigert, a German surgeon and pathologist, in 1886.

Approximately 100 cases of this condition have been reported to date.

### Case report

A 30-year-old female patient was hospitalized at the 1st Department and Clinic of General and Vascular Surgery, 2nd Faculty of Medicine, Medical University of Warsaw due to abdominal pain lasting for several days, located in the left hypochondriac region and middle-abdominal area. A few hours before hospitalization, the symptoms aggravated, with accompanying nausea, vomiting and fever up to 39°C. The patient had a history of two surgeries: splenectomy due to post-traumatic rupture of the spleen 13 years ago, and appendicectomy due to appendicitis 12 years ago. On the day of hospital

admission, the patient's condition was satisfactory, with symptoms of minor dehydration. Physical examination revealed tachycardia up to 102 beats per minute, normal arterial pressure (120/80), slightly bloated abdomen, abdominal pain in the hypochondriac region and middleabdominal area, with maintained peristalsis and no peritoneal symptoms. Biochemical tests showed a slight anaemia (Hbg 11.5g%, HCT 34%, RBC 3 920,000/mm<sup>3</sup>) and normal leukocyte and platelet count (WBC 9,300/mm<sup>3</sup>, PLT 293,000/mm<sup>3</sup>). The levels of electrolytes, glucose, urea, creatinine, transaminases, bilirubin, amylase, protein and coagulation parameters revealed no deviations from normal ranges. Ultrasonography revealed  $48 \times 39$  mm accessory spleen and  $80 \times 45$  mm vascular malformation adjacent to it, resembling an arteriovenous fistula on Colour Doppler examination. The ultrasound examination revealed the absence of free liquid in the abdomen. CT angiography showed an arteriovenous fistula between splenic artery and splenic vein in a bed left after spleen excision. The splenic vein was a varicose vein enlarged to 37 mm, and the remaining vessels of the portal venous system showed no signs of enlargement (Fig. 1, 2). No pathologies were found in the arterial system of the abdomen. Upper gastrointestinal endoscopy showed no deviations from normal. The abdominal pain got worse, the patient presented a poor response to analgesics, and became eligible for laparotomy. The peritoneal cavity was opened with a midline incision through the existing scarf. The splenic vein was found to be significantly enlarged to up to 4 cm, adhering with the greater curvature of the stomach and pulsating on palpation. Next, after the omental bursa had been opened, splenic artery and splenic vein were made visible in the section where the diameters of both vessels were normal. Moving along both vessels in the direction from the left side, both vessels were shown to be directly connected. After two independent bandings of splenic artery and splenic vein, the fistula was eliminated and separated with a section of fatty tissue. Two accessory spleens were found and left in the bed after the spleen excision. The surgery was completed with drainage of the left subphrenic region. No post-surgery

Correspondence to: Witold Woźniak, 1<sup>st</sup> Department and Clinic of General and Vascular Surgery, 2<sup>st</sup> Faculty of Medicine, Medical University of Warsaw, Kondratowicza-street 8, 03-242 Warsaw, Poland. E-mail: drww@neostrada.pl

Submission date: 06/07/2010 Acceptance date: 17/02/2011 W. Woźniak et al.

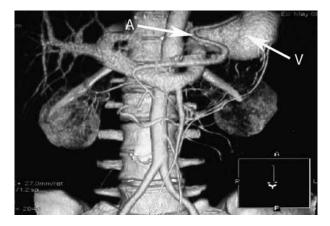


Fig. 1. — Abdominal angiography computed tomography reconstruction. Coronal reconstruction of splenic arteriovenous fistula (A-splenic artery, B-splenic vein).

complications were reported. On the 5<sup>th</sup> day after the surgery, the patient was discharged home with a healthy healing wound. Three months after the surgery, CT image of abdominal organs in a follow-up CT examination showed no abnormalities.

#### **Discussion**

Splenic arteriovenous fistula (SAVF) is a relatively rare cause of prehepatic portal hypertension. Its aetiology is not completely understood. According to the available literature data, SAVF can be caused by congenial, post-traumatic or iatrogenic factors (1-5). One third of all fistulae cases manifested in infancy and childhood, with concurrent Rendu-Osler-Weber syndrome or Ehlers-Danlos syndrome. The remaining two third occur in adults after vein injury by a splenic artery aneurysm or in case of post-traumatic or iatrogenic spleen injury (2,5). According to Blum, 37% of all SAVF cases occur in patients up to 35 years of age, and only 21% in patients above 60 years of age. SAVF occurs significantly more frequently in women (69%). Post-traumatic SAVF occurs in 45% patients, and post-splenectomy SAVF occurs in 15% (6). The high incidence of SAVF in women has been associated with predisposition of pregnant women and multiparas to splenic artery aneurysms. Similarly, a significant percentage of congenial and post-traumatic fistulae may explain the relatively rare incidence of SAVF in elderly patients.

Splenic arteriovenous fistula may manifest with abdominal pain after meals and physical activity, diarrhoea, malabsorption, oesophageal varices, bleeding in the digestive tract, abdominal dropsy, dyspnoea, hepatomegaly, and splenomegalia. Symptoms of liver damage and right heart failure may also occur, but are less likely. These symptoms are associated with increased portal hypertension caused by damage to the liver parenchyma and a significant increase of blood flow volume in portal vessels. Increased portal circula-



Fig. 2. — Abdominal angiography computed tomography reconstruction. Axial reconstruction of splenic arteriovenous fistula (A-splenic artery, B-splenic vein).

tion through the liver stimulates liver fibrosis, which in turn intensifies the already increased resistance in the portal system. This mechanism causes progressive liver failure. On the other hand, due to increased portal circulation, collateral circulation may occur, which quickly leads to the development of oesophageal varices. Increased portal pressure is also responsible for malabsorption and diarrhoea. The majority of splenic arteriovenous fistulae manifested by oesophageal variceal haemorrhages and massive abdominal dropsy with symptoms of right heart failure (1,2,4,7,8).

In this case study, the fistula developed after a trauma experienced in the past, after a splenectomy performed several years before. Apart from abdominal pain, no other symptoms occurred. This atypical course of the disease may be perhaps attributed its short duration, as the symptoms started several days before the patient's hospitalization. Detailed imaging diagnostics revealed abnormalities at the initial stage of the disease, before any complications occurred. Abdominal pain may be attributed to the splenic vascular ectasia and the related pressure symptoms.

As mentioned earlier in the text, there have been rare cases of post-splenectomy SAVF reported (9-15). According to literature data, the time of occurrence of SAVF varies from a few to dozens of years. In this case report, the patient underwent splenectomy 13 years earlier. It is extremely difficult to determine the cause of SAVF occurrence. The majority of researchers claim that SAVF develops as a result of common banding or puncturing of the splenic artery and splenic vein. Vargün (16) claims otherwise, and presents the results of spleen hilum closing with a linear stapler during laparoscopic splenectomy, without isolating the artery and the vein. Vargün examined the bed left after spleen excision in an ultrasonographic examination, but no fistula was found. However, the follow-up period was short and lasted approximately two years. In our case, apart from two independent vessel banding procedures, the vessels were separated with a strip of fatty tissue, to protect it from fistula recurrence.

The treatment of SAVF consists of splenic artery and vessel banding and splenectomy. With the progress of radiology and endovascular surgery during the last few years, SAVF can be now treated with minimally-invasive methods. Endovascular SAVF is possible if the fistula is located inside the parenchyma, following selective embolization of the respective ramifications of the splenic artery. If the splenic artery trunk is closed, spleen infarction may occur. Large numbers of endovascular repairs of SAVFs have been recently reported in literature (3,9,17-19). In patients who developed fistula with wide entrance, embolic material may relocate to the portal venous system and cause portal vein thrombosis. As a result, traditional surgical repair remains the optimal treatment method of SAVF. The patient underwent traditional surgical repair because of the wide entrance between the splenic artery and the splenic vein. If the entrance is narrow, an endovascular method may be considered, as the patient had a history of splenectomy. This particular treatment method was also selected due to vague clinical picture, which might suggest other pathologies that could require immediate measures. A traditional surgery is recommended if splenectomy needs to be performed, simultaneously to distal pancreatectomy or if endovascular treatment cannot be applied (20-22).

#### References

- JOHNSTON G.W., GIBSON J.B. Portal hypertension resulting from splenic arteriovenous fistulae. Gut, 1965, 6: 500-502.
- MC CLARY R.D., FINELLI D.S., CROKER B., DAVIS G.L. Portal hypertension secondary to a spontaneous splenic arteriovenous fistula: case report and review of the literature. Am. J. Gastroenterol., 1986, 81: 572-575.
- SIABLIS D., PAPATHANASSIOU Z.G., KARNABATIDIS D., CHRISTEAS N., KATSANOS K., VAGIANOS C. Splenic arteriovenous fistula and sudden onset of portal hypertension as complications of a ruptured splenic artery aneurysm: Successful treatment with transcatheter arterial embolization. A case study and review of the literature. World J. Gastroenterol., 2006, 12: 4264-4266.

- SHLEAPNIK M., SHPITZ B., SIEGAL A., DINBAR A. Bleeding esophageal varices and portal hypertension caused by arteriovenous fistula of splenic artery. HPB Surg., 1990, 3: 53-57.
- DI LECCE F., VIGANÒ P., BUSANI M., CAFORIO M., CALEFFI G., FRANCIA L., GERARD L., GHIDONI S., PATUZZO E., PULICA C. Splenic artero-venous fistula and portal hypertension. A case report with review of the literature. G. Chir., 2006, 27: 328-330.
- BLUM A., BAZIN C., KLEIN M., KAMINSKY P., BOISSEL P., CLAUDON M., BECKER S., REGENT D. Splenic arteriovenous fistula. Apropos of 2 cases and review of the literature. *J. Radiol.*, 1994, 75: 245-252.
- STRATE T., MANN O., KÖNIG A., BACHMANN K., IZBICKI J.R. Arterio-venous aneurysm of splenic vessels-a rare differential diagnosis for cardio-circulatory symptoms. *Zentralbl. Chir.*, 2007, 132: 77-80.
- WILLIAMS D.B., PAYNE W.S., FOULK W.T., JOHNSON C.M. Splenic arteriovenous fistula. *Mayo Clin. Proc.*, 1980, 55: 383-386.
- MADSEN M.A., FREVERT S., MADSEN P.L., EIBERG J.P. Splenic arteriovenous fistula treated with percutaneous transarterial embolization. Eur. J. Vasc. Endovasc. Surg., 2008, 36: 562-564.
- SILBERZWEIG J.E., MATISSEN D., KHORSANDI A.S. Splenic arteriovenous fistula with pseudoaneurysm as a complication of splenectomy. AJR Am. J. Roentgenol., 2006, 187: 548-549.
- GARTSIDE R., GAMELLI RL. Splenic arteriovenous fistula. J. Trauma, 1987, 27: 671-673.
- LACOMBE M., HANNOUN L. Arteriovenous fistula of splenic vessels after splenectomy. J. Chir. (Paris), 1984, 121: 159-162.
- HYDE G.L. Postsplenectomy arteriovenous fistula causing portal hypertension. J. Ky. Med. Assoc., 1979, 77: 113-115.
- BAYINDIR S., GRAEBNER H. Arteriovenous fistula between splenic artery and vein following splenectomy. A case report. Fortschr. Geb. Rontgenstr. Nuklearmed., 1966, 105: 279.
- FONTAN F., GALY J.C. Splenic arteriovenous aneurysm after splenectomy. *Arch. Mal. Appar. Dig. Mal. Nutr.*, 1963, 52: 580-582.
- VARGÜN R., GÖLLÜ G., FITÖZ S., YAGMURLU A. En-bloc stapling of the splenic hilum in laparoscopic splenectomy. *Minim Invasive Ther. Allied Technol.*, 2007, 16: 360-362.
- HUNG C.F., TSENG J.H., LUI K.W., WAN Y.L., TSAI C.C., SHEM C.H., WU C.S. Intractable oesophageal variceal bleeding caused by splenic arteriovenous fistula: treatment by transcatheter arterial embolization. *Postgrad. Med. J.*, 1999, 75: 355-357.
- OGUZ B., CIL B., EKINCI S., KARNAK I., AKATA D., HALILOGLU M. Posttraumatic splenic pseudoaneurysm and arteriovenous fistula: diagnosis by computed tomography angiography and treatment by transcatheter embolization. J. Pediatr. Surg., 2005, 40: 43-46.
- KAKIZAWA H., TOYOTA N., NAITO A., FUKUDA H., ITO K. Superselective transcatheter arterial embolization of a traumatic splenic arteriovenous fistula. *Radiat. Med.*, 2004, 22: 364-367.
- BROTHERS T.E., STANLEY J.C., ZELENOCK G.B. Splenic arteriovenous fistula. Int. Surg., 1995, 80: 189-194.
- PONS V., NAOURI C., NAOURI A., MENECIER P., DENIS J.M., LEVENQ P. Diagnosis and treatment of 2 forms of ruptured aneurysm of the splenic artery. *Gastroenterol. Clin. Biol.*, 1998, 22: 739-741.
- DUTTA U., BAPURAJ R., YADAV T.D., LAL A., SINGH K. Mesenteric ischemia and portal hypertension caused by splenic arteriovenous fistula. *Indian J. Gastroenterol.*, 2004, 23: 184-185.