## **CASE REPORT**

## A rare and life-threatening bleeding into a pancreatic pseudocyst

Una hemorragia rara y potencialmente mortal en un pseudoquiste pancreático

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## Abstract

Acute and chronic pancreatitis are among the most common diseases of the gastrointestinal tract. Most cases of acute pancreatitis are mild to moderate, but 15-20% of patients develop severe forms of pancreatitis that require surgery in 10.5% of cases. One of the complications of acute pancreatitis is the formation of pseudocysts. In rare cases, pseudocysts are associated with the development of pseudoaneurysms of the splenic, gastroduodenal, and pancreaticoduodenal arteries. Rupture of a pseudoaneurysm can lead to bleeding into various parts of the gastrointestinal tract, including the stomach, duodenum, pancreatic ducts, and biliary tree. The present case demonstrates bleeding from a pseudoaneurysm of the splenic artery into the gastrointestinal tract and then into the abdominal cavity. Bleeding into the ductal system of the pancreas can mimic upper gastrointestinal bleeding leading to misdiagnosis, inadequate treatment and even death.

Key words: Pancreatic pseudocyst, splenic artery pseudoaneurysm, bleeding, acute pancreatitis.

## Resumen

La pancreatitis aguda y crónica es una de las enfermedades más comunes del tracto gastrointestinal. La mayoría de los casos de pancreatitis aguda son de leves a moderados, pero entre el 15 y el 20% de los pacientes desarrollan formas graves de pancreatitis que requieren cirugía en el 10,5% de los casos. Una de las complicaciones de la pancreatitis aguda es la formación de pseudoquistes. En raras ocasiones, los pseudoquistes se asocian al desarrollo de pseudoaneurismas de las arterias esplénica, gastroduodenal y pancreaticoduodenal. La rotura de un pseudoaneurisma puede provocar hemorragias en diversas partes del tracto gastrointestinal, como el estómago, el duodeno, los conductos pancreáticos y el árbol biliar. El presente caso demuestra la hemorragia de un pseudoaneurisma de la arteria esplénica en el tracto gastrointestinal y luego en la cavidad abdominal. La hemorragia en el sistema ductal del páncreas puede simular una hemorragia digestiva alta, lo que puede conducir a un diagnóstico erróneo, un tratamiento inadecuado e incluso la muerte.

Palabras clave: Pseudoquiste pancreático, pseudoaneurisma de la arteria esplénica, hemorragia, pancreatitis aguda.

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## Introduction

The incidence of acute pancreatitis is 5-80 cases per 100,000 people, while chronic pancreatitis is encountered 5-12 cases per 100,000<sup>1,2</sup>. Most cases of acute pancreatitis are mild to moderate, but 15-20% of patients develop severe forms of pancreatitis that require surgery in 10.5% of cases<sup>3</sup>.

One of the complications of acute pancreatitis is the formation of pseudocysts – cavities containing fluid and surrounded by an inflammatory infiltrate. Pseudocysts form in 6-18.5% of cases in acute pancreatitis and in 20-40% of cases in chronic pancreatitis. Despite rare complications, pseudocysts are associated with the development of pseudoaneurysms of the splenic, gastroduodenal, and pancreaticoduodenal arteries<sup>4,5</sup>. Rupture of a pseudoaneurysm can lead to bleeding into various parts of the gastrointestinal tract, including the stomach, duodenum, pancreatic ducts, and biliary tree<sup>6</sup>.

The presented clinical case describes bleeding from a pseudoaneurysm of the splenic artery into the gastrointestinal tract, and then into the abdominal cavity mimicking upper gastrointestinal bleeding. Bleeding into the ductal system of the pancreas is rare and can mimic upper gastrointestinal bleeding. In this settings the patient will undergo endoscopy to find the source of bleeding, while splenic artery embolization or ligation are the optimal treatment strategies.

## **Case report**

A 38-year-old male, was urgently admitted to the hospital on November 20, 2019 with complaints of traces of blood in the stool. The patient considered himself ill since the summer of 2019, when for the first time after drinking alcohol he noted the appearance of girdle pain in the upper abdomen. In August he vomited blood and had black stool mixed with blood for two days. The patient was examined at the place of residence but upper gastrointestinal endoscopy and fibrocolonoscopy (FCS) did not reveal any source of bleeding. Upon admission his hemoglobin level was 79 g/l (normal reference 138-172 g/l), erythrocytes 3.3\*1012 (normal reference 4.3-5.9\*1012), hematocrit 25% (normal reference 41-50%). There were no other changes in laboratory tests.

Due to the clinical picture of gastrointestinal bleeding, he undergone esophagogastroduodenoscopy and FCS. During FCS, blood flow from the small intestine was noted but the source of bleeding was not visualized. The patient underwent computed tomography of the abdominal organs with intravenous contrast, which revealed a postnecrotic cyst of the pancreatic tail (**Figure 1**). At the time of the study, extravasation of the contrast agent was not detected (Figures 1 & 2). The patient underwent complex conservative therapy with a positive effect: the pain syndrome was relieved, laboratory parameters tended to normalize (increase in Hb level to 107 g/l). In order to search for a possible source of bleeding, the patient underwent enteroscopy, during which no data for an organic pathology of the small intestine were revealed. However, on December 8, 2019, the patient had clinical manifestations of hemorrhagic shock, clinical death and required resuscitation. An emergency ultrasound was performed and reveled free fluid in the abdominal cavity in the form of wide borders: subhepatic, parasplenic and in the pelvis (Figure 3). The was also a large mass with liquid (pseudocyst) that had a heterogeneous content (Figure 4).

Figure 1: CT scan of the abdomen. Splenic artery with an aneurysm (arrow).



Figure 2: CT scan of the abdomen reconstruction. Splenic artery with an aneurysm (arrow).



The patient was resuscitated and rushed to the operating room. A median laparotomy was performed under combined endotracheal anesthesia and in aseptic conditions. During revision there was fresh blood with clots in all parts of the abdominal cavity. Blood was reinfused using Cell Saver apparatus, with intraoperative autotransfusion of 3 liters of blood. During examination there was an active flow of arterial blood from the foramen of Winslow. The omental sac was opened through the gastrocolic ligament. Through the lesser omentum a hematoma was visualized and palpated in the region of the body-tail of the pancreatic cyst. The cyst was opened and in one of the walls there was an erosive area of the splenic artery, up to 1 cm, with active bleeding. The bleeding was stopped by stitching the artery with Prolen 4.0. Taking into account the severity of the patient's condition, previous resuscitation measures and the episode of clinical death, it was decided to refrain from expanding the surgical intervention. A drain was installed on the right through the foramen of Winslow into the omental bag, brought out through the counter-opening in the right mesogastrium. One drain was installed through the gastrocolic ligament, brought out through the counteropening in the right mesogastrium. Drainage into the pelvis was brought out through the counter-opening in the right iliac region. The wound was sutured layerby-layer. The patient was then transferred to intensive care unit. Given the high risk of recurrent bleeding from the eroded area of the splenic artery the patient undergone upper mesentericography, celiacography, angiography, and mechanical embolization of the splenic artery on December 11, 2019 (Figure 5). Upon stabilization of the condition, on December 16, 2019, the patient was transferred to the surgical department, where complex conservative therapy was continued: antiulcer, antibacterial, anti-inflammatory, infusions and pain management. The drains were removed upon stabilization and the patient was discharged for supervision at the place of residence.

Figure 3: Free fluid in the abdominal cavity (A - subhepatic, a pseudocyst area is also observed in the scanning area; B - in the pelvis).



Figure 4: Pseudocyst of the pancreas with a heterogeneous component (A - transverse section in the epigastric region; B - sagittal section, the left lobe of the liver is also observed in the scanning area).



Figure 5: Catheterization of the splenic artery and embolization of the aneurysm.



## **Discussion**

Pseudocysts form as a result of enzyme buildup and self-digestion of the pancreas in patients with acute pancreatitis. In chronic pancreatitis, pseudocysts form as a consequence of high intraductal pressure and destruction of blocked ducts<sup>7</sup>.

Although pseudocyst rupture is rare, it can be dangerous. Rupture usually occurs as a result of trauma, infection, or pancreatitis. The most dangerous complication of pseudocyst is bleeding, which occurs in 6-8% of cases and can be fatal<sup>8</sup>.

Bleeding can occur immediately after an episode of pancreatitis or after some time. Initially, inflammation and lysis of the elastic component of the vessel can occur, which leads to erosion and disruption of the wall structure. Then, the pseudocyst can cause vessel damage by compression, which leads to ischemia and further injury to the vessel wall. Finally, pseudocysts can lead to compression of the surrounding vessels, thrombosis and increased intravascular pressure, which also contributes to the violation of the integrity of the vascular wall. It is important to consider that the arteries supplying the pancreas vary as well as their diameter. Therefore, damage to large branches of the splenic artery can lead to rapid and profuse bleeding<sup>9,10</sup>. As a result, one of the possible complications may develop: bleeding into the cyst, bleeding into the gastrointestinal tract, and bleeding into the abdominal cavity. Various locations of bleeding are shown in Figure 6.

Figure 6: Localization of bleeding depending on the area where the aneurysm is emptied.



Intra-abdominal bleeding from pseudocysts is associated with high mortality (35.3-40% of cases)<sup>11,12</sup>. Pseudocysts are especially dangerous if they are localized near large vessels of the spleen or pancreas<sup>12,13</sup>. Diagnosis may require intravenous contrast-enhanced CT, ultrasound with Doppler imaging, and angiography. The optimal method of treatment is endovascular intervention with aneurysm embolization<sup>12</sup>. However, in hemodynamically unstable patients, the most rational method of treatment is midline laparotomy with ligation of the bleeding vessel<sup>14</sup>. Depending on the location of the aneurysm and the individual anatomy of the patient, splenectomy, partial resection of the cyst, ligation of the splenic artery or its branches may be required<sup>15</sup>.

The current case demonstrates that in rare cases splenic artery can bleed into the pseudocyst and the into the ductal system of the pancreas. CT with contrast enhancement does not always allow to diagnose extravasation even in large diameter vessels when they are compressed. The presence of an aneurism in patients with acute pancreatitis and symptoms of bleeding requires reevaluation of the main source of hemorrhage with subsequent embolization or ligation of the artery.

## Conclusions

Bleeding from an aneurysm of the splenic artery in the setting of acute pancreatitis is a rare but potentially life-threatening condition. Rarely, an aneurysm may empty into a pancreatic cyst and eventually into the gastrointestinal tract, giving the illusion of gastrointestinal bleeding. Depending on the hemodynamic parameters, the patient can undergo aneurysm embolization or emergency laparotomy and vessel ligation.

## **Author Contributions**

Conceptualization, AS and SC; formal analysis AS, VB, DD, AE, AK, NP, SC; investigation AS, VB, AE, AK, NP, SC; resources AS, VB, DD, AE, AK; data curation AS, VB, DD, AE, AK; writing—AE, AK, NP, SC; writing—review and editing AS, VB, DD, AE; visualization AK, NP, SC; supervision AS, VB, DD. All authors have read and agreed to the published version of the manuscript.

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#### **Informed Consent Statement**

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#### **Conflicts of Interest**

The authors declare no conflict of interest.

## **References**

1. Yadav D, Lowenfels AB. The epidemiology of pancreatitis and pancreatic cancer. Gastroenterology. 2013;144(6):1252-61.

2. Banks PA. Epidemiology, natural history, and predictors of disease outcome in acute and chronic pancreatitis. Gastrointestinal endoscopy. 2002;56(6 Suppl):S226-30.

3. Sarri G, Guo Y, Iheanacho I, Puelles J. Moderately severe and severe acute pancreatitis : a systematic review of the outcomes in the USA and European Union-5. BMJ Open Gastroenterology. 2019;6(1):e000248.

4. Carr JA, Cho JS, Shepard AD, Nypaver TJ, Reddy DJ. Visceral pseudoaneurysms due to pancreatic pseudocysts: rare but lethal complications of pancreatitis. Journal of vascular surgery. 2000;32(4):722-30.

5. Lerch MM, Stier A, Wahnschaffe U, Mayerle J. Pancreatic pseudocysts: observation, endoscopic drainage, or resection? Dtsch Arztebl Int. 2009;106(38):614-21.

6. Jorge Cerrudo J, Mansilla Roselló A, Paz Yañez A, Segura Jiménez I, Ferrón Orihuela JA. [Urgent vascular complications in acute pancreatitis]. Cirugia espanola. 2012;90(2):134-6. PubMed PMID: 21414617.

7. Kim HC, Yang DM, Kim HJ, Lee DH, Ko YT, Lim JW. Computed tomography appearances of various complications associated with pancreatic pseudocysts. Acta radiologica (Stockholm, Sweden : 1987). 2008;49(7):727-34.

8. Urakami A, Tsunoda T, Kubozoe T, Takeo T, Yamashita K, Imai H. Rupture of a bleeding pancreatic pseudocyst into the stomach. Journal of hepato-biliary-pancreatic surgery. 2002;9(3):383-5.

9. Covantev S, Mazuruc N, Belic O. A Rare Case of Vascularization of the Body and Tail of the Pancreas. Online J Health Allied Scs. 2017;16(3):10.

10. Covantev S, Mazuruc N, Belic O. The Arterial Supply of the Distal Part of the Pancreas. Surgery Research and Practice. 2019;2019:5804047.

11. Okamura K, Ohara M, Kaneko T, Shirosaki T, Fujiwara A, Yamabuki T, et al. Pancreatic Pseudocyst Ruptured due to Acute Intracystic Hemorrhage. Case Reports in Gastroenterology. 2017;11(3):755-62.

12. Kudaravalli P, Garg N, Pendela VS, Gambhir HS. Hemorrhagic pancreatic pseudocyst: A rare complication. The American Journal of Emergency Medicine. 2021;43:243-4.

13. Lunetta P, Penttilä A, Salovaara R, Sajantila A. Sudden death due to rupture of the arteria pancreatica magna: a complication of an immature pseudocyst in chronic pancreatitis. International journal of legal medicine. 2002;116(1):43-6.

14. Chiu HH, Chen CM, Wang KC, Lu YY, Mo LR. Pancreatic pseudocyst bleeding associated with massive intraperitoneal hemorrhage. American journal of surgery. 2006;192(1):87-8.

15. Botianu PVH, Dobre AS, Botianu A-MV, Onisor D. Pancreatic Pseudocyst with Splenic Artery Erosion, Retroperitoneal and Splenic Hematoma. Case Reports in Surgery. 2015;2015:981860.