Obstructive Jaundice Caused by Pancreaticoduodenal Artery Aneurysms Associated with Celiac Axis Stenosis: Case Report and Review of the Literature

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Pancreaticoduodenal artery aneurysm (PDA) is quite rare, which accounts for only approximate 2% of all visceral aneurysms. Besides, PDA is usually related to celiac axis stenosis (CAS) and prone to rupture. Advanced imaging examination can facilitate the disclosure of such peripancreatic masses, but most of them were seldom diagnosed until they rupture because of the nonspecific symptoms. Secondary to PDA, obstructive jaundice is however an extremely rare manifestation. A case of an 84-year-old man is reported here, who suffered from severe jaundice caused by a ruptured PDA associated with CAS. In addition, this review collects and organizes PDAs with jaundice by applying a MEDLINE search and discusses the pathogenesis and therapeutic options of these aneurysms leading to external compression over the bile duct. Consequently, the formation of PDA with obstructive jaundice is based on the specific anatomy of pancreaticoduodenal arcades. When there is a retroperitoneal mass around the head of the pancreas associated with unexpected jaundice, PDA should be considered, for which early aggressive therapy is required. The case report and literature review suggest that PDA associated with obstructive jaundice may be treated successfully by single transcatheter arterial embolization (TAE) without auxiliary biliary drainage, whether it ruptures or not.

Pancreaticoduodenal artery aneurysms (PDAs) are quite rare, which account for only 2% of all visceral artery aneurysms.[1] Since Sutton and Lawton [2] firstly put forward the relation between CAS and PDA, approximate 63% of reported cases had been found to be related to CAS.[3] More than 60% of PDAs are shown to be at rupture, and emergent treatments are required.[4] Although advanced imaging examination facilitates the disclosure of such peripancreatic masses, most of them were seldom diagnosed until they rupture because of the nonspecific symptoms. Secondary to PDA, Jaundice is however an extremely rare manifestation.

A case of an 84-year-old man is reported here, who suffered from severe jaundice caused by a ruptured PDA associated with CAS. In addition, this review collects and organizes PDA with jaundice by adopting a MEDLINE search and discusses the pathogenesis and therapeutic options of these aneurysms leading to external compression over the bile duct.

CASE REPORT

An 84-year-old man was admitted to the local hospital, who suffered from a sudden onset of abdominal pain in the right upper quadrant. There was no history of abdominal trauma, chronic pancreatitis, bile duct calculus, or alcohol abuse, but he had suffered from hypertension for 10 years. On admission, the blood pressure was 85/54 mm Hg, and an immediate ultrasound showed that

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there was a retroperitoneal mass. Subsequently, an urgent laparotomy was performed and the gigantic mass was found to be a retroperitoneal hematoma. The hematoma was located in the peripancreatic region without a clear limit out of the surrounding tissue, which had high surface tension with the tendency to rupture, but no bleeding focus was found. On account of the limitations in local technology and experience, further dissection around the mass was difficult and risky. Considering the patient safety, the surgeons set up a drainage tube and the operation was terminated.

The advanced age patient was intubated during the laparotomy and extubated the next day. Postoperative vital signs stabilized without any vasoactive substance, but he was transferred to the hospital because of unexpected jaundice. Subsequent laboratory analysis revealed that a serum total bilirubin was at 24.4 mg/dL, direct bilirubin at 21.5 mg/dL, alanine transaminase at 60 μ/L, and gamma glutamyl transpeptidase at 84 μ/L, which indicated severe obstructive jaundice. Additionally, abdominal contrast-enhanced computed tomography (CT) and CT angiography showed that retroperitoneal hematoma was secondary to ruptured pancreaticoduodenal artery aneurysm with celiac axis stenosis (Fig. 1A–C). Angiography under local anesthesia confirmed the existence of PDA with a diameter of 2 cm, which is derived from the first branch of the superior mesenteric artery on the right side (Fig. 2A–C).

In the meantime, the initial involvement of the celiac axis was obviously narrowed, which was compensated by vascular revascularization via the pancreaticoduodenal artery arcades. A guiding catheter was inserted to the branch of the superior mesenteric artery, and superselective arterial embolization was successful by placing 2 microcoils in the proximal portion of the aneurysm (Fig. 3A). However, it was infeasible to embolism the distal main vessel, for there was no definitive vessel that supplied blood to the aneurysm. Arteriography of SMA (Fig. 3B) shows that the aneurysm had disappeared, and selective angiography of the gastroduodenal artery via celiac axis confirmed similarly that there was no ''back'' bleeding from the distal pancreaticoduodenal artery branch (Fig. 3C).

The patient was returned to intensive care unit, and the jaundice subsided quickly after endovascular treatment. There were no further signs of bleeding from the aneurysm during admission. Unfortunately, the patient was subjected to multidrug-resistant respiratory infection and died from breathing failure 26 days after embolization.

DISCUSSION

As an uncommon vascular disease, PDA was first reported in 1895 by Ferguson.[5] These aneurysms can be divided into the true aneurysms frequently related to celiac axis stenosis and the pseudoaneurysms whose underlying cause is the peripancreatic inflammation or trauma involving chronic pancreatitis and abdominal injury. According to Kallamadi et al.,[6] abdominal pain is the most common initial symptom of PDA. However, it is difficult to establish an early diagnosis of PDA until it ruptures, which results in a mortality rate of 26%.[3] Most of them usually rupture into the retroperitoneal space around the head of pancreas, which simulates the peripancreatic mass. In addition, whether to rupture is hardly dependent on the size of aneurysms (the median diameter is 22.2 mm in patients with rupture, 21.4 mm to the contrary), but dependent on the propensity for rupture itself.[3,7–9] Both a gigantic aneurysm itself and a secondary retroperitoneal hematoma can produce oppression over surrounding tissues, which leads to obstructive jaundice uncommonly. In this regard, retroperitoneal mass around the head of the pancreas with jaundice can arouse the awareness of PDA, and early aggressive therapy is required. ''jaundice'', ''iterus'', ''pancreaticoduodenal'', ''aneurysm'' and ''pseudoaneurysm'' are regarded as key words and searched in the MEDLINE database by electronic and manual searches. As a result, totally 8 cases of PDA associated with jaundice are collected and organized in Table 1.[9–15]

Unexpected jaundice is based on the specific peripancreatic anatomy (Picture 1). The pancreaticoduodenal arcades are around the head of pancreas and formed by the branches of celiac axis and superior mesenteric artery (SMA), which supply corresponding tissues and organs. The anterior and posterior of the superior pancreaticoduodenal artery rooted in gastroduodenal artery are connected with the anterior and posterior of the inferior pancreaticoduodenal artery derived from SMA, respectively. There are several rami communicantes between the celiac axis and SMA, such as the Arc of Buhler and the Arc of Barkow.[17–19] Because of CAS and provided by the common hepatic artery, the flow to the liver, spleen and stomach, spleen artery, and left gastric artery is compensated by the arcades that serve as a retrograde bypass pathway. As a result, the accompanied arteries dilate, which is supposed to be PDA. It usually gathers on the duodenal ampulla along with the pancreatic duct behind the head of the pancreas, whereas the distal common bile duct tends to be compressed by the unruptured PDA or the hematomas caused by rupture. The bulky mass exerts direct pressure over the common bile duct, the head of the pancreas, and duodenum, which results in obstruction. As in our case, a giant retroperitoneal hematoma oppresses the extrahepatic duct, which