Portal Vein Pseudoaneurysm with Portoenteric Fistula: An Unusual Cause for Massive Gastrointestinal Hemorrhage

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ABSTRACT

Pancreaticoduodenectomy (Whipple's procedure) is a commonly performed procedure for the treatment of pancreatic malignancies. Postoperative bleeding from adjacent arteries is a well-established complication of this procedure. This article describes an unusual case of massive gastrointestinal bleeding following pancreaticoduodenectomy due to the development of a portal vein pseudoaneurysm with a portoenteric fistula. The diagnosis was confirmed with multidetector computed tomography angiography; the different endovascular treatment options are discussed.

KEYWORDS: Portal vein, gastrointestinal hemorrhage, portoenteric fistula, pancreaticoduodenectomy complications

Objectives: Upon completion of this article, the reader should be able to summarize the endovascular treatment options for patients with portal vein pseudoaneurysms.

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Acute gastrointestinal hemorrhage is a common cause for hospital admission, with more than 300,000 hospitalizations per year in the United States. Most cases resolve spontaneously, but up to 25% of these cases require image-directed treatment due to massive hemorrhage or recurrence.¹ Clinical assessment, endoscopy, scintigraphy, abdominal angiography, and surgery are part of the workup as dictated by the pattern and rate of bleeding. A site-specific diagnosis is important in determining the most appropriate therapy and impact morbidity and mortality significantly.² In this article, a rare case of massive gastrointestinal hemorrhage resulting from portal vein pseudoaneurysm in a patient who

recently had undergone a pylorus-sparing pancreaticoduodenectomy is presented with discussion of the various diagnostic imaging and interventional procedures employed.

CASE REPORT

A 68-year-old woman presented to the emergency department (ED) with altered mental status and rectal bleeding. She had been discharged from the hospital 2 weeks earlier after having undergone pylorus-sparing pancreaticoduodenectomy for mucinous cystadenocarcinoma of the pancreatic head. This was complicated by

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anastomotic leak at the pancreaticojejunal anastomosis requiring exploratory laparotomy and abdominal wash-out. Also included in her surgical history was a right hemicolectomy performed 2 years prior to admission for colon cancer.

At the time of her presentation to the ED, her blood pressure was 110/60 mm Hg and heart rate was 125 beats per minute. On physical examination, her abdomen was soft and nondistended, and she was passing dark gross blood per rectum. Pertinent laboratory values included hemoglobin of 2.9 g/dL, hematocrit of 9.2%, prothrombin time of 19.6 seconds (normal, 11 to 14 seconds), and an international normalized ratio of 1.7. The patient was admitted to the intensive care unit, and upper endoscopy was performed. On endoscopy, blood was identified within the stomach and proximal jejunum, but an active bleeding source was not identified.

Given the patient's tenuous clinical findings, the initial radiographic evaluation consisted of visceral angiography. Selective catheterization and angiography of the celiac and superior mesenteric arteries demonstrated no evidence for arterial leak or pseudoaneurysm. However, on venous phase imaging, there was an abnormal double density overlying the main portal vein, just distal to the splenic-superior mesenteric venous confluence (Fig. 1A). This was concerning for a portal vein pseudoaneurysm, and the patient was sent to computed tomography (CT) for further evaluation. Multidetector CT (MDCT) angiogram with arterial and venous phase images demonstrated a 1.4-cm portal vein pseudoaneurysm with contrast extending from portal vein to the jejunum (Fig. 1B).

In light of the MDCT findings, different treatment options were discussed, and it was decided to proceed with surgical management. To temporize the

bleeding until the portal vein could be repaired, the surgeon requested the placement of an occlusion balloon across the portal vein defect. Therefore, transhepatic portal vein access was obtained. Direct portal venogram confirmed the portal vein pseudoaneurysm and demonstrated active extravasation into the loop of jejunum (Fig. 2). A 10-mm angioplasty balloon (Ultra-thin; Boston Scientific, Natick, MA) was placed across the portal vein defect and inflated; venography with the balloon inflated demonstrated adequate hemostatic control.

The patient was taken to the operating room for definitive surgical repair. At surgery, extensive dense adhesions were noted throughout the entire peritoneal cavity requiring 4 hours for lysis. The pancreaticojejunostomy anastomosis had nearly completely dehiscd. Once the portal vein was dissected free, a 5-cm defect along the lateral wall was discovered. Using the transhepatic balloon catheter for hemostatic control, the defect was repaired with a cadaveric vein patch. After repairing the portal vein, the patient was taken to the intensive care unit for further resuscitation and monitoring. A short time later, the patient expired secondary to continued coagulopathic hemorrhage despite resuscitative efforts.

DISCUSSION

Over the past decade, the overall mortality rate associated with pancreaticoduodenectomy has improved to 1.5%, down from historic highs of 25%.³ However, postoperative complications from pancreaticoduodenectomy remain common (43%) and include cardiopulmonary events, pancreatic fistula, delayed gastric emptying, and sepsis.³ Bleeding complications are reported to occur in 7 to 20% of patients,⁴⁻⁶ and delayed postoperative

Figure 1 (A) Venous imaging during celiac angiography demonstrates double density (arrow) at the splenomesenteric confluence suggestive of portal vein pseudoaneurysm. Abnormal increased density is seen within adjacent loop of jejunum (arrowhead). (B) Venous phase imaging during multidetector computed tomography angiography demonstrates contrast enhancement of portal vein and loops of adjacent jejunum (arrowheads). Fistula between portal vein and jejunum is clearly visualized (arrow). No oral contrast had been given.

Figure 2 (A) Transhepatic portal venogram demonstrates pseudoaneurysm at the confluence of the superior mesenteric vein and splenic vein (arrow). (B) Fistulous communication between portal vein and jejunum is demonstrated (arrow). (C) A 10-mm balloon is inflated across the fistula (arrowhead) and into the gastrocolic trunk (arrow).

bleeding, defined as bleeding after postoperative day 5, often results from pancreatic leak and pseudoaneurysm formation of the major abdominal arteries.⁷ To our knowledge, this is the first reported case of a portal vein pseudoaneurysm and portoenteric fistula presenting as a complication from pancreaticoduodenectomy.

Fistulae between the portal venous system and the gastrointestinal tract are exceedingly rare. Only two case reports are known to exist in the literature, each describing a fistulous communication between the portal vein and duodenum resulting from complications following biliary surgery⁸ and penetrating peptic ulcer.⁹ In this case, a chronic leak at a pancreaticojejunostomy anastomosis requiring laparotomy, abdominal washout, and placement of a surgical drain near the portal vein may have contributed to injury of the portal vein from both

proteolytic injury and mechanical forces from adjacent surgical drain placement (Fig. 3).

The diagnosis of portal vein pseudoaneurysm was initially suggested on the venous phase of a visceral angiogram. However, it was cross-sectional imaging that fully demonstrated the lesion, documented the portoenteric fistula, and facilitated treatment planning. MDCT angiography has become the preferred imaging modality for evaluation of nontraumatic emergent abdominal vascular complications.¹⁰ High spatial resolution volumetric image data can be obtained at a single breath hold, and perfusion in adjacent organs can be evaluated. In postoperative patients, CT angiography has the advantage of demonstrating anatomy and potential postsurgical complications. Given these advantages, the MDCT holds great promise in the

Figure 3 Computed tomography angiography of the abdomen performed 1 month earlier demonstrates a Jackson-Pratt surgical drain adjacent to the portal vein (arrow). The drain was placed during laparotomy and abdominal washout for pancreaticojejunostomy leak.

evaluation of gastrointestinal bleeding, and its utility is currently under investigation. A recent study based on a porcine model suggests that CT performed with a rapid contrast bolus can detect hemorrhage at a rate of < 0.1 mL/minute.¹¹ Another series suggests enhanced CT with water oral contrast as a useful alternative to more invasive studies in the workup of obscure gastrointestinal bleeding.¹² In this particular case, MDCT played an integral role in localizing the site of hemorrhage and guiding therapy.

Interventional radiologists play a critical role in reducing the postoperative morbidity of patients following pancreaticoduodenectomy. A retrospective review of 1061 pancreaticoduodenectomy patients reveals that 471 patients (44%) had one or more interventional radiologic procedures.¹³ Within this group, postoperative interventional procedures were performed on 129 patients (12%), including aspiration/catheter drainage of intra-abdominal abscess, biloma, or lymphocele and diagnosis and intervention for postoperative hemorrhage. Percutaneous abscess drainage was highly effective with only 4 of 84 (4.8%) patients requiring surgical drainage or repair of anastomotic leak. However, embolization of postsurgical gastrointestinal bleeding is not as effective. Within this series, 4 of 18 patients (22%) treated with angiography and embolization for hemobilia or gastrointestinal bleeding required subsequent definitive surgical repair.

The present case, though, differs from the study just cited in that the source of bleeding was from the portal vein rather than an adjacent artery. Surgical repair is currently considered the "standard" treatment for portal vein aneurysms,^{14,15} but with the current availability of a variety of endovascular tools, endovascular

repair options may be considered. Due to the location of the injury in the main portal vein, near the confluence with the superior mesenteric vein, embolization of the portal vein was considered an unattractive treatment option. An alternative to embolization is the use of a covered stent. Covered stents are accepted in the treatment of vascular injuries in other locations¹⁶ and have been used successfully in the portal vein.¹⁷ However, thus far, the use of covered stents in the portal vein have mostly been in the setting of portal vein invasion by hepatic malignancies or in the setting of transjugular intrahepatic portosystemic shunt creation^{18,19} rather than for control of acute portal vein hemorrhage.

In the current case, a covered stent might have provided short-term control of the active hemorrhage, but there were several other significant considerations. The effect of the local environment of proteolytic enzymes would have on the integrity of the fabric around the stent is uncertain. The continued exposure of the fabric of the stent could result in fabric breakdown, placing the patient at risk for recurrent hemorrhage. There is currently very little information in the medical literature confirming or refuting this assertion. In a similar situation, a hepatic artery pseudoaneurysm with an associated enteric fistula following pancreaticoduodenectomy was treated with the use of a covered stent.²⁰ In this instance, the stent graft controlled the hemorrhage; the patient had no further episodes of bleeding at 2-year follow-up.

In addition to the continued exposure to pancreatic enzymes, the associated enteric fistula introduces the risk of contamination of the stent: This may result in long-term bacterial infection. The use of covered stents in the setting of enteric fistulae is controversial. There are reports of the successful use of covered stents in such settings; these reports are mostly in the setting of the use of aortic stent grafts for aortoenteric fistulae.²¹⁻²³ These reports have shown it is possible to treat this devastating condition by endovascular means if one uses an aggressive and lifelong antibiotic regimen.²³ Nonetheless, stent-graft infection remains problematic. In a series of 7 patients with aortoenteric fistula treated with aortic stent grafts, 1 patient died of sepsis and 2 others required additional procedures due to sepsis. Given these results, the use of a covered stent would have likely only been a temporizing measure, and the patient would more than likely have required further intervention once the clinical condition stabilized.

Although neither embolization nor covered-stent placement were performed, interventional radiology played a vital role in the care of this patient. By placing a balloon catheter across the portal vein defect, the acute hemorrhage was controlled until the portal vein could be surgically repaired. This concept of using temporary balloon occlusion prior to surgery is not new; it has been used successfully in a variety of situations,

including providing acute control in patients with ruptured abdominal aortic aneurysms and prior to cesarean section in patients with placental abnormalities.^{24,25} Temporary balloon occlusion has also been used in the setting of massive hemorrhage following hepato-pancreato-biliary surgery. In one series, an aortic occlusion balloon was placed above the celiac artery, then alternatively inflated and deflated while the source of bleeding was surgically identified and repaired.²⁶ Using this technique, the hemorrhage was successfully controlled in 70% of patients.

In summary, we have described an unusual case of gastrointestinal hemorrhage resulting from portal venous pseudoaneurysm and portoenteric fistula. MDCT angiography played a critical role in localizing the site of hemorrhage and guiding therapy; this modality shows promise as an effective tool in the radiographic assessment of unusual gastrointestinal hemorrhage. In this case, surgical treatment was preferred over endovascular management. Nonetheless, interventional radiology still played a critical role by providing temporary hemostatic control with balloon occlusion of the portal vein to assist with operative repair.

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