Inferior Pancreaticoduodenal Artery Aneurysm With Superior Mesenteric Artery Occlusion - An Extremely Rare Case Of Visceral Artery Aneurysms

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Citation

Abstract
Background: Although visceral artery aneurysms (VAA) are rare, they have a high potential for fatal rupture with a very high mortality rate if not promptly diagnosed and treated. Case presentation: We report an interesting case of inferior pancreaticoduodenal artery aneurysm (IPDAA) with massive retroperitoneal hemorrhage in a 71-year-old man who presented with persistent epigastric and periumbilical pain for one month. Contrast-enhanced computed tomography (CT) revealed a ruptured IPDAA with superior mesenteric artery (SMA) occlusion. The SMA occlusion had caused necrosis of the small intestines. The ruptured IPDAA was confirmed by selective abdominal angiography. Percutaneous transcatheter coil embolotherapy (PTCE) was successfully employed to treat the patient. All previously described cases of IPDAA have been associated with celiac artery occlusion or stenosis. To the best of our knowledge, this is the second reported case of IPDAA aneurysm with superior mesenteric artery (SMA) occlusion in the medical literature. The subject of this article is to present our experience with an exceedingly rare case of IPDAA and to review the medical literature on management of visceral artery aneurysms. Conclusion: Hemorrhage from VAA requires prompt diagnosis and management to prevent or decrease the morbidity and mortality associated with these aneurysms.

INTRODUCTION
Visceral artery aneurysms (VAAs) are rare but life-threatening abnormalities of the splanchnic vasculature. About 22-23% of VAAs eventually rupture resulting in an overall mortality of 8%-9% (1) The most common detected VAAs involve the splenic artery aneurysms (60%), followed by hepatic artery aneurysm (20%) (2-3). The majority of VAAs are usually seen in elderly patients with co-morbidities including hypertension and coronary artery disease. Smaller visceral vessel aneurysms such as the pancreaticoduodenal artery aneurysm (PDA) are uncommon and account for only 2-3% of VAAs (4-6). Furthermore, IPDA aneurysms are extremely rare and due to their distinct and often fatal presentation, they pose a diagnostic and management challenge to surgeons especially in emergency settings (7-16). The incidence of patients presenting with a ruptured aneurysm is usually as high as 60-75% with an operative mortality rate of 50% (17-22). PDA aneurysms are reported as either pseudoaneurysm or false aneurysm or true aneurysm depending on the nature of the aneurysm. False PDAAs are more common than true aneurysms and are often associated with acute or chronic pancreatitis, laparoscopic cholecystectomy, septic emboli and abdominal trauma; whereas true PDAAs are reported with celiac artery stenosis or occlusion and account for only 2% of VAAs (23). Pseudoaneurysms usually have a higher risk of rupture than true aneurysms. Studies have shown that larger diameter aneurysms have a higher risk for rupture. Patients with PDAAs usually present with abdominal pain, tachycardia, and jaundice. The ruptured aneurysm can lead to hemorrhage in the peritoneal cavity, retroperitoneal space, in the biliary tract or in the gastrointestinal tract (9, 11-13, 15) Due to the developments in interventional radiology and the widespread use of CT and angiography, the detections rates of IPDAAs have significantly increased. Although, surgical treatment involving either bypass or ligation has been an effective tool in the management of VAAs in the past (20-22, 24), recently, endovascular management techniques have become the treatment of choice in patients with VAAs, especially in emergency cases (25-32).
In this case report, we present a patient with inferior pancreaticoduodenal artery (IPDA) aneurysm and rupture successfully managed with PTCE.

**CASE REPORT**

A 71-year-old man presented to our hospital emergency department with persistent spasmodic epigastric and periumbilical abdominal pain associated with one episode of vomiting. The pain was not associated with indigestion. His past medical history included hypertension, atrial fibrillation, and alcohol abuse which had resulted in alcoholic cardiomyopathy. However, the patient had been abstinent from alcohol for the past two years. He had no history of pancreatitis or previous hospitalizations for similar abdominal pain. The patient denied fever, chills, diarrhea, constipations, weight changes, and previous similar episodes. On physical examination, he was found to be in severe distress and hypertensive. Upon abdominal examination, moderate epigastric tenderness on palpation with no guarding or rigidity and normo-active bowel sounds were noted. Both rectal examination and stool for occult blood were negative. Electrocardiography showed atrial fibrillation. CT without intravenous (IV) contrast material of the abdomen revealed an ill-defined area in the head of the pancreas with normal body and tail of the pancreas. Upon a subsequent drop in the patient’s hematocrit, a CT with IV contrast was performed which showed a retroperitoneal hematoma near the head of the pancreas and an occlusion in the distal part of the SMA. Extensive necrosis of the small intestines due to SMA thrombosis was also noted. Selective SMA angiography revealed a ruptured IPDA aneurysm.

The patient was successfully treated with percutaneous transcatheter coil embolotherapy (PTCE) and resection of the necrotic bowel. He was discharged from the hospital and was transferred to a rehabilitation facility and then to home.

**Figure 1**

Figures 1 and 2: Contrast-enhanced computed tomography (CT) images of the abdomen showing a retroperitoneal hematoma with IPDA aneurysm.
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DISCUSSION

Splanchnic vasculature aneurysms are uncommon surgical entities with a risk for rupture and life-threatening complications due to their elusive clinical presentation. Although treatment is proposed for both symptomatic and asymptomatic VAAs due to the unpredictable nature of aneurysm rupture, the approach to symptomatic and asymptomatic aneurysm is different (7, 8, 33). Treatment is indicated for asymptomatic VAAs with the exception of splenic artery aneurysm which are treated only when the diameter is larger than 2.5cm or when they occur in women of childbearing age or pregnant patients (34)

The gold standard for treatment of VAAs is surgery (35-37); however, endovascular techniques such as the PTCE have

Figure 3
Figure 3: CT showing the SMA clot.

Figure 4
Figures 4 and 5: “Pre” and “Post” angiograms showing SMA embolism and coil embolizations of the feeding vessels.
now become the mainstay of VAA management. Embolization has become the method of choice for symptomatic or ruptured VAA (38-41), and the first line of therapy in an emergency setting with rare fatalities and complications (40-42). It is a safer alternative to open surgical management of VAA which are associated with a high operative mortality; 36% for splenic artery aneurysms (41, 43) and 21% for hepatic artery aneurysms (44). Similarly, the operative mortality for pseudoaneurysm is 30%-50% (18).

Some of the factors leading to poor outcome of VAA include location of the aneurysm, general health status of the patients, systemic risk factors, and difficulty with surgical exposure. Although, percutaneous techniques have become very popular in the management of VAA due to their low risk involvement, it has a few disadvantages such as sepsis due to the presence of a foreign body or the embolization coils. However, success rates for PTCE have been reported to be as high as 80% (18).

In our patient, selective catheterization and microcoil embolization for the aneurysm was carried out using the femoral artery percutaneous cannulation guided with mesenteric angiography. The feeding vessels were selectively embolized using microcoils from the SMA.

CONCLUSION
The high morbidity and mortality from acute hemorrhage of ruptured VAA aneurysms emphasizes the importance of early diagnosis and endovascular management of VAA. Though prognosis of VAA depends on the aneurysm location, co-morbidity factors, and the hemodynamic stability of the patients, mortality rates still remain high for ruptured PDA and SMA aneurysms, especially after treatment (11-12, 45).

Endovascular management of VAA has shown a more favorable prognosis than open surgical repair of VAA. PTCE is an attractive therapeutic modality that has not only shown to be safer than open surgical repair but has also achieved promising results.

References
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