

This article is licensed under the Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC) (<http://www.karger.com/Services/OpenAccessLicense>). Usage and distribution for commercial purposes requires written permission.

Single Case

Spontaneous Resolution of Hemorrhagic Pseudocyst-Associated Pseudoaneurysm in Groove Pancreatitis

Gwang Mo Kim^a Soon Young Ko^{a, b} Joon Ho Wang^{a, b}

^aDepartment of Internal Medicine, Konkuk University Chungju Hospital, Chungju, Republic of Korea; ^bDepartment of Internal Medicine, Konkuk University School of Medicine, Chungju, Republic of Korea

Keywords

Groove pancreatitis · Hemorrhagic pseudocyst · Pseudoaneurysm

Abstract

Hemorrhagic pseudocyst (HP) and pseudocyst-associated pseudoaneurysms (PPs) are complications of pseudocyst. Angiography with embolization has been advocated as the first-line intervention for HP. A 47-year-old man with groove pancreatitis combined with HP near the pancreatic head was treated conservatively. He had relapsed pancreatitis with a newly identified pseudoaneurysm; however, the pseudocyst size was reduced. Although pseudoaneurysm was identified, angiography was not performed because there was no evidence of ongoing bleeding, and he was in a stable condition. Sphincterotomy and stent insertion in the pancreatic duct was applied to prevent relapsed pancreatitis with facilitation of the flow of pancreatic juice. He has done well during the 10-month follow-up, without recurrent pancreatitis. Angiography as an initial approach in HP and PPs may need to be more selective depending on the clinical presentation of the patient. A lysed clot within the strictured pancreatic duct during the healing process has been thought to be the cause of relapsed pancreatitis, and pancreatic sphincterotomy and stent insertion should be the preferred treatment methods.

© 2020 The Author(s)
Published by S. Karger AG, Basel

Introduction

Groove pancreatitis, which is commonly associated with alcohol abuse, is chronic and affects the groove area [1]. Pseudocysts, which are frequently found in the head of the pancreas, are complicated by intracystic hemorrhage and pseudoaneurysm [1, 2]. Angiography with embolization has been advocated as the first-line intervention for hemorrhagic pseudocyst (HP) and pseudoaneurysm in patients with a stable condition and contributes to lower mortality [3, 4]. Prompt diagnostic methods include abdominal computed tomography (CT) and ultrasonography (US) with Doppler imaging, which could detect an ongoing HP with high sensitivity [3]. Endoscopy is useful to preclude and find sources of hemorrhage, in addition to making an approximate assessment of the amount of blood with visualization of bleeding from the papilla. These findings emphasize the role of esophagogastroduodenoscopy (EGD) before angiography. Angiography as an initial approach in HP may need to be more selective depending on the presentation of the patient in light of previous findings [5–7].

Recurrent pancreatitis is related with pseudocyst, chronic pancreatitis, and stricture of the pancreatic duct. Recurrent pancreatitis is defined as a separate attack 3 months after acute pancreatitis with complete resolution [7]. Relapsed pancreatitis is said to be a re-attack within 3 months, which has been suggested to be associated with complications of first attack before complete healing [7]. Herein, we report the case of a 47-year-old man who presented with HP and pseudocyst-associated pseudoaneurysms in groove pancreatitis and was treated conservatively without angiography with embolization. In this case, relapsed pancreatitis could be considered to have occurred during the healing process of HP, which resolved after pancreatic duct stent implantation.

Case Report/Case Presentation

A 47-year-old man presented with acute epigastric pain, nausea, and vomiting for 1 day. He had undergone right nephrectomy due to renal cell cancer 18 years ago. He is a chronic alcohol drinker, that is, drinking four times per week. He was diagnosed with alcoholic chronic pancreatitis and pseudocyst and followed for several months. He visited a hospital because of abdominal discomfort, where he underwent EGD, colonofiberscopy, and abdominal CT. He was told that he had a pseudocyst (3.2 cm) near the pancreatic head and chronic pancreatitis, had been placed under observation, and then discharged. The above symptoms developed 1 day after discharge, and he was admitted through our emergency department. His blood pressure was 140/80 mm Hg, body temperature was 36.8°C, pulse rate was 78 beats per minute, and respiration was 20 breaths per minute. Mild abdominal distension was found with direct tenderness in the epigastric and mid-abdomen. Laboratory findings were as follows: white blood cells (WBCs), 14,300/mm³; hemoglobin (Hb), 12.1 g/dL; hematocrit (Hct), 36.1 %; total protein, 7.3 g/dL; albumin, 3.1 g/dL; platelets (Plt), 212,000/mm³; total bilirubin (TB), 0.5 mg/dL; aspartate aminotransferase (AST), 23 IU/L; alanine aminotransferase (ALT), 12 IU/L; blood urea nitrogen (BUN), 9.8 mg/dL; creatinine (Cr), 1.4 mg/dL; C-reactive protein (CRP), 3.31 g/dL; amylase, 92 (range 37–220) U/L; and alkaline phosphatase, 165 (range 103–335) IU/L. CT revealed a pseudocyst near the pancreatic head. The pseudocyst was a 4.1-cm ovoid lesion with a thick wall and cystic mass not enhanced by contrast near the pancreatic head (Fig. 1). Doppler US revealed a cystic mass and a thick cystic wall without blood flow, which suggested hematoma. EGD showed extrinsic compression in the second duodenum; however, the scope could pass, and there was no evidence of hemorrhage on the duodenal mucosa and

papilla. He was diagnosed with chronic pancreatitis with HP in the groove. His abdominal pain subsided. He refused transfer to other hospital for angiography and was discharged. Twenty days later, he experienced epigastric pain in the mid-abdominal area (VAS score was 7) and was readmitted through the emergency department. He denied drinking and the pain occurred the previous day after dinner. He had a pulse rate of 80 beats per minute, and his blood pressure was 130/80 mm Hg. He did not have fever. His physical examination revealed direct tenderness in the epigastric area. The amylase level was 2,561 U/L and lipase (13–60) was 4,610 U/L. Other laboratory findings including WBC, Hb, Hct, Plt, AST, ALT, BUN, Cr, and CRP were unremarkable. CT showed reduced size of the pancreatic HP with an enhanced vessel within the pseudocyst (Fig. 2). The symptoms and amylase level caused by pancreatitis were improved with conservative management. Seven days later, pancreatitis recurred with epigastric pain during admission. The amylase level was 1,575 U/L, and WBC, Hb, Hct, AST, ALT, TB, BUN, and Cr levels were unremarkable. CT showed reduced pseudocyst size at 2.2 cm; however, there was central contrast collection within the pseudocyst near the pancreatic head (Fig. 3a), suggesting pseudoaneurysm arising from a branch in the gastroduodenal artery (Fig. 3b). The pancreatic duct had mild dilatation with proximal stricture. Endoscopic retrograde pancreatography was performed. Pancreatography did not show communication between the pancreatic duct and pseudocyst, but it showed a small filling defect which was thought to be associated with the lysed material from the HP during pancreatography (Fig. 3b). However, there was no particular finding in the pancreatic juice. He was diagnosed with relapsed pancreatitis with a small pseudoaneurysm within the reduced pseudocyst. To prevent pancreatitis and facilitate the flow of pancreatic juice from the strictured duct, a pancreatic stent was inserted (7 Fr, 7 cm) and was changed to a pancreatic plastic stent to prevent obstruction by 3 months. He had not experienced recurrent pancreatitis since the pancreatic stent was inserted. Follow-up CT showed that the size of the pseudocyst was reduced to 2 cm, and there was no evidence of bleeding after 3 months (Fig. 3c) and with almost complete resolution after 6 months (Fig. 3d). He has done well during the 10-month follow-up, without recurrent pancreatitis.

Discussion/Conclusion

Pseudocyst associated with chronic pancreatitis has been reported to occur mostly in acute recurrence on chronic pancreatitis, which may lead to pseudoaneurysm [8]. In this report, the patient frequently consumed alcohol and had chronic pancreatitis and pseudocyst with a thick wall in the groove, which was found several months ago, suggesting difficult spontaneous resolution. These findings combined with the thick wall of the second duodenum and dilatation of the pancreatic duct are consistent with groove pancreatitis according to imaging findings. Extrinsic compression by the pseudocyst in the groove caused narrowing of the duodenum, and the patient presented with nausea, vomiting, and duodenal outlet obstruction.

Hemorrhage of the pancreatic pseudocyst shows various clinical presentations, ranging from asymptomatic to melena, gastrointestinal bleeding, hemobilia, and hemosuccus pancreaticus, as well as abdominal pain [3]. Generally, a low volume of hemorrhage may be associated with bleeding in a small branch of the artery as in this case, which can lead to a small expansion of the pseudocyst, manifested by hematoma formation within the pseudocyst and the patient may remain in a stable condition [5]. Angiography may not always be mandatory in patients who have no clinical signs of ongoing bleeding when HP is detected by US and CT [9]. This suggests that conservative treatment can be considered if bleeding is not ongoing and

originated from a small artery branch in a patient with a stable condition. In this case, the initial CT showed a cystic mass not enhanced by contrast media within the pseudocyst near the pancreatic head (Fig. 3a). Doppler US did not show arterial flow within the pseudocyst. In addition, the EGD finding and the stable patient condition suggested the absence of ongoing bleeding.

In this case, chronic pancreatitis with dilatation of the pancreatic duct and HP could be the causes of relapsed pancreatitis, and the lysed clot within the pseudocyst combined with the strictured duct has been strongly thought to lead to relapsed pancreatitis. In this case, abdominal CT showed the decreased size of the pseudocyst near the pancreatic head, indicating resolution of the hematoma (Fig. 2). The lysed clot could be drained from the pseudocyst into the pancreatic duct, which in turn increased the pressure in the duct, and could have led to relapsed pancreatitis, although a small newly identified pseudoaneurysm was found. However, it was uncertain whether the pseudoaneurysm existed from the beginning. The pseudoaneurysm was in a small branch arising from the gastroduodenal artery (Fig. 3a). In this case, although a connection between the main pancreatic duct and pseudocyst was not found during pancreatography, the finding that the size of the pseudocyst was decreased supported that the pseudocyst was connected with the pancreatic duct, and the dissolved hematoma drained into the pancreatic duct. Thus, it was expected that stent insertion in the pancreatic duct through the papilla could facilitate the flow of pancreatic juice to drain from the duct. Endoscopic treatment including sphincterotomy with a plastic stent of 5–7 Fr in diameter in the pancreatic duct can be considered a therapy of choice [4, 7]. The pancreatic stent was inserted (7 Fr, 7 cm) and changed to prevent obstruction of the duct by 3 months. He had no recurrent pancreatitis. CT showed that the size was reduced to 2 cm after 3 months, pseudoaneurysm was not observed (Fig. 3c), and there was almost complete resolution after 6 months, but the thickening of the duodenum in the groove was retained, which needs close observation for rebleeding and malignancy (Fig. 3d).

In conclusion, angiography as an initial approach to treat HP may need to be more selective depending on the clinical presentation of the patient. Relapsed pancreatitis in this patient with HP could have occurred during the healing process of HP. The lysed clot and strictured pancreatic duct have been thought as the cause of relapsed pancreatitis, and pancreatic sphincterotomy and stent insertion should be considered the preferred treatment methods.

Statement of Ethics

Since this is simple retrospective case report, only written informed consent was obtained from the patient for the publication of this report.

Disclosure Statement

The authors have no conflicts of interest to declare.

Funding Sources

The authors declare that no funding was received for the preparation of the manuscript.

Author Contributions

All authors contributed to the preparation, review, and editing of the manuscript.

References

- 1 Tezuka K, Makino T, Hirai I, Kimura W. Groove pancreatitis. *Dig Surg*. 2010;27(2):149–52.
- 2 Chantarojanasiri T, Isayama H, Nakai Y, Matsubara S, Yamamoto N, Takahara N, et al. Groove pancreatitis: endoscopic treatment via the minor papilla and duct of Santorini morphology. *Gut Liver*. 2018 Mar;12(2):208–13.
- 3 Zyromski NJ, Vieira C, Stecker M, Nakeeb A, Pitt HA, Lillemoe KD, et al. Improved outcomes in postoperative and pancreatitis-related visceral pseudoaneurysms. *J Gastrointest Surg*. 2007 Jan;11(1):50–5.
- 4 Carr JA, Cho JS, Shepard AD, Nypaver TJ, Reddy DJ. Visceral pseudoaneurysms due to pancreatic pseudocysts: rare but lethal complications of pancreatitis. *J Vasc Surg*. 2000 Oct;32(4):722–30.
- 5 Sand JA, Seppänen SK, Nordback IH. Intracystic hemorrhage in pancreatic pseudocysts: initial experiences of a treatment protocol. *Pancreas*. 1997 Mar;14(2):187–91.
- 6 Castillo-Tandazo W, Ortega J, Mariscal C. Spontaneous regression of splenic artery pseudoaneurysm: a rare complication of acute pancreatitis. *Int Med Case Rep J*. 2013 Apr;6:17–20.
- 7 Khurana V, Ganguly I. Recurrent acute pancreatitis. *JOP*. 2014 Sep;15(5):413–26.
- 8 Pang TC, Maher R, Gananadha S, Hugh TJ, Samra JS. Peripancreatic pseudoaneurysms: a management-based classification system. *Surg Endosc*. 2014 Jul;28(7):2027–38.
- 9 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 Jan;86(1):29–32.

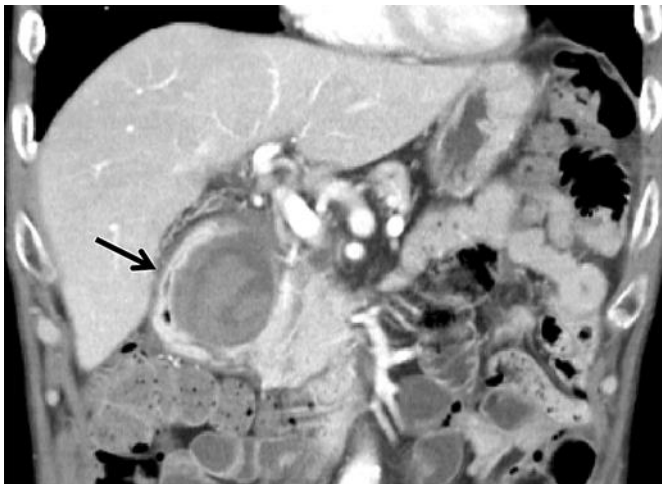


Fig. 1. Contrast-enhanced computed tomography image showing a 4.1-cm ovoid lesion without contrast enhancement near the pancreatic head with mild dilatation of the pancreatic duct.

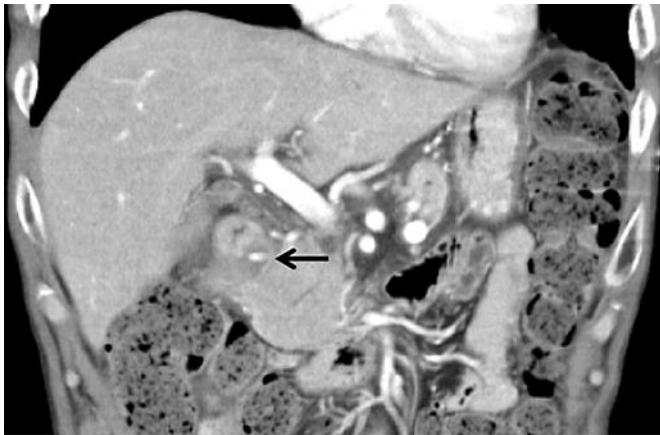


Fig. 2. Contrast-enhanced computed tomography image showing a 2.2-cm ovoid low-density lesion with an enhanced vessel near the pancreatic head, suggesting pseudoaneurysm.



Fig. 3. **a** Contrast-enhanced computed tomography image showing a 2-cm ovoid low-density lesion with central collection near the pancreatic head in the arterial phase, suggesting pseudoaneurysm arising from a branch of the gastroduodenal artery. **b** Pancreatogram showing that the proximal portion of the major pancreatic duct was narrowed with slight dilation at the mid- and distal portions. **c** F/U contrast-enhanced computed tomography image showing no evidence of bleeding. **d** Contrast-enhanced computed tomography image showing an almost reduced cystic lesion near the pancreatic duct. The stent was noted in the main pancreatic duct.