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Case Report

Spontaneous cessation of postpancreatectomy hemorrhage in a patient with celiac artery stenosis☆

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ABSTRACT

Postpancreatectomy hemorrhage (PPH) is a severe complication of pancreatic surgery. This condition often develops on the background of pancreatic fistula. In our report, we demonstrate an unusual case of spontaneous cessation of severe postpancreatectomy common hepatic artery (CHA) hemorrhage after distal pancreatectomy in a patient with celiac artery stenosis (CAS). A 64-year-old male diagnosed with pancreatic tail tumor underwent extended distal pancreatectomy. He developed pancreatic fistula and was discharged with an abdominal drain, and was readmitted with severe postpancreatectomy hemorrhage from a pseudoaneurysm of the CHA. The bleeding stopped spontaneously due to CHA thrombosis. The patient developed no ischemic symptoms. Spontaneous severe postpancreatectomy hemorrhage cessation is an extremely rare phenomenon. Vascular anomalies must be considered when attempting interventional radiology treatment. CHA probably may be sacrificed with no ischemic consequences in case of severe hemorrhage in patients with CAS.

Introduction

Intraabdominal hemorrhage is one of the deadliest complications in pancreatic surgery. According to International Study Group of Pancreatic Surgery classification, postpancreatectomy hemorrhage (PPH) should be divided into early, occurring during the first 24 hours after initial operation, and late [1]. Early hemorrhage is usually related to inadequate intraoperative hemostasis and managed mostly via relaparotomy.

Late hemorrhage develops usually with the underlying pancreatic fistula or biliary leak [2], and its management involves interventional radiology, that is preferred over operative management due to unacceptably high mortality rates after relaparotomy in these patients (22% vs 47%, respectively) [3]. The incidence of late postpencreatectomy hemorrhage is 3%-16% (mean, 5%) [4]. Conservative management is usually ineffective in the late hemorrhage [5]. There are no guidelines for the management of PPH, although different authors proposed their treatment algorithms [5–7].

Celiac artery stenosis (CAS) has been found in 2%-7.6% in patients undergoing pancreatic surgery [8]. In cases of pancreatic head resection, CAS often requires some type of arterial reconstruction to reestablish hepatic arterial flow after disrupted superior mesenteric artery (SMA)—pancreaticoduodenal arcs—gastroduodenal artery collateral axis. The significance of CAS in cases of distal pancreatic resection has not been described. Here, we present the case of distal pancreatectomy in a patient with CAS, who developed late PPH from the common hepatic artery that stopped spontaneously.

This case has been reported in the line with the SCARE Criteria [9].

Case description

A 64-year-old male without individual and family history of malignancy was referred to our department with nonspecific complaints of mild upper abdominal pain. He underwent abdominal US in his regional hospital, which revealed a mass in his pancreatic tail, followed by contrast-enhanced multiphase CT—a hypoenhancing 3.5 × 2.5 cm lesion in the pancreatic tail with irregular contour confirmed the diagnosis of pancreatic tumor (Fig. 1). His CA-19-9 level was 102.6 U/ml (reference range 0-37). Upper endoscopy was normal, colonoscopy finding were colonic polyps amenable to endoscopic excision. Polypectomy was decided to be performed after pancreatic tumor treatment. CBC, liver and kidney function tests, coagulation test were within normal ranges. After multidisciplinary discussion the patient was offered surgical treatment. During preoperative CT-scan evaluation the replaced right hepatic artery from the superior mesenteric artery (Michel's type 3), celiac artery stenosis, as well as pancreaticoduodenal arcades hypertrophy were noted (Fig. 2).

Radical antegrade modular pancreatosplenectomy (modified distal pancreatectomy) was performed. Operation time was 4 hours, blood loss 400 ml. Pancreatic stump was closed in "fish-mouth" fashion. Pathology revealed moderately-differentiated pancreatic ductal adenocarcinoma of the pancreatic tail, without metastases in regional lymph nodes.

Patient received standard postoperative care: early initiation of enteral feeding, mechanical and pharmacologic thromboprophylaxis, NSAIDs. On postoperative day 3, he developed abdominal lymphorrhea (confirmed by high triglyceride level in milky-appearance abdominal drain fluid), which was successfully managed with low-fat diet. On postoperative day 5, the patient developed delayed gastric empting symptoms and an episode of hyperthermia up to 38.3'C. CT-scan detected
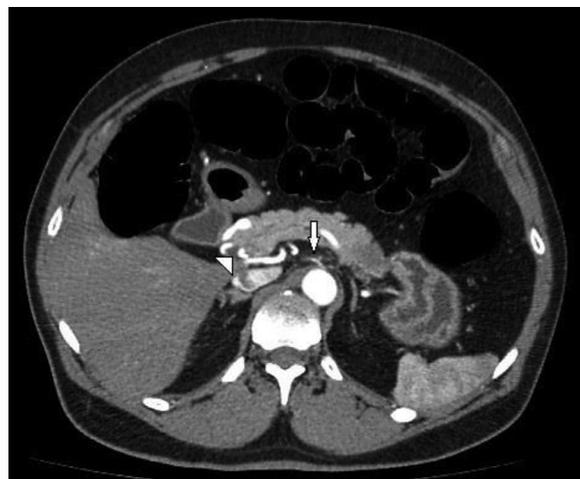
Fig. 1 – Contrast-enhanced CT-scan, pancreatic phase: a hypoenhancing mass in the pancreatic tail without vascular invasion; celiac artery stenosis (*arrow*); enlarged, tortuous pancreaticoduodenal arteries (*arrowhead*).
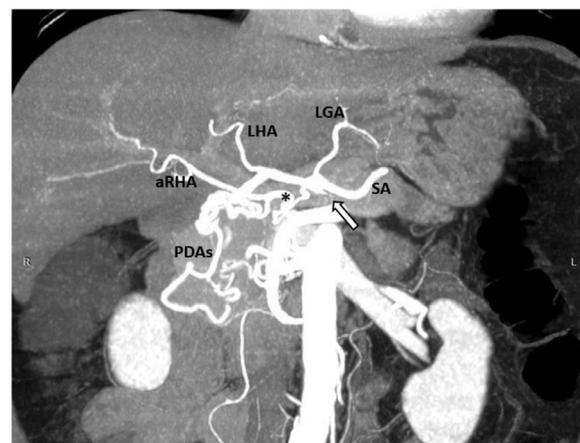
Fig. 2 – Preoperative CT-angiography. Celiac artery stenosis (*arrow*) with apparent enlargement of pancreaticoduodenal arcades (PDAs) providing collateral flow from superior mesenteric artery to common hepatic artery. Aberrant right hepatic artery (aRHA; *asterisk* – an origin of the vessel) consisting with Michel's type 3 anatomy. LGA, left gastric artery; SA, splenic artery.

peripancreatic fluid collection, which was drained. Serial amylase levels in the exudate confirmed postoperative pancreatic fistula diagnosis. The patient was discharged on postoperative day 13 with the drain.

On postoperative day 21, the patient developed hemorrhage through the drain with dizziness and Hb drop from 11 to less than 8 g/dL. He was readmitted and conservative treatment with red blood cells transfusion, fresh frozen plasma, tranexamic acid started. The hemorrhage continued, although non-massive. On day 22, he underwent angiography in an interventional radiology unit. We revealed the source of bleeding
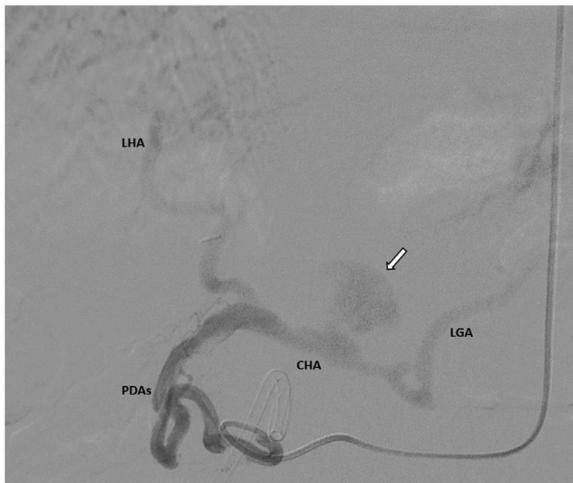
Fig. 3 – Angiography, an angiocatheter in introduced into inferior pancreaticoduodenal artery. Apparent contrast filling of common hepatic artery (CHA) and left hepatic artery (LHA) through pancreaticoduodenal arcades (PDAs). A pseudoaneurysm of common hepatic artery is visualized (*arrow*). LGA, left gastric artery.

Fig. 4 – Angiography, an angiocatheter in introduced into the origin on aberrant right hepatic artery (aRHA) arising from superior mesenteric artery. Communicating vessels between aRHA and pancreaticoduodenal arcades (PDAs) are apparent, representing additional collaterals to common hepatic artery (CHA). LHA, left hepatic artery; *arrow* – pseudoaneurysm.

to be a pseudoaneurysm of the CHA, but it was unavailable for catheterizing due to celiac artery stenosis (Figs. 3 and 4). Surprisingly, after angiography, although being non-curative, the bleeding stopped. On postoperative day 31, CT-scan revealed a total thrombosis of common hepatic artery with well-developed arterial collaterals from the superior mesenteric artery to the proper hepatic artery in the pancreatic head, and hematoma under the left lateral section of liver (Fig. 5). The patient was followed up for additional 5 days and discharged home without the abdominal drain.
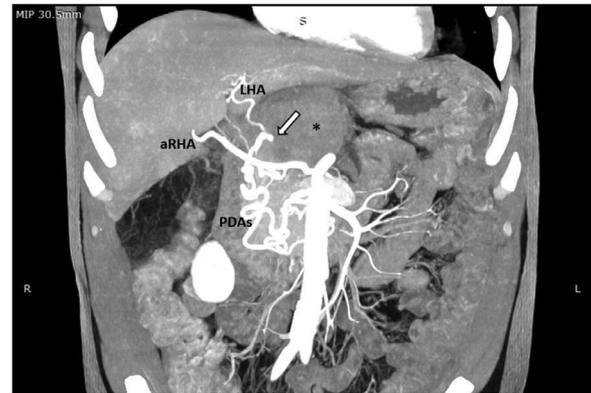
Fig. 5 – Postoperative CT (MIP 30.5 mm window). A large hematoma (*asterisk*) under the left lateral liber section. Common hepatic artery is not contrast enhanced (thrombosed) and only its terminal segment is seen as a blunt-ended vessel (*arrow*). Left gastric artery is also occluded. PDAs, pancreaticoduodenal arcades; aRHA, aberrant right hepatic artery; LHA, left hepatic artery.

After 6 weeks postoperation, the patient started adjuvant mFOLFIRINOX chemotherapy and received 12 cycles. Control CT-scans after 3 and 6 months showed the completely occluded common hepatic artery and an even increased vascularity of pancreatic head collaterals (Fig. 7). After 12 months of follow-up, the patient is asymptomatic and without evidence of recurrence.

Discussion

As stated above, late PPH is a dangerous complication rarely treated with conservative measures. Yekebas et al. [5] published their experience of treatment of PPH in the group of 1669 pancreatic resections. Conservative treatment was initiated in 17 patients, but only in 4 it was effective, while in others initially successful hemostasis appeared to be the "time gap" after the sentinel bleeding. Thus, interventional radiology is stated to be the procedure of the first choice by most authors, given that it provides comparable rate of effective hemostasis and significantly lower mortality that relaparotomy [3]. There are no guidelines regarding the method to be preferentially used for angiographic hemostasis, although most published data (yet being nonrandomized) suggest stent-grafts to be more effective in common hepatic artery pseudoaneurysms than embolization techniques [10–13]. Contraindications for stent-graft implantation are believed to be short and large-neck pseudoaneurysms, tortuous, stenotic or small vessels [13]. We didn't succeed in angiographic hemostasis in the case because of inability to catheterize the CHA due to CAS. Although, Ikeda et al. [14] described their experience of hepatic artery chemoembolization in hepatocellular carcinoma in patients with CAS. In 5 cases, proper hepatic artery was catheterized via dilated pancreaticoduodenal arcades.

Fig. 6 – Postoperative CT (MIP 30.4 mm window). An apparent communication (*arrow*) between aberrant right (aRHA) and left (LHA) hepatic arteries

Fig. 7 – Postoperative CT after 3 months (MIP 46.3 mm window). Common hepatic artery occlusion remains (*arrow*). Active collateral flow through pancreaticoduodenal arcades (PDAs) into left hepatic artery (LHA). Patent aberrant right hepatic artery (aRHA, *asterisk* – the vessel's origin).

Spontaneous hepatic artery thrombosis is an extremely rare emergency, presenting with liver infarction. To the date, there are only 4 cases described in literature [15–18]. Rate of asymptomatic thromboses is unknown, although it is clear that liver arterial exclusion without sufficient collaterals leads to severe and often fatal consequences.

Collateral arterial pathways in patients with CAS have been extensively studied by Song et al. [19]. They stated that the most common pathway both in normal and aberrant hepatic arterial anatomy is pancreaticoduodenal arcade that is present in 95%. The number of anterior and posterior arcades variates from 1-4 that is prominent in our patient who clearly has multiple arterial axes in his pancreatic head. The second most common pathway is via the dorsal pancreatic artery. It can be clearly seen in the patient's preoperative CT as an anastomosis between celiac artery and superior mesenteric artery, although is absent on postoperative CT and angiography, probably being disrupted intraoperatively. Less common and unique for the aberrant right hepatic artery originating from the SMA is the presence of intrahepatic interlobular collaterals, also seen in our patient (Fig. 6).

Normally, these vascular abnormalities used to lead to a reverse bloodstream in the CHA towards the splenic artery to supply the pancreatic tail and the spleen. The phenomenon of reverse flow in CHA and gastroduodenal artery in CAS was shown by Mano et al by the means of contrast-enhanced 3-dimensional magnetic resonance angiography [20]. After the distal pancreatectomy, the bloodstream through the CHA seized and it turned to a blunt-ended vessel. The well-established collateral liver supply and the absence of axial bloodstream through the pseudoaneurysm-bearing vascular segment provided a possibility for spontaneous asymptomatic arterial thrombosis.

The patient's final arterial hemodynamics resemble those after modified Appleby procedure (distal pancreatectomy with celiac axis resection). Specific complications following this procedure are partial liver infarction (7%) and ischemic gastropathy (29%) [21]. Holoyda et al. reported a case of modified Appleby procedure in a patient with preceding CAS and pancreaticoduodenal collaterals, and stated that such patients are the best candidates for this surgery due to the intact hepatic flow [22].
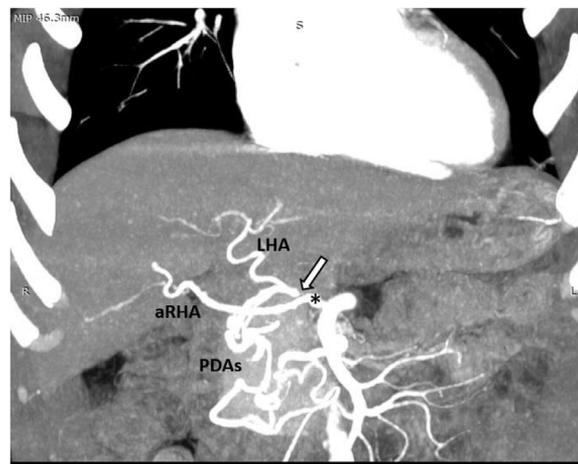
Long-term vascular outcomes after modified Appleby procedure have not been studied yet due to relatively small number of long-term survivors. A well-known complication of CAS is pancreaticoduodenal artery aneurysm formation, with the incidence ranging from 45% to 67% [23]. These aneurysms prone to spontaneous rupture, that occurs in 7%-15% [24] that is associated with significant mortality (up to 75%) [25]. Although no guidelines exist regarding the follow-up of patients with CAS and those after celiac artery resection, one should be familiar with this complication and it must be treated as detected.

The management of our patient's postoperative complication was carried out in accordance with existing recommendations and literature data, although we received nonpredicted results due to the vascular anomaly. We can't state that conservative "watch and wait" approach should be utilized in cases of bleeding from CHA in patients with collateral hepatic flow due to CAS, but can conclude that CHA may be sacrificed (either by IR distal and proximal embolization or operatively) in these patients. Tactics should base on hospital resources and anatomical and clinical features of the patient.

Patient consent

The written patient's consent has been obtained.

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