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Intrapancreatic pseudoaneurysm causing massive gastrointestinal hemorrhage and chronic pancreatitis

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ABSTRACT

Pseudoaneurysms of the splenic artery is a well-known complication of pancreatitis. However, to the best of our knowledge, a subcentimetric intrapancreatic pseudoaneurysm (without an associated pseudocyst) causing massive gastrointestinal bleeding and distal main pancreatic duct obstruction is a rare clinical phenomenon. Herein, we present such a unique complication with computed tomography images and subsequent successful endovascular treatment.

Keywords: Intrapancreatic pseudoaneurysm, pancreatitis, gastrointestinal bleeding, splenic artery aneurysm, embolization

Case Report

INTRODUCTION

Pseudoaneurysm of the main peripancreatic arteries is a well-known complications of acute or chronic pancreatitis, with a reported incidence of 10%–20% in acute pancreatitis (1). A hemorrhagic pseudocyst and pseudocyst with a pseudoaneurysm may occur, and intrapancreatic ductal bleeding (hemosuccus pancreaticus) with subsequent gastrointestinal bleeding has also been reported (2). However, formation of an intrapancreatic pseudoaneurysm associated with hemosuccus pancreaticus without an associated pseudocyst is a rare finding (2-4) as a pseudoaneurysm usually occurs by digestion of an artery adjacent to a pseudocyst (4). Woods et al. (4) reviewed the literature regarding gastrointestinal hemorrhage secondary to pancreatitis in 210 patients and revealed that the majority of these patients had pseudocysts associated with pseudoaneurysms.

The natural course of an intrapancreatic pseudoaneurysm is unpredictable and spontaneous rupture may cause mortality with high rates, particularly in morbid post-surgical patients (5). Therefore, treatment of an identified pseudoaneurysm as soon as possible is recommended (5). Endovascular treatment is the first-line option for a pseudoaneurysm, particularly in hemodynamically stable patients, with a reported success rate ranging from 79% to 100% and recurrent bleeding rate ranging from 18% to 37% (6,7). The surgical treatment of a pseudoaneurysm is recommended in unstable patients or those with angiographic failures. However, the deep location of the injured vessel and distortion of the pancreatic anatomy due to the pancreatic inflammation episodes contribute to complexity of the surgical treatment (7). Pancreatic resection is needed in cases with failed proximal ligation of a bleeding vessel, and the mortality rate because of pancreatic resection in a bleeding lesion has been reported to approach 43% (6).

In this article, we present the imaging findings of a rare subcentimetric intrapancreatic pseudoaneurysm (without an associated pseudocyst) and its subsequent endovascular treatment in a patient with acute pancreatitis and profuse lower gastrointestinal tract bleeding, necessitating transfusion.

CASE PRESENTATION

A 62-year-old man presented to the emergency department with severe, sudden-onset abdominal pain-followed by nausea, vomiting, and hematochezia. On

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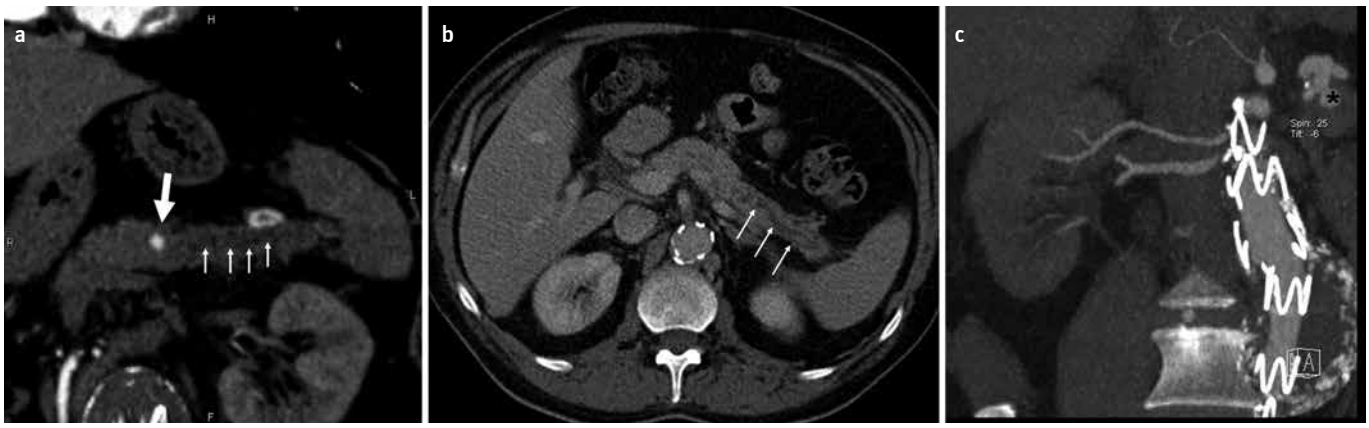

Figure 1. a-c. Arterial **(a)** and venous **(b)** phase images from an abdominal CT. An intrapancreatic splenic artery aneurysm (big arrow) is seen in a close association with the main pancreatic duct. Also note the dilation of the main pancreatic duct and changes suggesting chronic pancreatitis along the tail of the pancreas (small arrows) distal to the pseudoaneurysm. **(c).** MPR image for documenting the patency of the right renal arteries showing the splenic artery aneurysm (*) in this patient who previously underwent EVAR.

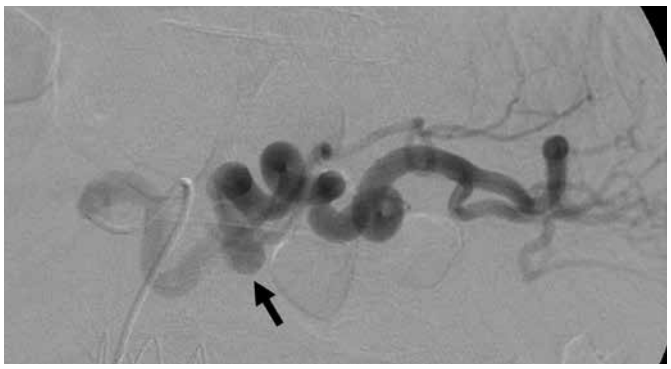

Figure 2. Selective splenic artery injection shows the tortuous splenic artery with a subcentimetric pseudoaneurysm (arrow), which can easily be overlooked considering the tortuous splenic artery anatomy.

initial examination, he was found to be severely pale with prominent orthostatic hypotension. The initial laboratory studies revealed profound anemia with a hemoglobin value of 10 mg/dL. The serum amylase level was 188 U/L (normal range: 28–100 U/L), pancreatic amylase level was 154 U/L (normal range: 17-115 U/L), and aspartate aminotransferase level was 79 U/L (normal <37 U/L). He had no history of chronic liver disease or gastric varices. His had a history of acute pancreatitis 3 years ago, which was treated medically with no discernible etiology. He also had a history of uneventful endovascular abdominal aortic aneurysm repair (EVAR) 9 months ago. He was admitted to the hospital with the same complaints 4 months ago and was treated with red blood cell transfusion (2 units) because upper gastrointestinal system endoscopy and colonoscopy revealed no finding.

The patient was immediately referred to our department with a presumptive diagnosis of acute pancreatitis and gastrointestinal bleeding with possible aorto enteric fistula. The primary sonographic examination was unrevealing because of his extreme discomfort and bowel superimposition. On abdominal computed tomography (CT), the distal pancreatic duct was found to be dilated, and a highly vascular nodular lesion was found at the uppermost part of the dilated duct (Figure 1). In addition, there was inflammatory stranding in the peri-pancreatic fat tissue. The main vascular structures around the pancreas, as well as the other intra-abdominal solid organs, were within normal limits. At first glance, the lesion in the pancreas was presumed to be an occluding neurovascular tumor; however, the density of the lesion was unusually high and it was as bright as the arterial structures in the CT images. On rescanning with CT angiography (CTA) using thin slices, the lesion was found to arise from the splenic artery on multiplanar reformat (MPR) images with a broad neck and dissect the pancreatic tissue to occlude the pancreatic duct. There was no endoleak and no evidence of aorto-enteric fistula on the CT angiography (CTA) images. With a diagnosis of intrapancreatic pseudoaneurysm and secondary hemosuccus pancreaticus, he underwent angiography for further diagnosis and possible endovascular treatment. Informed consent was obtained from the patient.

Under light sedation and local anesthesia, a femoral arterial puncture was made with subsequent abdominal aorta and celiac axis injections to identify the exact location of the splenic artery pseudoaneurysm. As it was known from CTA that the pseudoaneurysm was arising in the caudal direction at the proximal part of the tortuous splenic artery, it was easy to locate it precisely on angiography (Figure 2). A decision was made to embolize the proximal part of the splenic artery distal and proximal to the pseudoaneurysm without embolizing the pseudoaneurysm itself mainly for avoiding further pancreatic duct compression. Using detachable microcoils, the splenic artery was embolized without putting a coil into the aneurysm sac. Control arteriogram after the successful proximal the splenic artery embolization was performed, immediate collateral perfusion of the spleen was confirmed from the other main arterial branches particularly from the gastroduodenal artery (Figure 3). Groin hemostasis was obtained by manual compression, and the patient had no pain related to the solid organ embolization after the procedure, most likely because of the immediate collateralization.

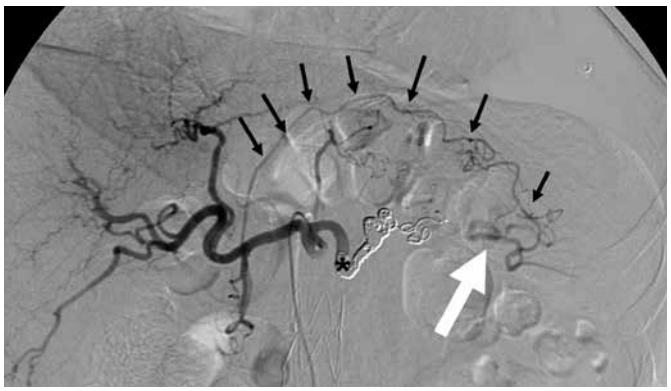

Figure 3. Control celiac artery injection after proximal splenic artery embolization shows the occluded splenic artery (*), with all the coils within the splenic artery itself (no coils in the pseudoaneurysm). Distal reconstruction (white arrow) of the splenic artery immediately developed through the gastroduodenal collaterals (black arrows).

The patientneeded a total of 2 units of red blood cell transfusion. His gastrointestinal bleeding and the drop in the hemoglobin level ceased after the procedure, and he was discharged from the hospital 2 days after presentation with an uneventful course. Three months after the procedure, the patient was symptom free on a normal physical examination. Complete blood count and laboratory tests were normal, with no evidence of splenic and pancreatic abnormalities.

DISCUSSION

The complications of acute pancreatitis are varied and may be of several grades depending on the clinical severity. Pseudocyst formation, sepsis, fatal hemorrhage, and multi-organ failure are among the well-known complications of acute pancreatitis. Pseudoaneurysm formation is a rare occurrence during the course and progression of acute pancreatitis, and the incidence is reported to be between 10% and 20% (1). A pseudoaneurysm is presumed to occur" due to the autodigestion of the wall of the blood vessels secondary to the spillage of pancreatic digestive enzymes. The final point of this process is the formation of a pseudocyst or secondary hemorrhagic pseudocyst. Hemorrhage into the pancreatic duct (hemosuccus pancreaticus) and secondary gastrointestinal hemorrhage may also occur in such patients, as observed in the presented case, to the extent of causing hemodynamic instability (2). In such patients, the pseudocyst must not be percutaneously drained before treatment of the pseudoaneurysm to prevent potentially fatal bleeding. The most commonly involved vessels are the splenic, gastroduodenal, pancreaticoduodenal, left gastric, and common hepatic arteries, in decreasing frequency (3,8-10). In the majority of the presented hemorrhages, pseudoaneurysms are detected with pseudocysts (2,3). Although we believe that the splenic artery pseudoaneurysm was most likely a complication of the existing pancreatitis in the present case, as a vicious cycle, the pseudoaneurysm itself may have subsequently become the reason of chronic pancreatitis by obstructing the main pancreatic duct.

In the present case, ductal dilatation was a worrisome finding for the presence of pancreatic neoplasia. Although pancreatitis is a rare presentation of pancreatic cancer, histological evidence of pancreatitis is more common in resected specimens (11). The pseudoaneurysm was also thought to be an obstructing hypervascular neuroendocrine tumor of the pancreas at first glance. However, contrast enhancement similar to that of the neighboring arterial structures and detection of the relation with the neighboring splenic artery on MPR images was diagnostic. The pseudoaneurysm was thought to be secondary to the previous pancreatitis episode.

Treatment of pseudoaneurysms of the pancreas is a challeng that remains to be solved. Several different suggestions have been reported in the medical literature depending on the different aspects of pseudoaneurysms (12). Surgical treatment, particularly in hemorrhagic pseudocysts, is a well-known and highly successful approach in experienced hands. However, the dense inflammatory adhesions in the surgical bed and high operative mortality rate may sometimes cause concern and hesitation in using this option (12,13). The location of the pseudoaneurysm in the pancreas is also mentioned as a potential indicator of surgery (2). It must also be kept in mind that the operative morbidity and mortality may also increase in hemodynamically unstable patients.

With recent advances in the techniques and technology of endovascular treatment of vascular pseudoaneurysms, this option appears to be the first line of treatment, with high success rates in such patients (2). In proximally located pseudoaneurysms, particularly in highly tortuous splenic arteries, it may be hard to perform selective catheterization, and complete splenic artery embolization may be indicated. In such a clinical scenario, the splenic infarct and abscess formation in the spleen appear to be important, clinical concerns. Although the risk of a splenic infarct is lower in proximal embolization than in splenic artery embolization at the hilum, selective embolization was never considered in the present case to prevent further compression of the main pancreatic duct with a filled aneurysm sac. Having a well-formed collateral vascularization of the splenic parenchyma, proximal splenic arterial embolization appears to be a safe procedure considering that distal pancreatectomy is the surgical alternative. In the present case, the patient's pain and overall clinical condition significantly improved immediately after the procedure, and the blood pressure returned to normal levels. We also think that the decision and plan of treatment of intrapancreatic pseudoaneurysms must not be based on strict criteria; rather, they must be based on the clinical status of the patient. Based on the results of our case, it is obvious that subcentimetric pseudoaneurysms may also be potentially catastrophic and fatal if appropriate measures are not taken promptly and adequately.

Another point we want to emphasize is the use of CTA with MPR images in the diagnosis of subcentimetric pseudoaneu-

rysms of the splenic artery, particularly when they are located intrapancreatically. Apart from excluding the other possible underlying etiologies such as aortoenteric fistulae or pancreatic neoplasms, acquisition of thin slices with multidetector CT definitely improves the diagnostic accuracy of hypervascular lesions of the pancreatic parenchyma and allows discerning neuroendocrine tumors from rare intrapancreatic pseudoaneurysms.

To conclude, endovascular treatment with complete occlusion of the proximal splenic artery may be a useful approach, considering our short follow-up period, in bleeding intrapancreatic pseudoaneurysms, particularly for hemodynamically unstable patients. One also has to keep in mind that pseudoaneurysms may occur without an associated pseudocyst and that reformatted images in different planes must be used to make an accurate diagnosis.

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REFERENCES

1. Storr M, Weigert N, Weiss W, Allescher HD, Berger H. Hemosuccus pancreaticus (bleeding into the pancreatic duct). A rare complication of the acute pancreatitis. Dtsch Med Wochenschr 2001; 126: 108-12. **[CrossRef]**
2. Masatsugu T, Yamaguchi K, Yokohata K, Mizumoto K, Chijiiwa K, Tanaka M. Hemorrhagic pseudocyst and pseudocyst with pseudoaneurysm successfully treated by pancreatectomy: Report of three cases. J Hepatobiliary Pancreat Surg 2000; 7: 432-7. **[CrossRef]**
3. Stabile BE, Wilson SE, Debas HT. Reduced mortality from bleeding pseudocysts and pseudoaneurysms caused by pancreatitis. Arch Surg 1983; 118: 45-51. **[CrossRef]**
4. Woods MS, Traverso LW, Kozarek RA, Brandabur J, Hauptmann E. Successful treatment of bleeding pseudoaneurysms of chronic pancreatitis. Pancreas 1995; 10: 22-30. **[CrossRef]**
5. Saad NE, Saad WE, Davies MG, Waldman DL, Fultz PJ, Rubens DJ. Pseudoaneurysms and the role of minimally invasive techniques in their management. Radiographics 2005; 25 Suppl 1: S173-S189. **[CrossRef]**
6. Hsu JT, Yeh CN, Hung CF, et al. Management and outcome of bleeding pseudoaneurysm associated with chronic pancreatitis. BMC Gastroenterol 2006; 6: 3. **[CrossRef]**
7. Zyromski NJ, Vieira C, Stecker M, et al. Improved outcomes in postoperative and pancreatitis-related visceral pseudoaneurysms. J Gastrointest Surg 2007; 11: 50-5. **[CrossRef]**
8. Golzarian J, Nicaise N, Deviere J, et al. Transcatheter embolization of pseudoaneurysms complicating pancreatitis. Cardiovasc and Intervent Radiol 1997; 20: 435-40. **[CrossRef]**
9. Elton E, Howell DA, Amberson SM, Dykes TA. Combined angiographic and endoscopic management of bleeding pancreatic pseudoaneurysms. Gastrointest Endosc 1997; 46: 544-9. **[CrossRef]**
10. Srivinas M, Kataria R, Bhatnagar V, et al. Intrapancreatic splenic artery pseudoaneurysm: a rare complication of chronic calcific pancreatitis. Pediatr Surg Int 1998; 13: 171-4. **[CrossRef]**
11. Hashimoto M, Watanabe G, Matsuda M, Mori M. Intrapancreatic pseudoanerysm associated with pancreatitis mimicking pancreatic tumor. J Gastroenterol Hepatol 2005; 20: 159-61. **[CrossRef]**
12. Trastek VF, Pairlero PC, Joyce JW, Hollier LH, Bernatz PE. Splenic artery aneurysms. Surgery 1982; 91: 694-9.
13. Boudghene F, L'Hermine C, Bigot JM. Arterial complications of pancreatitis: diagnostic and therapaeutic aspects in 104 cases. J Vasc Interv Radiol 1993; 4: 551-8. **[CrossRef]**