Available online at www.sciencedirect.com

ScienceDirect

journal homepage: http://www.elsevier.com/locate/poamed

Case report

Superior mesenteric and portal vein thrombosis following appendectomy – A case report

Michał Grabala [a], *Paweł Grabala* [b,*], *Piotr Malinowski* [c], *Leszek Adadyński* [a]

[a] *Department of General Surgery and Transplantology, Regional Specialized Hospital in Olsztyn, Poland*
[b] *Department of Orthopaedic Surgery, Regional Specialized Children's Hospital in Olsztyn, Poland*
[c] *Clinical Department of Vascular Surgery, Regional Specialized Hospital in Olsztyn, Poland*

ARTICLE INFO

Article history:
Received 8 March 2016
Received in revised form
12 May 2016
Accepted 27 June 2016
Available online 24 August 2016

Keywords:
Superior mesenteric vein thrombosis
SMVT
Portal vein thrombosis
PVT

ABSTRACT

Introduction: Superior mesenteric vein thrombosis (SMVT) and portal vein thrombosis (PVT) are rare, early complications of surgically treated acute appendicitis. They develop secondary to ongoing inflammation in the peritoneal cavity, in organs that drain blood via the portal vein. Early diagnosis can be difficult due to the lack of specific symptoms.
Aim: Description of a rare complication following surgical treatment of acute appendicitis.
Case study: We present the case of a 26-year-old patient who returned to our hospital 7 days after appendectomy for acute appendicitis, and 5 days after discharge, with severe pain in the epigastrium and vomiting. Ultrasonography and computed tomography (CT) with contrast (angio-CT) of the abdominal cavity were performed, revealing SMVT and PVT.
Results and discussion: We administered unfractionated heparin in the therapeutic range and antibiotics, followed by low-molecular-weight heparin from 2nd day of treatment. The pain completely disappeared with an associated decrease in D-dimer levels. On the 7th day of treatment, a repeat angio-CT scan showed numerous thrombi within the lumen of the portal vein, superior mesenteric vein, and its branches. Inflammatory infiltrations in the adipose tissue surrounding the mesenteric vein had decreased. The patient was discharged home well on 8th day of treatment.
Conclusions: Appendicectomy for appendicitis is one of the most commonly performed surgical procedures, with a low rate of major complications. SMVT and PVT are rare but potentially fatal complications, and this case highlights the importance of early diagnosis and introduction of appropriate treatment.

* *Correspondence to*: Żołnierska 18A, 10-561 Olsztyn, Poland. Tel.: +48 89 675 51 13; fax: +48 775 466 324; mobile: +48 787 111 100.
E-mail address: pgrabala@wp.pl (P. Grabala).

http://dx.doi.org/10.1016/j.poamed.2016.06.008

1. Introduction

Appendicectomy for appendicitis is one of the most commonly performed surgical procedures, with a low rate of major complications. The most common complications are abscesses, perforations, fecal fistulae, peritonitis, mechanical obstruction of the gastrointestinal tract, and postoperative wound infection. Superior mesenteric vein thrombosis (SMVT) and portal vein thrombosis (PVT), early complications of surgically treated acute appendicitis, are rare. They develop due to ongoing inflammation in the peritoneal cavity in organs that drain blood into the portal vein. Initially, there is edema, and hyperemia, and later, intestinal necrosis may occur. The mortality is estimated at 30%,[1,2] and the recurrence rate is approximately 25%.[3] Early diagnosis may be difficult due to the lack of specific symptoms. The best examination that facilitates making a rapid diagnosis is computed tomography (CT) with administration of contrast agent (angio-CT).[4,5] The accuracy of ultrasound with color flow imaging depends on the experience of the sonographer.[6,7]

2. Aim

Description of a rare complication following surgical treatment of acute appendicitis.

3. Case study

Our patient was 26 years-old, 1.72 m tall, weighing 67 kg, and with a BMI of 22.65 kg/m^2 and a history of appendectomy for acute appendicitis 7 days previously. Re-presentation, 5 days after discharge, was due to severe epigastric pain and vomiting. The symptoms started approximately 24 h earlier, with significant intensification in the previous 6–8 h. Physical examination revealed that the abdomen was tight, distended, and tender, especially in the epigastrium and mesogastrium. There were signs of peritonism. Vital signs included a blood pressure of 130/85 mmHg, and a heart rate of 96 beats per minute. Laboratory tests showed an elevated D-dimer of 2.3 μg/mL, white blood count 10 720/μL, C-reactive protein 11.25 mg/dL, fibrinogen 587 mg/dL, and lipase 89 U/L. Radiological examination of the abdomen showed no evidence of obstruction or perforation. Ultrasonography (USG) and angio-CT of the abdominal cavity revealed SMVT and PVT (Fig. 1). A thrombus was observed within the superior mesenteric vein and its branches, with venous dilatation up to 14 mm over a length of approximately 11 cm. Furthermore, a thrombus was seen in the central and distal parts of the portal vein, with dilatation up to 15 mm over a length of approximately 4.5 cm. The branches of the portal vein to the right lobe of the liver were thrombosed, with an approximately 4-cm-long thrombus. It must be emphasized the patient did not receive any type of antithrombotic treatment/prophylaxis before and at the time of the first operation.

Informed consent was obtained from the participant included in the study.

4. Results

We started treatment with an infusion of unfractionated heparin, used low-molecular-weight heparin (*Enoxaparinum natricum*) from 2nd day at a dose of 2× 70 mg, subcutaneously, and antibiotics (piperacillin + tazobactam). There was a significant clinical improvement with resolution of the symptoms of diffuse peritonitis within a few hours of treatment. During the first 48 h of treatment, the patient did

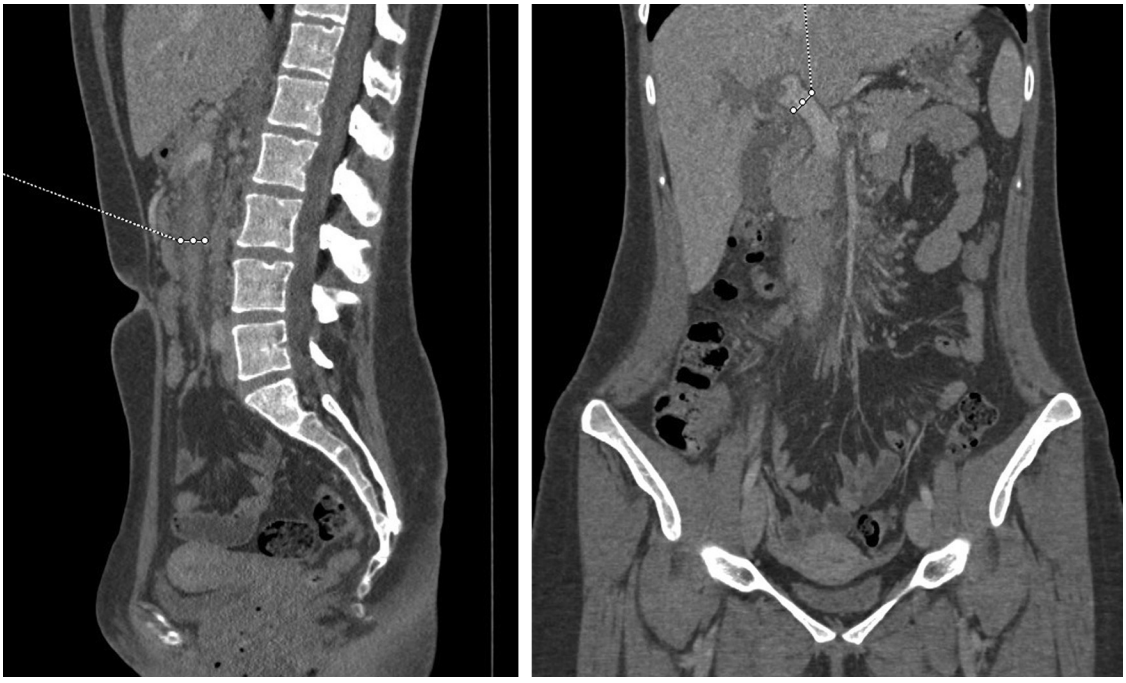

Fig. 1 – Computed tomography with contrast (angio-CT) of the abdominal cavity revealed SMVT and PVT.

not receive solid foods. A hematologist, vascular surgeon, cardiologist, and gynecologist were consulted in order to exclude causes of SMVT and PVT other than the history of appendectomy. There were not found any other causes.

A repeat angio-CT scan on 7th day of treatment showed numerous thrombi within the portal vein and the superior mesenteric vein and its branches, with reduced inflammatory infiltrations in the adipose tissue surrounding the mesenteric vein. The patient was discharged home well on 8th day of treatment. Because the family history included maternal scleroderma, a maternal aunt with systemic lupus erythematous, and paternal pulmonary embolism, our patient was referred for further, in-depth hematologic diagnostic tests, which were negative for other causes of thrombosis. She was discharged home with one enoxaparin per day for 3 months, which was subsequently extended for another 3 months by a hematologist. A further angio-CT scan of the abdominal cavity 6 months later demonstrated normal and unobstructed superior mesenteric and portal veins with no presence of thrombi (Fig. 2).

5. Discussion

PVT is defined as the presence of a thrombus narrowing or occluding the lumen of this vessel. Apart from the portal vein trunk, the intravascular coagulation process may involve intrahepatic branches of this vessel, as well as the superior mesenteric and splenic veins.[8] Thrombosis, according to Virchow's triad, is caused by changes in the vessels, especially damage to the endothelium (e.g. trauma or surgery); changes in blood composition (e.g. thrombophilia); and stasis. Depending on the duration and intensity of the accumulation of symptoms the disease can have an acute, sub-acute, or chronic course.[6,9–11] There are numerous factors that cause thrombosis of the vessels supplying the portal vein. The most common causes are cirrhosis and liver cancer.[10,11] In such cases, thrombosis is caused by stasis of portal venous blood caused by pathological lesions in the liver cytoarchitecture, and the process normally starts in intrahepatic portal vein branches.[10,12] Other factors that may cause portal vein thrombosis include abdominal trauma, intraoperative injuries (e.g. post-splenectomy), ongoing inflammation in the peritoneal cavity within organs from which blood flows into the portal vein, conditions of thrombophilia (e.g. polycythemia, oral contraceptive), cancers, and abdominal tumors (e.g. pancreatic tumors).[10,11,13]

Massive SMVT and PVT, which completely blocks the outflow of blood from the gastrointestinal tract, may occur at any age. The clinical manifestation of PVT depends on the dynamics of the underlying disease process.

Rapidly progressing obstruction of the portal system causes blood stasis in the venous bed, resulting within a few hours in passive intestinal congestion, followed by necrosis. It usually begins suddenly, causing severe abdominal pain, vomiting, bloody diarrhea, and peritonitis due to hemorrhagic intestinal necrosis. The mortality rate is up to 30–50%,[4,14] and depends on the anatomical location and the extent of the thrombotic lesions. Parietal thrombosis, which does not close the entire lumen of a vessel, or involves a single venous trunk, allows for the outflow of blood from small vessels in the bowel mesentery, and has a better prognosis. Evacuating stagnant portal vein blood protects the intestines from necrosis.

In recent years, the incidence of PVT and SMVT has increased likely due to wider access to portal system imaging techniques.[15] Moreover, in the last 15 years, the number of diagnoses in the acute phase of the disease has significantly increased.[16]

SMVT and PVT, early complications of surgically-treated acute appendicitis, are relatively rare. Early diagnosis can be difficult due to the lack of specific symptoms. The acute form of the disease often involves epigastric pain, radiating to the lumbar spine. Pain of this nature may be incorrectly attributed to gastritis or pancreatitis. Passive intestinal congestion can lead to the appearance of a small volume of fluid in the peritoneal cavity. Laboratory tests show leukocytosis, increased level of C-reactive protein (CRP), and D-dimers.

Basic diagnostic tests in portal system thrombosis include USG with blood flow measurements, and CT.[4,17]

USG is the most accessible and harmless examination and allows for multiple repetitions in the same patient. Similar to CT, it allows for the evaluation of the size and structure of the

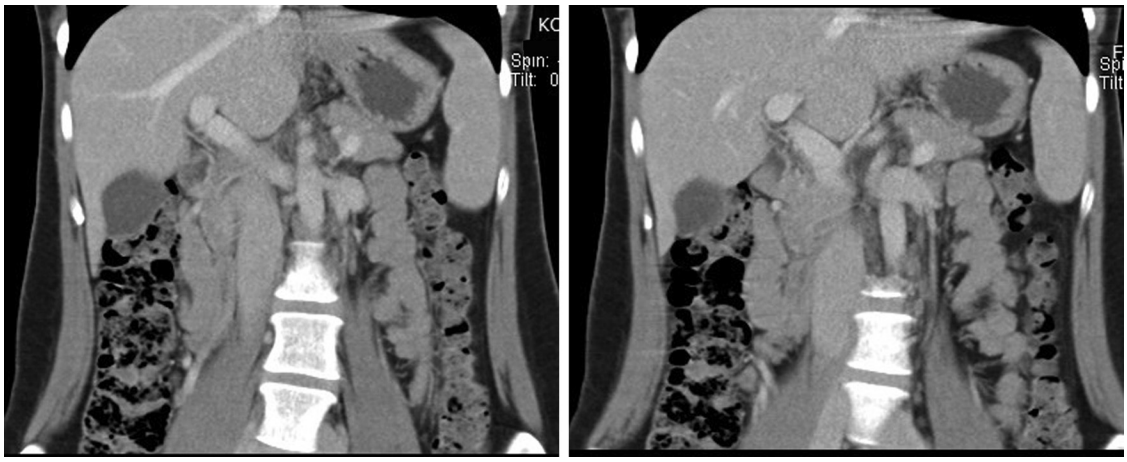

Fig. 2 – Angio-CT scan of the abdominal cavity 6 months later demonstrated normal and unobstructed superior mesenteric and portal veins with no presence of thrombi.

liver and spleen, detection of lesions typical of cirrhosis, as well as detection of cancers and inflammatory tumors in the abdominal organs, which may cause PVT. It enables accurate imaging of the hepatic veins, intrahepatic branches of the portal vein, its trunk, and supplying vessels. It allows for the visualization of portal system thrombosis and can detect collateral circulation.[6,7] Abdominal CT with contrast is the most effective way to diagnose SMVT with a sensitivity of 90–100%.[6,18] Thrombi in the mesenteric vein are imaged directly and the limits of thrombosis can be defined, as well as the exclusion of other diseases that cause gastrointestinal symptoms.[19]

In the literature of the past two decades, there are only a few case reports and a few papers concerning this rare disease and its causes. Plessier et al.[20] presented the results of a multicenter study conducted in 7 European countries over 3 years. In 21% of cases, there was a local risk factor, commonly acute pancreatitis or biliary tract infection, and in 52% of cases, there was a systemic risk factor, usually a myeloproliferative disease (e.g. polycythemia vera, myelofibrosis, chronic myeloid leukemia). No cause was found in 27% of cases. The basis of the therapy was antithrombotic treatment, first with unfractionated, fractionated, or low-molecular-weight heparin, followed by oral medication for a period of 4–12 months. Severe complications were rare: in 2 patients, acute mesenteric ischemia developed on days 6 and 12 after onset, respectively, requiring laparotomy and excision of the necrotic small intestine. There were no deaths during the treatment.

Condat et al.[21,22] presented the results of treatment of 136 patients with PVT, or thrombosis of supplying vessels, conducted over 15 years in a single center. In 67 patients (49%), the primary symptom was abdominal pain; in 24 (18%), there was bleeding from esophageal varices; and in 7 (5%), there was acute mesenteric ischemia requiring laparotomy. The remaining 38 patients had other unusual symptoms. Among the analyzed patients with 'fresh' PVT, there was a local risk factor of an intra-abdominal infection (acute appendicitis, liver abscesses, cholangitis, etc.) in 11, and a systemic risk factor in 13. Five women in this latter group were taking oral contraceptives, a recognized risk factor. Treatment comprised initial administration of low-molecular-weight heparin, followed by warfarin, and the results were evaluated after an average of 5 months. Unblocking of the portal vein was observed in 25 patients (76%) – complete in 10 and partial in 15. No bleeding into the gastrointestinal tract was observed.[22]

Acute SMVT and PVT require symptomatic treatment (rehydration, analgesics) and the use of low-molecular-weight heparin in therapeutic doses. This significantly decreases the risk of thrombosis extension, and consequently, intestinal complications, while providing time for the formation of a collateral circulation. After preliminary treatment with heparin, an oral anticoagulant should be administered for 3–6 months. In the case of thrombophilia, this treatment may be permanent. There is insufficient evidence to assess the clinical usefulness of local or systemic thrombolytic therapy.[23]

Anticoagulant therapy is an essential form of therapy in this disease, giving a chance to unblock the superior mesenteric and portal veins. Success is greater if the treatment starts in the early phase of thrombosis, and when the lesions involve only a part of the portal venous system.[22,24,25] Patients who do not receive anticoagulants are less likely to unblock the portal system, and usually a cavernous transformation of the vein and portal hypertension, and its consequences, eventuate.[21] There are also other methods of treatment including thrombolysis, surgical thrombectomy, and transjugular intrahepatic portosystemic shunt, but they carry risks.[24,25] The prognosis for patients with SMVT and PVT is quite good, provided that the treatment is adequate and the cause is not a degenerative disease. Survival at 5 and 10 years for patients without cirrhosis or cancer is 90% and 80%, respectively.[26]

6. Conclusions

Appendicectomy is one of the most commonly performed surgical procedures, currently with a low rate of major complications. SMVT and PVT are rare, but potentially fatal complications.

The basis of therapy is anticoagulation, initially with unfractionated, then fractionated or low-molecular-weight heparin, and subsequently with oral medication for a period of 4–12 months. Our patient, received enoxaparin once per day for 6 months after discharge, and a repeat abdominal CT scan revealed patent superior mesenteric and portal veins free of thrombi.

This case – because of its rarity and difficulty in diagnosis – demonstrates the importance of early diagnosis and introduction of appropriate treatment. It may be instructive and helpful for surgeons involved in the treatment of acute abdominal diseases.

Conflict of interest

None declared.

REFERENCES

1. Baril N, Wren S, Radin R, Ralls P, Stain S. The role of anticoagulation in pylephlebitis. *Am J Surg*. 1996;172(5):449–452.
2. Plemmons RM, Dooley DP, Longfield RN. Septic thrombophlebitis of the portal vein (pylephlebitis): diagnosis and management in the modern era. *Clin Infect Dis*. 1995;21(5):1114–1120.
3. Divino CM, Park IS, Angel LP, Ellozy S, Spiegel R, Kim U. A retrospective study of diagnosis and management of mesenteric vein thrombosis. *Am J Surg*. 2001;181(1):20–23.
4. Balthazar EJ, Gollapudi P. Septic thrombophlebitis of the mesenteric and portal veins: CT imaging. *J Comput Assist Tomogr*. 2000;24(5):755–760.
5. Nishimori H, Ezoe E, Ura H, et al. Septic thrombophlebitis of the portal and superior mesenteric veins as a complication of appendicitis: report of a case. *Surg Today*. 2004;34(2):173–176.
6. Rhee RY, Gloviczki P, Mendonca CT, et al. Mesenteric venous thrombosis: still a lethal disease in the 1990s. *J Vasc Surg*. 1994;20(5):688–697.
7. Desai SR, Cox MR, Martin CJ. Superior mesenteric vein thrombosis: computed tomography diagnosis. *Aust N Z J Surg*. 1998;68(11):811–812.

8. Bayraktar Y, Harmanci O. Etiology and consequences of thrombosis in abdominal vessels. *World J Gastroenterol*. 2006;12(8):1165–1174.
9. Kumar S, Sarr MG, Kamath PS. Mesenteric venous thrombosis. *N Engl J Med*. 2001;345(23):1683–1688.
10. Muszak J. The impact of selected vasoactive factors on vascular functions. *Pol Ann Med*. 2013;20(2):149–153.
11. Targoński R, Cygański P, Kuciel-Lisiska G, Drozdowski M, Kaleczyc M, Sadowski J. Perioperative disorders of coagulation and fibrinolysis in patients subjected to colorectal cancer resection. *Pol Ann Med*. 2011;18(1):52–65.
12. Aldrete JS, Slaughter RL, Han SY. Portal vein thrombosis resulting in portal hypertension in adults. *Am J Gastroenterol*. 1976;65(3):236–246.
13. Bekkhoucha S, Boulay-Colleta I, Turner L, Berrod JL. Pylephlebitis in the course of diverticulitis. *J Chir (Paris)*. 2008;145(3):284–286 [in French].
14. Dean JW, Trerotola SO, Harris VJ, Snidow JJ, Hawes D. Percutaneous management of suppurative pylephlebitis. *J Vasc Interv Radiol*. 1995;6(4):585–588.
15. Ogren M, Bergqvist D, Björck M, et al. Portal vein thrombosis: prevalence, patient characteristics and lifetime risk: a population study based on 23,796 consecutive autopsies. *World J Gastroenterol*. 2006;12:2115–2119.
16. Rajani R, Björnsson E, Bergquist A, et al. Epidemiology and clinical features of portal vein thrombosis: a multicenter study. *Aliment Pharmacol Ther*. 2010;32(9):1154–1162.
17. Ponziani FR, Zocco MA, Campanale C, et al. Portal vein thrombosis: insight into physiopathology, diagnosis, and treatment. *World J Gastroenterol*. 2010;16:143–155.
18. Rijs J, Depreitere B, Beckers A, Van Lerberghe E, Vollon H, Swinnen F. Mesenteric venous thrombosis. Diagnostic and therapeutic approach. *Acta Chir Belg*. 1997;97(5):247–249.
19. Soulier K, Delèvaux I, André M, et al. Mesenteric venous thrombosis: early diagnosis with abdominal CT scan and screening for coagulopathies. Six observations. *Rev Med Interne*. 2001;22(8):699–705 [in French].
20. Plessier A, Darwish-Murad S, Hernandez-Guerra M, et al. Acute portal vein thrombosis unrelated to cirrhosis: a prospective multicenter follow up study. *Hepatology*. 2010;51 (1):210–218.
21. Condat B, Pessione F, Hillarie S, et al. Current outcome of portal vein thrombosis in adults: risk and benefit of anticoagulant therapy. *Gastroenterology*. 2001;120(2):490–497.
22. Condat B, Pessione F, Helene Denninger M, Hillaire S, Valla D. Recent portal or mesenteric venous thrombosis: increased recognition and frequent recanalization on anticoagulant therapy. *Hepatology*. 2000;32(3):466–470.
23. de Franchis R, Baveno VF. Revising consensus in portal hypertension: report of the Baveno V consensus workshop on methodology of diagnosis and therapy in portal hypertension. *J Hepatol*. 2010;53(4):762–776.
24. Sogaard KK, Astrup LB, Vilstrup H, Gronbaek H. Portal vein thrombosis: risk factors, clinical presentation and treatment. *BMC Gastroenterol*. 2007;7:34.
25. Janssen HL, Wijnhould A, Haagsma EB, et al. Extrahepatic portal vein thrombosis: aetiology and determinants of survival. *Gut*. 2001;49(5):720–724.
26. Primignani M. Portal vein thrombosis, revisited. *Dig Liver Dis*. 2010;42:163–170.