Coil embolization of a ruptured gastroduodenal artery pseudoaneurysm presenting with hemosuccus pancreaticus

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ABSTRACT

Hemosuccus pancreaticus is a rare form of upper gastrointestinal bleeding that accounts for roughly 1 in 1500 cases. It is characterized by hemorrhage from the ampulla of Vater secondary to rupture of a peripancreatic pseudoaneurysm or visceral artery pseudoaneurysm. Among the visceral artery pseudoaneurysms, gastroduodenal artery pseudoaneurysms are among the rarest. In this case report, we describe a successful coil embolization of a large ruptured gastroduodenal pseudoaneurysm in a patient with massive gastrointestinal bleeding. (J Vasc Surg Cases and Innovative Techniques 2020;6:67-70.)

Keywords: Hemosuccus pancreaticus; Visceral artery pseudoaneurysms; coil embolization

Hemosuccus pancreaticus, hemorrhage from the ampulla of Vater, is a rare form of upper gastrointestinal bleeding (incidence of 1/1500 cases).¹ Visceral artery pseudoaneurysms secondary to chronic pancreatitis can lead to potentially life-threatening hemosuccus pancreaticus. Visceral artery pseudoaneurysms can occur in up to 10% of patients with chronic pancreatitis. However, rupture into the pancreatic duct occurs in only a minority of cases.² Gastroduodenal artery (GDA) pseudoaneurysms are among the rarest forms of visceral artery pseudoaneurysms (<2%).³ Here, we report a case of successful coil embolization of a ruptured large GDA pseudoaneurysm in a patient with massive gastrointestinal bleeding. The patient's consent was obtained for publication of this report.

CASE REPORT

A 68-year-old man with a history of chronic pancreatitis and cirrhosis presented to the emergency department with a 7-day history of melena that was associated with intermittent severe abdominal pain and shortness of breath. The patient denied any recent history of trauma or instrumentation of the abdomen, including endoscopic procedures as well as surgery.

Author conflict of interest: A.A. is a consultant for W. L. Gore and Medtronic.

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Physical examination was significant for tachycardia and hypotension: laboratory data revealed severe anemia (hemoglobin level, 4.2 g/dL) and hyperbilirubinemia (total bilirubin concentration, 2.7 mg/dL). The patient was aggressively resuscitated with transfusions and underwent an emergent esophagogastroduodenoscopy (ECD) that failed to reveal the source of bleeding. Next, computed tomography angiography (CTA) of the abdomen and pelvis was performed. It demonstrated a 4.5-cm GDA pseudoaneurysm without active extravasation. The patient was taken to the operating room for aortography and possible coil embolization under monitored anesthesia care.

Transfemoral selective common hepatic artery and superselective gastroduodenal angiography was performed. The angiogram revealed a large GDA pseudoaneurysm with a pancreatic duct fistula as the source of the gastrointestinal bleed (Fig 1). Extravasation of contrast material into the duodenum was demonstrated on delayed images and captured on cine loop (Fig 1). A 5F catheter (Clidecath; Terumo, Somerset, NJ) was placed in the common hepatic artery, and a 3F microcatheter (Renegade; Boston Scientific, Marlborough, Mass) was placed in the aneurysm sac. The aneurysm sac was packed with Interlock Fibered IDC (Boston Scientific) coils delivered through the microcatheter. Subsequently, the GDA was embolized with Fibered IDC 3-mm coils; coils were placed across the origin of the pseudoaneurysm and continued to exclude the proximal and distal portions of the GDA. Postembolization common hepatic angiography revealed successful exclusion of the sac, occlusion of the GDA, and patent hepatic arteries (Fig 2). Selective celiac and superior mesenteric artery (Fig 3) angiography revealed patent vessels with no retrograde flow into the visceral artery pseudoaneurysm. Fig 4 depicts an overview of the case, illustrating the GDA pseudoaneurysm with resultant hemosuccus pancreaticus and successful coil embolization of the pseudoaneurysm.

The patient had an unremarkable postoperative course without any further bleeding. He was discharged on postoperative day 5. Follow-up CTA confirmed continued exclusion of the CDA pseudoaneurysm. He followed up in clinic at 4 months, 1 year, and 3 years after intervention and has remained asymptomatic.

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Fig 1. Intraoperative angiography with catheter placed in the gastroduodenal artery (GDA) pseudoaneurysm **(A)**, revealing contrast material in the GDA pseudoaneurysm **(B)** and noted to be spilling into the pancreas **(C)** and extravasating into the duodenum **(D)** through the ampulla of Vater (hemosuccus pancreaticus).

DISCUSSION

Gastrointestinal hemorrhage from the ampulla of Vater through the pancreatic duct was first described in 1931⁴ and termed hemosuccus pancreaticus in 1970.⁵ Only 153 cases of hemosuccus pancreaticus have been reported in the literature, mostly case reports or limited series. Visceral artery pseudoaneurysms that develop in a setting of acute relapsing pancreatitis or chronic pancreatitis are the most common causes of hemosuccus pancreaticus (most common are splenic artery aneurysms; only five cases of GDA aneurysms have been described).⁶⁻¹⁰ Other causes of hemosuccus pancreaticus include bleeding from pancreatic parenchyma; pancreatic duct stones; iatrogenic pancreatic duct injury; and arterial fistulas that follow fine-needle aspiration or pancreatic surgery, trauma, pancreatic malignant neoplasm, and metastatic carcinoma. We describe a giant GDA pseudoaneurysm (4.5 cm) that was successfully treated by an endovascular approach with the longest reported postintervention follow-up (without relapse). The diagnosis is challenging and is usually delayed

because of the rarity of the diagnosis and the usual presence of intermittent bleeding. Intermittent epigastric pain followed by melena, hematochezia, or hematemesis within 30 to 40 minutes is a characteristic clinical presentation.¹¹ Jaundice develops secondary to pancreaticobiliary reflux due to clot formation in the pancreatic duct and is present in a few cases. As a result of intermittent bleeding of small volume, hemodynamic instability is not a common finding in most cases in hemosuccus pancreaticus. However, similar to our patient, most cases of GDA pseudoaneurysm-related hemosuccus pancreaticus presented with shock. Endoscopic visualization of bleeding from the ampulla of Vater during EGD is missed in most cases because of the intermittent nature of the bleeding, but it is characteristic when identified.¹¹ A normal finding on EGD aids in diagnosis as it rules out other causes of upper gastrointestinal bleeding. A triple-phase CTA scan of the abdomen is an excellent diagnostic modality. It demonstrates features of chronic pancreatitis, presence of pancreatic duct contrast material, and presence of



Fig 2. A, Selective common hepatic artery angiography reveals gastroduodenal artery (GDA) pseudoaneurysm. **B**, The postembolization angiogram shows exclusion of the pseudoaneurysm sac and flush occlusion of the GDA.

pseudoaneurysms/aneurysms or pancreatic mass. Selective arteriography is highly sensitive (96%)¹⁻⁶; identification of pancreatic duct hemorrhage with endoscopic retrograde cholangiopancreatography, endoscopic ultrasound, and visceral artery aneurysm on ultrasound has also been reported.⁴⁻⁸

The conventional approach has been surgical resection of the pseudoaneurysm, ligation of arteries, and pancreatic resection (pancreaticoduodenectomy or distal pancreatectomy) to treat the acute gastrointestinal bleed and to prevent recurrence. However, there is a 28% to 56% mortality rate after surgical treatment of hemosuccus pancreaticus.¹² Mortality is related to the location of the pseudoaneurysm (head vs tail of pancreas, 43% vs 16%) and recurrent postoperative bleeding (doubled in patients with rebleeding).⁶⁻¹² Coil embolization and stent graft repair have shown excellent results in multiple series for the treatment of visceral artery aneurysms and pseudoaneurysms.^{13,14} Microcoils, detachable coils, and liquid embolic materials have been reported for embolization treatment in the setting of visceral artery aneurysms and pseudoaneurysms. Coil embolization in treatment of visceral artery aneurysms and pseudoaneurysms has achieved success rates between 75% and 100%. Similar success has not been replicated in treating GDA pseudoaneurysms presenting with hemosuccus pancreaticus.¹⁵ Reinterventions and open repairs are frequently required after coil embolization in GDA pseudoaneurysms. In one series, coil embolization was attempted in four of the five cases of GDA aneurysms with hemosuccus pancreaticus. However, success was achieved in only two cases (40%), one with a small GDA pseudoaneurysm $(15 \text{ mm})^7$ and one originating from a branch of the GDA.¹⁰ Bohl et al⁶ performed coil

embolization of a large (4 cm) GDA pseudoaneurysm with hemosuccus pancreaticus. However, two additional procedures with further coil embolization in 4-month intervals were needed for recurrent bleeding. Despite the two reinterventions, the pseudoaneurysm increased in size and led to rebleeding, necessitating laparotomy, which revealed a large pseudoaneurysm arising from the GDA with active bleeding despite the presence of coils within the neck of the pseudoaneurysm.

Pancreaticoduodenectomy and ligation of GDA was performed to treat the recurrent bleeding and obstructive jaundice in the patient. We treated this large ruptured GDA pseudoaneurysm by packing the pseudoaneurysm sac with coils to obtain complete thrombosis followed by occlusion of the GDA with coils to prevent recurrence. Confirmation of absence of filling by retrograde flow was also performed during the initial treatment to avoid need for reintervention. We believe that in the setting of large pseudoaneurysms, packing the sac along with the feeding arteries is necessary to ensure successful thrombosis and treatment of the active bleeding by lessening the likelihood of recanalization of flow to the pseudoaneurysm. Worsening of pancreatitis as well as rebleeding after coil embolization has been reported; therefore, close follow-up is needed. Artifact caused by the coils on the computed tomography scan hinders accurate evaluation of persistent flow in the pseudoaneurysm. However, regression of the sac size and absence of anemia can be used as measures of success at follow-up. Our follow-up consisted of CTA of the abdomen and pelvis before discharge at the index hospitalization and at 1 month after the procedure, then physical examination and complete blood count thereafter.



Fig 3. Selective superior mesenteric artery angiography reveals successful exclusion of gastroduodenal artery (GDA) pseudoaneurysm as evidenced by lack of contrast material opacification within pseudoaneurysm sac.



Fig 4. Illustration depicting the gastroduodenal artery (GDA) pseudoaneurysm resulting in a pancreatic duct fistula with hemorrhage into the duodenum (hemosuccus pancreaticus; *left*) and successful coil embolization (*right*).

CONCLUSIONS

Visceral artery pseudoaneurysms should be included in the differential diagnosis of patients presenting with gastrointestinal bleeding. Coil embolization using microcatheter techniques is a suitable treatment option for this challenging clinical condition. Advanced embolization techniques should be included in the training and practice of modern vascular surgeons.

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