Surgical Treatment of Splenic Artery Pseudoaneurysm with Digestive Tract Communication – Presentation of Two Cases

Mihai Adrian Eftimie, Oana Maria Stanculea, Leonard David, Vasile Lungu, Simona Dima, Iulian Mosteanu, Luiza Tirca, Irinel Popescu

Center for General Surgery and Liver Transplantation “Dan Setlacec”, Fundeni Clinical Institute, Bucharest, Romania

Corresponding author:
Mihai Adrian Eftimie, MD
Center for General Surgery and Liver
Transplantation Dan Setlacec
Fundeni Clinical Institute, Bucharest
Romania
E-mail: mihai_eftimie84@yahoo.com

Rezumat

Tratamentul chirurgical al pseudoanevrismelor arterei splenice ce prezintă comunicare cu tracul digestiv - două prezentări de caz

Spleno-pancreatectomia efectuată pentru tratamentul pseudo-anevrismelor arterei splenice localizate la nivelul unui pseudochist pancreatic ce comunică cu tubul digestiv (stomac sau colon) reprezintă una din indicațiile rare, puține astfel de proceduri fiind descrise în literatură. Tratamentul pseudo-anevrismelor peri-pancreatice este complex și dificil. Tratamentul chirurgical este curativ și poate include rezecția altor organe. Intervențiile chirurgicale pot fi executate în condiții de urgență, dacă pacientul prezintă sângerare cataclismică sau în condiții electivă în cazul în care diagnosticul se pune accidental sau dacă pacientul reușește să depășească episodul inițial de sângerare. În această lucrare prezentăm două cazuri de pseudochist pancreatic ce asociază pseudo-aneurism al arterei splenice și fistula cu tubul digestiv (comunicare pseudochisto-colonică și pseudochisto-gastrică). Amândoi pacienții sunt bărbați, cunoscuți cu pancreatită cronică și pseudochiste pancreatice. Primul pacient a fost operat electiv după investigații amănunțite. Cealaltă intervenție chirurgicală a fost efectuată în condiții de urgență pentru a controla un episod de sângerare cataclismică. Concluzionăm ca abordul chirurgical rămâne principala metodă pentru tratamentul acestor cazuri. Poate fi folosit ca primă linie de tratament sau secundar abordului endovascular.

Cuvinte cheie: splenopancreatectomie, pseudoaneurism, arteră splenică, rezecție pancreatică

Received: 20.03.2017
Accepted: 12.04.2017

Abstract

Splenopancreatectomy performed for pancreatic pseudo-cyst with splenic artery pseudo-aneurysm and communication with the digestive tract (stomach or colon) is a very rare indication and a small number of these procedures are described in literature. Managing peri-pancreatic pseudo-aneurysm is complex and can be challenging. Surgical treatment is of curative intent and can involve multiple visceral resections. Surgery can be performed in an emergency setting, if the patient presents cataclysmic bleeding, or in a planned manner if the pseudo-aneurysm is discovered incidentally or if the patient manages to overcome the initial bleeding. In this paper we present two cases of pancreatic pseudo-cysts with splenic artery pseudo-aneurysms and communication with the digestive tract (one with pseudo-cystic-colonic communication and the other one with gastric communication). Both patients were males, suffered from chronic pancreatitis and were known to have pancreatic pseudo-cysts. For the treatment of the first patient, surgery was performed in an elective setting, after intensive investigations. The other patient presented with cataclysmic bleeding and emergency surgery was performed in order to control the bleeding. We conclude that surgery remains the main option of treatment for these patients. It can be used as a first line of treatment or secondary to endovascular procedures.

Key words: spleno-pancreatectomy, pseudo-aneurysm, splenic artery, pancreatic resection

Introduction

Peri-pancreatic pseudo-aneurysms represent a rare clinical entity that is commonly associated with pancreatitis and pancreatic surgery. It is different from a real aneurysm. Its walls do not contain all of the components of a true artery, instead the wall is formed out of fibrotic tissue that expands, due to continuous aggression from the enzyme-rich peri-pancreatic fluid that is present inside the pseudo-cyst. The splenic artery is involved in up to 50% of the total number of pseudo-aneurysms. (1)

Preoperative evaluation is routinely performed by contrast enhanced CT exam. This examination provides conclusive information about the location of the pseudo-aneurysm, its dimensions and about the arterial branch involved.

There is no way to predict how a pseudo-aneurysm can evolve, but is a known fact that if they are left untreated can lead to life threatening bleeding. Most of these cases present with uncontrollable bleeding so increased mortality is associated with this pathology. If the first episode is self-controlled the risk of recurrent bleeding is present and

can occur days to years after the initial presentation. (2)

Their optimal treatment is controversial and no consensus is present. Only small series of cases are described in literature so a standard way of treatment isn't described. Surgery remains the gold standard of treatment. Selective trans-catheter arterial embolization and arterial stenting are modern ways of care but are these treatments of curative intent or a bridge therapy towards curative surgical treatment? (2)

Case report 1

55 year-old male with childhood history of accidental swallowing of acid, type II diabetes with oral medication since 2012, and a known episode of acute pancreatitis in December 2015, is investigated in a territorial hospital for hematochezia. At that time the patient presented with a hemoglobin level of 7 g/dl, so vivid hematic resuscitation was initiated. After the patient was stabilized a colonoscopy was performed and a protrusive hemorrhagic lesion was revealed at 40 centimeters from the anal verge. (*Fig. 1*) Biopsies did not show any signs of malignancy. A contrast enhanced computer

Figure 1. Colonoscopy - protrusive hemorrhagic lesion at 40 centimeters from the anal verge

tomography was performed and an ulcerated splenic artery pseudo-aneurysm located in a pancreatic tail pseudo-cyst with communication with the splenic flexure of the colon was found. (Figs. 2-4) After the initial episode, the patient did not show any signs of recurrent bleeding; the hemoglobin level remained constant so he was referred to our center for further investigations and treatment.

At time of admission the patient was stable, GCS of 15, no signs of bleeding, hemoglobin level of 13 g/dl with a hematocrit of 43.5%, leucocytes and thrombocytes were within normal range, normal coagulation, normal liver and renal tests. Tumoral markers CEA, CA19-9 and α FP were within normal values. A gastroscopy was performed and did not reveal any significant findings.

Our interventional radiological team evaluated the CT exam previously performed and decided to redo the contrast enhanced CT. Their conclusion was that the pseudo-cyst was located in close contact with the hilum of the spleen, between arterial and venous branches of the splenic vasculature and had no clear margins with the splenic flexure of the colon. The pseudo-aneurysm had one source, the splenic artery; had thin walls and no signs of active bleeding were present. (Figs. 2-4) A specific procedure of embolization or transarterial stenting was not feasible because of its close contact with the splenic vasculature and risk of spleen infarction.

The patient was operated after careful

Figure 2. Contrast enhanced CT scan – pseudo-aneurysm of the splenic artery in close contact with vascular pedicles of the spleen hilum

Figure 3. Contrast enhanced CT scan - pseudo-aneurysm of the splenic artery in close contact with vascular pedicles of the spleen hilum

Figure 4. Contrast enhanced CT scan – pseudo-cyst of the pancreatic body and tail in close contact/communication with the left colonic flexure

preoperative management and investigations. The intraoperative diagnosis was that of a pancreatic tail pseudo-cyst with communication with the splenic flexure of the colon and splenic artery pseudo-aneurysm. (Figs. 5-7) A medial to lateral spleno-pancreatectomy en bloc with resection of the splenic flexure of the colon was performed. The digestive tract was rebuilt using a side-to-side; two layer PDS 4-0 colonic anastomosis. The pancreas was transected using a 60 mm green cartridge Endo-GIA Stapler® with Tri-Staple Technology® after the splenic artery was identified at the superior border of the pancreas and ligated. The splenic vein was included in the stapling line along with the pancreatic tissue. No supplementary sutures were applied over the staples in order not to excessively manipulate the pancreatic stump. After surgery the patient was admitted in the postoperative-ICU.

On the first postoperative day, the patient developed a mild pancreatic reaction, with elevated blood values of amylase and lipase.

On the second postoperative day the patient presented bleeding from the pancreatic stump, exteriorized via the intra-peritoneal drainage tube. A hemoglobin drop of 2 g/dl was present.

The patient was operated and supplementary hemostasis was performed using Prolene 4-0 sutures that reinforced the stapled line of the pancreatic stump.

On the third postop day he developed a grade I pancreatic leakage exteriorized through the intraperitoneal drainage tube, in spite the fact that he received Sandostatin® and PPI treatment. There was no need for other specific treatments, the leakage remitted after 6 days. The patient was discharged on the 14 postoperative day, completely recovered.

The pathological exam did not reveal any signs of malignancy and confirmed the radiological and intraoperative findings.

A one-month postoperative physical examination was performed along with blood tests and an ultrasound exam. No significant findings were revealed.

Figure 5. Surgical specimen

Figure 6. Surgical specimen – pseudo-cysts with calcified walls and signs of bleeding from a ruptured pseudo-aneurysm of the splenic artery

Figure 7. Surgical specimen – point of communication between the pancreatic pseudo-cyst and the colon

Case report 2

59 year-old obese male, alcoholic, with a known history of myocardial infarction in 2007, with aorto-coronary by-pass in the same year and chronic pancreatitis since 2012, with several acute episodes treated conservatively and complicated with type-two diabetes since 2013. In the same year the patient was diagnosed with toxic hepatopathy and F3 fibrosis.

Since 2014 the patient was diagnosed with a 5 by 5 centimeters pancreatic pseudo-cyst of the body and tail that was in close contact with the posterior wall of the stomach. No signs of acute or chronic bleeding were present at that time. Also no signs of communication with the stomach were present. The patient neglected his chronic pathology and continued alcohol consumption.

He was admitted to our internal medicine department for hematochezia.

At admission he was conscious (GCS of 15), hemodinamically stable with a blood pressure of 145 over 70, heart rate of 72 bpm, no fever. Hemoglobin level was 9 g/dl, leucocytes 13810 with thrombocytes of 262.000 per ml. Renal and liver function were normal. Coagulation was between normal values. After twelve hours the patient was admitted to the Intensive Care Unit for severe dyspnea and hypotension. Fresh blood, about 500 cc, was exteriorized through the nazo-gastric tube.

An upper gastrointestinal endoscopy was performed and revealed fresh blood and cloths without a clear source of bleeding.

An emergency CT exam revealed a 7 by 8 centimeters pancreatic body and tail pseudo-cyst with compression on the posterior wall of the stomach. In the center of the pseudo-cyst signs of bleeding were identified. (Figs. 8-10) Blood was present in the small intestine and the colon. A left pneumonic reaction was identified.

After the CT exam the patients' state deteriorated, he became unstable and hemoglobin level dropped to 5 g/dl. He was rushed to the OR for emergency surgery.

After transecting the gastro-colic ligament, the short gastric vessels and the gastro-splenic

Figure 8. Contrast enhanced CT scan - pseudo-aneurysm of the splenic artery and pancreatic body pseudo-cyst that is in close contact with the posterior aspect of the stomach

Figure 9. Contrast enhanced CT scan - pseudo-aneurysm of the splenic artery and pancreatic body pseudo-cyst that has a communication with the stomach

Figure 10. Contrast enhanced CT scan - pancreatic body pseudo-cyst that is in close contact with the posterior aspect of the stomach

ligament, the pancreatic region was visualized. A pancreatic body and tail pseudo-cyst with communication with the posterior wall of the stomach was identified. The communication was near the great curvature of the stomach. (Fig. 11) The operative surgeon decided to perform a medial to lateral spleno-pancreatectomy en bloc with partial gastric resection.

The splenic artery was identified at the superior edge of the pancreas. After ligation and sectioning of the splenic artery a tunnel was made between the posterior aspect of the pancreatic body so that a 60 mm green cartridge Tri-Staple Technology® Endo-GIA Stapler® could be introduced for the sectioning of the pancreas and splenic vein. A linear two-staple GIA Stapler® was used for the sectioning of the stomach at the level of its great curvature, so an en-bloc resection was performed. Medial to lateral dissection was performed with minimal blood-loss. The specimen was extracted and at close examination the source of the bleeding was identified as a pseudo-aneurysm of the splenic artery ruptured in the cavity of the pseudo-cyst. (Fig. 12)

The stapled pancreatic stump was reinforced with separated Prolene 4-0 sutures. The stapled gastric margin was reinforced with a continuous PDS 4-0 suture.

After surgery the patient was admitted in the ICU for recovery. From the surgical point of view the patient presented no pancreatic or gastric leak, liquid food was administered through the naso-enteric feeding tube from the second postop day. A control CT exam was performed on the seventh postoperative day and showed no signs of intra-abdominal complications.

The patients state deteriorated and he developed severe pneumonia with *Acinetobacter* sp. and then the CT scan and lumbar puncture diagnosed him with sub-arachnoid bleeding.

He progressively developed multiple organ failure and died on the 50-th postoperative day in spite of Intensive Care Therapy.

The final pathological exam confirmed the intraoperative findings and concluded that the patient suffered from a ruptured pseudo-aneurysm of the splenic artery into a pseudo-

Figure 11. Intraoperative aspect

Figure 12. Surgical specimen

cyst of the pancreas that had a communication with the posterior aspect of the stomach.

Discussion

Peripancreatic pseudo-aneurysms can be associated with several abdominal pathologies but most frequent they can be seen in conjunction with pancreatitis both chronic and acute and with pancreatic surgery, due to intensive vessel dissection and postoperative complications. (2) They are frequently associated with pseudo-cysts.

The splenic artery is involved in more than 50% of cases.

Looking through multiple papers that cover

this topic we concluded that this pathology is more frequent in males and the mean age is about 56-57 years. (2-4,5-12)

Pseudo-aneurysm can rupture directly into the digestive tract, the peritoneal cavity, the retroperitoneum, the pancreatic duct (haemosuccus) or indirectly via the pseudo-cyst. In literature the risk of pseudo-aneurysmal rupture is as high as 37% with a mortality rate of near 90% for ruptured cases. (13,14) No relation was found between the size of the splenic pseudo-aneurysm and the risk of rupture. (15)

A non-surgical patient often can present only mild abdominal pain but when the pseudo-aneurysm ruptures into the digestive tract or the pancreatic duct he can present hematemesis or haematochesia. This episode can be self-limiting or can lead to a major bleeding. Operated patients often bleed directly into the abdominal cavity and fresh blood is exteriorized through the drainage tubes. (2) In both case scenarios, if the patient presents signs of bleeding his evolution is marked by increased rates of mortality and morbidity. Patients who are discovered accidentally with this pathology can benefit from minimally invasive approach; they are haemodynamically stable and associate lower mortality and morbidity.

If there is time, the patient does not present signs of bleeding or he is stable, he must be evaluated properly. The best way to evaluate such a patient is by contrast enhanced computer tomography that can describe the location of the pseudo-aneurysm, its dimensions, contact or communications with other surrounding organs and can pin point the arterial branch that is involved.

Selective angiography can be used and it is both a diagnosis tool and a treatment option. In literature it is considered to be the gold standard for diagnosis and provide deferential diagnosis from a simple pseudo-cyst or peri-pancreatic fluid collections. (16,17) Control of the involved artery can be obtained by embolization or stenting. This can be performed even in patients with massive bleeding in order to control the bleed and thus hopping to reduce the complication rate. A single procedure of

embolization or stenting has a high rate of failure and often several attempts are done in order to completely manage the aneurysm. (2)

These specialized procedures are highly dependent on the endowment of the medical facility in which the patient is treated. Ideally for the patient is to receive treatment in an angio-surgical-setting under general anesthesia. The radiological team is the first one who tries to solve the problem. If it fails, the surgical team can perform an invasive procedure in order to control and solve the problem.

The risk for recurrent bleeding from an embolized pseudo-aneurysm remains present for days even months after the initial bleeding and is closely linked with exposure to pancreatic juices and local sepsis. (2,9,11,18)

Gambiez et al. (19) presented in his study that the majority of his patients were surgically treated and no signs of recurrent bleeding were present after definitive pancreatic surgery. He also concluded that angiographic methods might associate other severe complications such as necrosis of the duodenum or the development of spleen abscesses.

Management of these patients is complicated. In many cases they are unstable, so treatment must be prioritized. First the bleeding must be controlled, if possible, and then fluid resuscitation must be applied. Third comes definitive treatment of the underlying pathology.

Tony C.Y. Pang et al. (2) classified peri-pancreatic pseudo-aneurysms in accordance with the type of artery involved, communication with gastrointestinal tract and exposure to pancreatic juices, in order to ease up proper management. (2)

If the patient is stable and can be transported, definitive treatment must be applied in high-volume centers for pancreatic surgery because of better resources, higher expertise and better intensive care management. Here the patient can benefit from several types of treatment:

- Trans-catheter embolization with coils, thrombin or gelfoam;
- Trans-catheter stenting of the involved artery, by-passing the pseudo-aneurysm

and proper blood flow is maintained to distal organs;

- Surgical treatment can be performed using both open or minimally invasive techniques (robotic or laparoscopic approach);
- Proximal ligation of the splenic artery and then resection of the pseudo-aneurysm;
- Resection of the pseudo-aneurysm and the corporeo-caudal region of the pancreas with spleen preservation;
- En-bloc resection of the pseudo-aneurysm, of the spleen and of the pancreatic body and tail.

Conclusion

Peripancreatic pseudo-aneurysm of the splenic artery can be deadly. The patient is at risk of bleeding at any given time, even if the initial bleeding episode is managed by endovascular treatment or surpassed on its on.

Management of these patients involves a multidisciplinary team that is usually found in a specialized center for pancreatic surgery. It's usually formed out of a surgeon, interventional radiologist, endoscopist and intensive care doctor.

Surgery remains an important treatment option and is the only feasible procedure in case of an unstable patient or for patients with communication solutions with the digestive tract. Surgery is also indicated for treatment of most postoperative pseudo-aneurysmal bleeding.

Bleeding patients are linked with high rates of mortality and morbidity.

A standardized treatment sequence cannot be found in literature. Treatment must be adapted to each patient and all encountered pseudo-aneurysm must be treated, regardless of his or her size.

Disclosure

None of the authors has a conflict of interest.

References

1. Lupașcu C, Ursulescu C, Andronic D. Emergency pancreatico-

- duodenectomy for bleeding pancreatic pseudoaneurysm in patient with common mesentery and replaced right hepatic artery. *Chirurgia (Bucur)*. 2013;108(6):910-4.
2. Pang TC, Maher R, Gananadha S, Hugh TJ, Samra JS. Peripancreatic pseudoaneurysms: a management-based classification system. *Surg Endosc*. 2014;28(7):2027-38. doi: 10.1007/s00464-014-3434-9. Epub 2014 Feb 12.
3. Kalva SP, Yeddula K, Wicky S, Fernandez del Castillo C, Warshaw AL. Angiographic intervention in patients with a suspected visceral artery pseudoaneurysm complicating pancreatitis and pancreatic surgery. *Arch Surg*. 2011;146(6):647-52. doi: 10.1001/archsurg.2011.11. Epub 2011 Feb 21.
4. Flati G, Andrén-Sandberg A, La Pinta M, Porowska B, Carboni M. Potentially fatal bleeding in acute pancreatitis: pathophysiology, prevention, and treatment. *Pancreas*. 2003;26(1):8-14.
5. Okuno A, Miyazaki M, Ito H, Ambiru S, Yoshidome H, Shimizu H, et al. Nonsurgical management of ruptured pseudoaneurysm in patients with hepatobiliary pancreatic diseases. *Am J Gastroenterol*. 2001;96(4):1067-71.
6. Fujii Y, Shimada H, Endo I, Yoshida K, Matsuo K, Takeda K, et al. Management of massive arterial hemorrhage after pancreatobiliary surgery: does embolotherapy contribute to successful outcome? *J Gastrointest Surg*. 2007;11(4):432-8.
7. Miura F, Asano T, Amano H, Yoshida M, Toyota N, Wada K, et al. Management of postoperative arterial hemorrhage after pancreatobiliary surgery according to the site of bleeding: re-laparotomy or interventional radiology. *J Hepatobiliary Pancreat Surg*. 2009;16(1):56-63. doi: 10.1007/s00534-008-0012-3. Epub 2008 Dec 26.
8. Lee HG, Heo JS, Choi SH, Choi DW. Management of bleeding from pseudoaneurysms following pancreaticoduodenectomy. *World J Gastroenterol*. 2010;16(10):1239-44.
9. Schäfer M, Heinrich S, Pfammatter T, Clavien PA. Management of delayed major visceral arterial bleeding after pancreatic surgery. *HPB (Oxford)*. 2011;13(2):132-8. doi: 10.1111/j.1477-2574.2010.00260.x.
10. de Perrot M, Berney T, Deléaval J, Bühler L, Mentha G, Morel P. Management of true aneurysms of the pancreaticoduodenal arteries. *Ann Surg*. 1999;229(3):416-20.
11. de Perrot M, Berney T, Bühler L, Delgadillo X, Mentha G, Morel P. Management of bleeding pseudoaneurysms in patients with pancreatitis. *Br J Surg*. 1999;86(1):29-32.
12. de Perrot M, Bühler L, Deléaval J, Borisch B, Mentha G, Morel P. Management of true aneurysms of the splenic artery. *Am J Surg*. 1998;175(6):466-8.
13. LiPuma JP, Sachs PB, Sands MJ, Stuhlmiller S, Herbener TE. Angiography/interventional case of the day. Splenic artery pseudoaneurysm associated with pancreatitis. *AJR Am J Roentgenol*. 1997;169(1):259, 262-3.
14. Huang IH, Zuckerman DA, Matthews JB. Occlusion of a giant splenic artery pseudoaneurysm with percutaneous thrombin-collagen injection. *J Vasc Surg*. 2004;40(3):574-7.
15. Tessier DJ, Stone WM, Fowl RJ, Abbas MA, Andrews JC, Bower TC, et al. Clinical features and management of splenic artery pseudoaneurysm: case series and cumulative review of literature. *J Vasc Surg*. 2003;38(5):969-74.
16. Goldberg RF, Maley W, Kennedy EP, Yeo CJ, Lavu H. Giant splenic artery pseudoaneurysm. *J Gastrointest Surg*. 2011;15(6):1063-6. doi: 10.1007/s11605-010-1388-3. Epub 2010 Nov 25.
17. Agrawal GA, Johnson PT, Fishman EK. Splenic artery aneurysms and pseudoaneurysms: clinical distinctions and CT appearances. *AJR Am J Roentgenol*. 2007;188(4):992-9.
18. Hsu JT, Yeh CN, Hung CF, Chen HM, Hwang TL, Jan YY, et al. Management and outcome of bleeding pseudoaneurysm associated with chronic pancreatitis. *BMC Gastroenterol*. 2006;6:3.
19. Gambiez LP, Ernst OJ, Merlier OA, Porte HL, Chambon JP, Quandalle PA. Arterial embolization for bleeding pseudocysts complicating chronic pancreatitis. *Arch Surg*. 1997;132(9):1016-21.