

中文題目：脾動脈假性血管瘤 — 一個急性胰臟炎致命併發症的個案討論

英文題目：Splenic artery pseudoaneurysm as a fatal complication of acute pancreatitis

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**Introduction:** Splenic artery pseudoaneurysm (SAP) is a rare but potentially fatal complication of acute pancreatitis.

**Case Presentation:** We present a 67-year-old female with ruptured SAP as a complication of acute pancreatitis. The patient had mild clinical symptoms on admission, thus was difficult to identify for severe complications. However, she had experienced two episodes of hypovolemic shock on the 10th day after admission. Abdominal computer tomography scan and angiography revealed evidence of splenic artery injury possibly due to acute pancreatitis. The patient underwent transcatheter embolization of splenic artery and total spleen. However, she was still hemodynamically unstable and eventually expired due to severe sepsis. This case raised our clinical awareness of SAP as a rare but life-threatening complication of acute pancreatitis even in patients with short clinical courses and initially mild symptoms.

**Conclusion:** SAP should be kept in mind during image study or in hemodynamically unstable cases of acute pancreatitis.

**Keywords:** Acute pancreatitis, splenic artery aneurysm, splenic artery pseudoaneurysm

## Introduction

Acute pancreatitis is an acute inflammatory process of the pancreas. Most patients present with a mild disease course and can be treated with conservative measures like bowel rest with parenteral nutritional support and adequate fluid resuscitation.<sup>1</sup> By these supportive treatments, the majority of patients recover without complications or organ failure. Splenic artery pseudoaneurysm (SAP) is a rare complication of acute pancreatitis but is life-threatening. The incidence of pseudoaneurysm from pancreatitis is around 1.3% to 10%.<sup>2</sup> Moreover, pseudoaneurysms usually do not occur immediately after an episode of pancreatitis.<sup>2</sup> The time lag was reported to range from 3 weeks to 9 years. This case report described a patient with ruptured SAP as a complication of acute pancreatitis with initially mild symptoms.

## Case Report

A 67-year-old female with ectopic adrenocorticotrophic hormone (ACTH)-secreting tumor and Cushing syndrome went to the emergency department due to decreased appetite, diarrhea and exacerbated general weakness for the past few weeks. Blood tests showed elevated blood glucose (408 mg/dL), amylase (119 U/L) and lipase (483 U/L) levels. Elevated serum liver biochemistry tests (aspartate aminotransferase AST 310 U/L, alanine aminotransferase ALT 588 U/L, alkaline phosphatase Alk-P 395 U/L,  $\gamma$ -Glutamyl transpeptidase rGT 338 U/L, respectively) were also noted. Upper abdominal computed tomography (CT) scan revealed mild swelling appearance of the pancreatic tail with peripancreatic fat stranding and thickening of left anterior renal fascia (**Figure 1**). Results of laboratory tests and image study indicated acute pancreatitis with hyperglycemic hyperosmolar state (HHS). The patient was admitted for further evaluation and management.

Abdominal pain subsided after bowel rest and parenteral nutritional support. We had applied several scoring systems to investigate the clinical severity of this patient, and the results were as following: the Bedside Index of Severity in Acute Pancreatitis (BISAP) score was 2 (low risk with mortality rate < 2%), the initial modified CT severity index was 2 (mild degree with a rate of organ failure around 1%). Serum C-reactive protein (CRP) level was 20 mg/L on admission and decreased to 8.3 mg/L 48 hours later. She started to have a good appetite 2 days after admission. So, the patient started oral feeding.

However, she had decreased level of consciousness on the 10th day after admission, accompanying with hypotension, tachycardia, and desaturation. Hemoglobin level decreased from 9.1 g/dL to 3.6 g/dL, suggesting hypovolemic shock. Resuscitation was done at bedside and catheter embolization was arranged immediately. However, a second shock episode was found right before CTA, so blood transfusion and resuscitation were applied again, and embolization was slightly postponed until her vital signs were stabilized. Ultrasonography at bedside was performed in the morning before consciousness change and after first resuscitation. Only a small amount of ascites was found, and there was no safety window for tapping. Hematoma formation was noted at the adjacent pancreatic tail to peri-splenic region with extravasation on lower abdominal CT scan (**Figure 2**). Abdominal angiography revealed decreased splenic artery size with beaded-appearance and several aneurysms (**Figure 3A**). Acute pancreatitis with splenic artery injury was suspected, thus gelfoam cubes embolization at dorsal pancreatic artery was performed. Nester coils were used to embolize the total spleen and aneurysms the following day (**Figure 3B**). Meanwhile, we prescribed meropenem as empirical antibiotics right after the infection survey at the first shock episode. Blood culture, central line tip culture and blood fungus culture all showed negative results. Sputum and blood sample for viral survey revealed positive cytomegalovirus test by real time polymerase chain reaction (PCR) (>4000 copies/ml), so Ganciclovir was added. Despite resuscitation with antibiotics and antiviral treatment, multiple organ failures with progressing disseminated intravascular coagulation (DIC) appeared. Her family refused major invasive treatment including operation, so we kept conservative treatment. The patient died of multiple organ failures on the 25th day of admission.

## Discussion

Complications of acute pancreatitis can be divided into three groups: local complications, systemic complications, and organ failure. Local complications include acute peripancreatic fluid collection, pancreatic pseudocyst, acute necrotic collection, walled-off necrosis, gastric outlet dysfunction, splenic and portal vein thrombosis, and colonic necrosis. A systemic complication of acute pancreatitis is defined as an exacerbation of an underlying comorbidity by the revised Atlanta classification.<sup>3</sup>

Respiratory, cardiovascular and renal systems are assessed by the modified Marshall scoring system. Any of these three systems with a score 2 or more is defined as organ failure.

We had applied several scoring systems to investigate the clinical severity of this patient in the hope to detect early signs of such severe complication, as mentioned in the case report section. However, evaluated by these scoring systems, we did not expect such rapid deterioration to happen on the patient.

Arterial pseudoaneurysm is a rare yet life-threatening complication of pancreatitis. Visceral aneurysm is most often found at splenic artery (60%), as in our patient.<sup>4</sup> Common etiologies of SAPs include trauma and acute or chronic pancreatitis. Splenic artery pseudoaneurysm is more commonly associated with chronic (46%), rather than acute pancreatitis(6%),<sup>5</sup> and usually do not occur immediately after an episode of pancreatitis. The time lag was reported to range from 3 to 5 weeks by Dorffel et al, 2 months to 8 years (mean, 2.3 years) by Balthazar et al, and 1 to 9 years (mean, 4 years) by Bretagne et al, respectively.<sup>6-8</sup>

Under the setting of pancreatitis, the release of pancreatic enzymes may cause necrotizing arteritis, resulting in vessel wall weakening and rupture. The risk of SAP rupture is around 2% to 10%, which is increased to 76% to 83% in symptomatic patients. If the aneurysm ruptures, the mortality can be as high as 76%.<sup>4,9</sup> The patient had experienced pseudoaneurysm rupture 10 days after admission due to acute pancreatitis, significantly rapid progressing compared with the literature reviewed.

The most common imaging tools to diagnose SAPs include abdominal ultrasound (US), CT scan with intravenous contrast, magnetic resonance imaging, endoscopic US, and contrast angiography (CA). Contrast CT is helpful in diagnosing small SAPs, while the CA is the gold standard for diagnosis and may also be therapeutic. Our patient had initially mild clinical symptoms, and contrast CT scan on admission revealed no evidence of visible SAPs. Splenic artery injury was diagnosed by abdominal CT angiogram after hypovolemic shock due to suspected SAP rupture.

Treatment options for SAP include open abdominal surgery, laparoscopic surgery, endovascular treatment (coil embolization or stent), and medical treatment.<sup>10</sup> Though surgical or endovascular elimination should be performed for aneurysms greater than 2 cm in size, endovascular intervention is a popular option in managing aneurysms.<sup>10</sup> In

this case, the patient was under hemodynamically unstable status, and SAP was not found in the abdominal CT before rupture, which favored smaller size of the lesion. Thus, endovascular approach was selected, and coil embolization of dorsal pancreatic artery and total spleen was performed.

## **Conclusion**

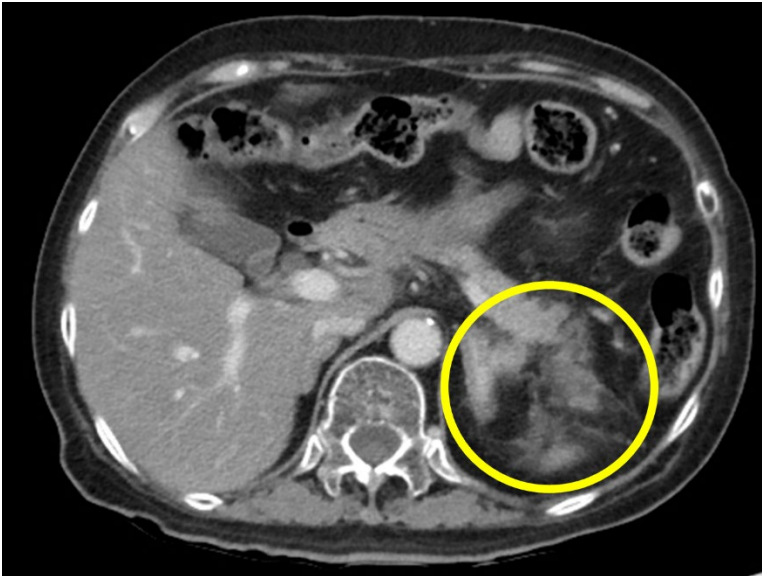
We report a rare complication of acute pancreatitis as ruptured SAP. Despite early transcatheter embolization of splenic artery and total spleen embolization, the patient expired due to severe infection. This case is difficult for early detection of such severe complication due to initially mild clinical symptoms, and her clinical course was significantly rapid progressing compared with the literature reviewed, with the development and rupture of SAP occurring in 10 days after her acute pancreatitis episode. Clinical physicians should keep a high index of suspicion of this complication when treating patients with acute pancreatitis.

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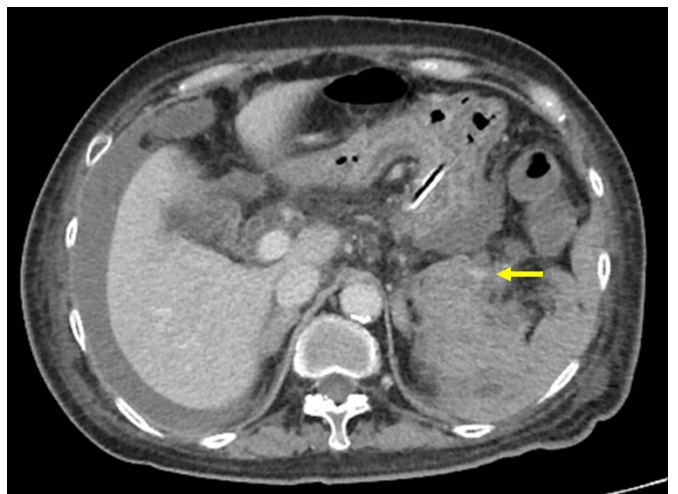
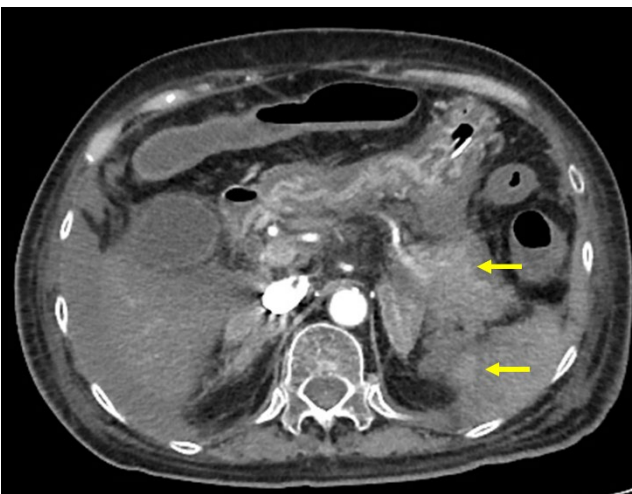
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## Figure Legends

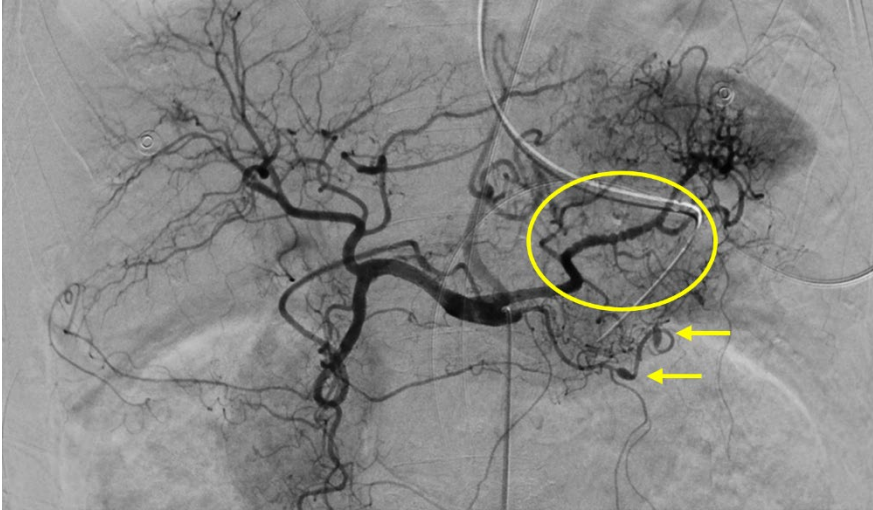
**Figure 1.** CT scan on admission revealed signs of acute pancreatitis.



**Figure 2.** Lower abdominal CT revealed hematoma formation adjacent to the spleen with contrast extravasation (arrows).



**Figure 3.** (a) Abdominal angiography revealed decreased splenic artery size with beaded appearance (circle area) and two aneurysms (arrows).



**Figure 3.** (b) Nester coils (arrows) were used to embolize the total spleen and aneurysms.

