



Hemobilia due to an Arterio biliary Fistula Following Duodenopancreatectomy: A Case Report and Literature Review

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Introduction

Hemobilia, defined as the presence of blood in the bile ducts, is a rare condition that poses a diagnostic challenge due to its ability to mimic other acute abdominal conditions. Its clinical manifestations often include Quincke's triad: jaundice, abdominal pain, and gastrointestinal bleeding. The etiology of hemobilia is diverse, ranging from trauma, malignancies, and inflammatory processes to medical procedures like liver biopsies or surgical interventions. Bleeding can also result from a vascular injury that connects the hepatic artery or other blood vessels to the biliary system. The intricate nature of the biliary and vascular systems makes the pathophysiology of hemobilia particularly complex. A multidisciplinary approach, involving imaging techniques like CT scans, MRIs, and angiography, is often necessary to identify the bleeding source [1].

Postpancreatectomy hemorrhage (PPH) is a serious complication that can occur after pancreatic surgery, especially pancreatoduodenectomy (PD). Due to the complex vascular anatomy around the pancreas and bile ducts, PPH can present in various forms, sometimes mimicking other conditions [2,3]. This article presents a detailed case study that highlights the complexities of diagnosing and managing PPH when it manifests as hemobilia, offering insights into its clinical implications and the importance of a thorough postoperative evaluation.

Case Presentation

We report the case of a 70-year-old male admitted with new-onset, painless jaundice and elevated liver enzymes. Examinations suggested a possible distal cholangiocarcinoma of the common bile duct. The patient had a history of insulin-dependent type 2 diabetes mellitus and arterial hypertension. The interdisciplinary tumor board recommended a pylorus-preserving pancreatic head resection, which was performed as a pylorus-preserving PD (PPPD). The procedure revealed an anatomical variant with a large common hepatic duct and an additional posterior bile duct, both of which were transected.

The surgical reconstruction included a pancreaticojejunostomy, a hepaticojejunostomy, and a duodenojejunostomy. The right hepatic artery had to be mobilized due to its close proximity to the common hepatic duct. The bile duct anastomosis was performed 15 cm distal to the pancreatic anastomosis by suturing the two ducts together to form a common ostium. Two Jackson-Pratt drains were placed, one for the biliodigestive anastomosis and one for the pancreatic anastomosis. The initial postoperative period was uneventful, and the drains were removed on the seventh day. Histological analysis confirmed adenocarcinoma of the ampulla of Vater pT3 pN1 (1/10) cM0 R0. However, discharge was delayed due to unstable blood sugar levels. On the 13th postoperative day, the patient's condition worsened, with decreased appetite, dyspnoea, productive cough, vomiting, and elevated inflammatory markers. A CT scan ruled out pneumonia but revealed a large intra-abdominal fluid collection (58 mm × 62 mm) with air bubbles near the residual pancreas, suggesting an abscess or postoperative pancreatic fistula (POPF) and ascites. Intravenous antibiotics were started, and sonographically guided drainage of ascites revealed a lymphatic fistula. At this time a puncture of the collection was because of technical difficulty and necessarily transhepatic puncture not performed. A follow-up CT on day 16 showed the fluid collection was stable, but inflammatory markers remained high. On the 17th postoperative day, a transhepatic abscess puncture was attempted but aborted after aspirating blood. An emergency CT revealed a new contrast-enhancing structure, 10 mm in diameter, suggestive of a right hepatic artery aneurysm (Figure 1). No active hemorrhage or hematoma was detected. The oesophagus and stomach appeared to be fluid-filled. A

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Figure 1: Pseudoaneurysm of the right hepatic artery.

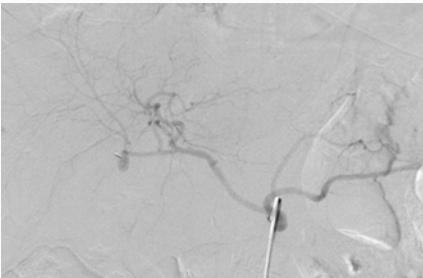


Figure 2: Pseudoaneurysm of the right hepatic artery.

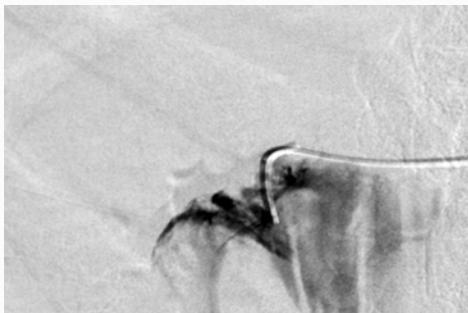


Figure 3: Perforation of the aneurysm into the surrounding soft tissues.

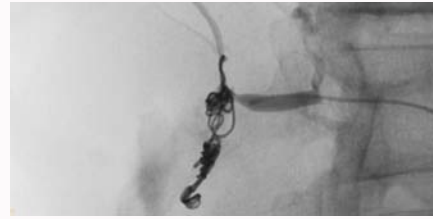


Figure 4: Coil loops extravascular.

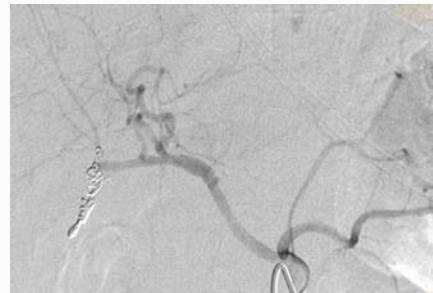


Figure 5: Final examination after first embolization.

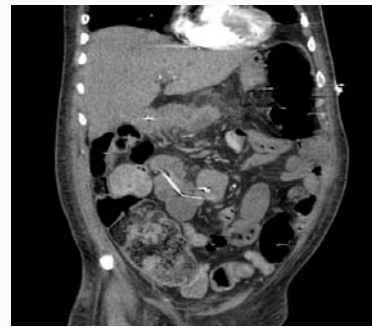


Figure 6: Dislocation of the angiographically placed coils into the biliopancreatic jejunal loop.

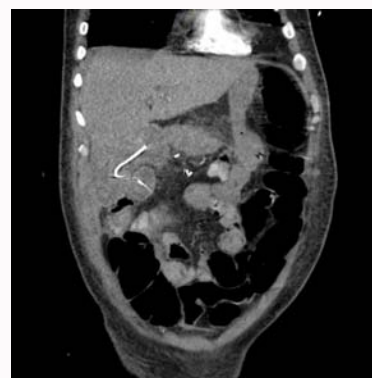


Figure 7: Suspicion of anastomotic insufficiency at the biliodigestive anastomosis.

nasogastric tube revealed blood in the stomach, indicating an upper gastrointestinal hemorrhage. Emergency gastroscopy showed clotted blood but no active bleeding source. Angiography was performed, and a pseudoaneurysm of the right hepatic artery was embolized (Figure 2). During the intervention, a perforation of the aneurysm occurred into the surrounding soft tissues (Figure 3), resulting in several coil loops being extravascular (between the small intestine and the liver, and in the aneurysm); there was no contrast observed in the intestinal loops (Figure 4). Individual coil loops were noted in the right hepatic artery, while the vessel was strongly contrasted and appeared unchanged upon final examination (Figure 5). A follow-up CT on day 18 showed no bleeding sources but a stable fluid collection near the pancreas. We initiated a CT-guided retroperitoneal drainage of the abscess. During the intervention in the prone position, the patient was intubated. The CT-guided retroperitoneal drainage was aborted due to the patient's deteriorating condition and increased blood loss through the gastric tube. A subsequent CT showed a growing hematoma in the hepatic hilum and dislocated coils in the biliopancreatic jejunal loop (Figure 6). Considering these findings, we indicated the need for relaparotomy due to suspicion of anastomotic insufficiency at the biliodigestive anastomosis (Figure 7). Intraoperatively, no acute

hemorrhage or anastomotic insufficiency was found. The dislocated coils were successfully extracted. Despite stabilization, bloody discharge from the gastric tube recurred on day 22. A CT scan again showed no active bleeding. An endoscopy revealed bleeding from the afferent loop and a gastric ulcer. A second relaparotomy was performed due to recurrent intraluminal bleeding in the region of



Figure 8: Pseudoaneurysm of the right hepatic artery with a fistula into the bilio-digestive anastomosis.

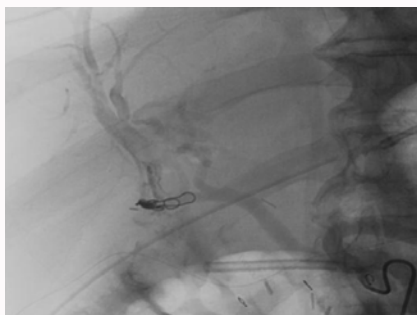


Figure 9: Final aspect after second embolization.

the biliopancreatic loop. The pancreatic anastomosis was disrupted, and an external pancreatic duct drainage was placed. On day 24, a drop in hemoglobin levels prompted another angiography. This time, the angiogram directly visualized the contrast medium flowing from the pseudoaneurysm into the bile duct and jejunal loop, confirming a fistula between the pseudoaneurysm of the right hepatic artery and the biliodigestive anastomosis (Figure 8). The pseudoaneurysm was successfully managed with interventional radiological embolization (Figure 9). The patient's condition stabilized, and he was discharged on the 46th postoperative day. A control CT scan showed normal liver perfusion. After discharge, he underwent six cycles of chemotherapy. Six months later, the pancreatic duct drainage was accidentally lost. A subsequent CT showed no pancreatic duct dilation or fluid collections. Given the patient's insulin-dependent diabetes and the potential functional loss of the pancreas, a conservative approach was chosen over another major surgery. The patient has been under follow-up care for five years with no recurrence or metastasis.

Discussion

Hemobilia is the presence of blood within the biliary tree, often resulting from vascular injuries during surgical procedures [1]. This rare but significant complication presents diagnostic and management challenges. Our case highlights the association between hemobilia and postpancreatectomy hemorrhage (PPH), which warrants comprehensive discussion.

PPH is a life-threatening complication of pancreatic surgery. Studies have shown that performing pancreatic surgeries in high-volume centers improves outcomes. Postoperative pancreatic fistula and bile leakage are major risk factors for severe PPH [4-6].

The definition of PPH is based on three parameters: onset, location, and severity. The timing of PPH is classified as early (within 24 hours) or late (after 24 hours). Hemorrhage can be intraluminal or extraluminal, and its severity is graded as mild or severe. An objective, universally accepted definition and clinical grading system for PPH are essential for guiding appropriate management and interventions [7].

Early PPH is typically caused by inadequate hemostasis, while late PPH is often associated with erosive factors like anastomotic insufficiency or intra-abdominal infections [8,9].

Late PPH is further categorized into erosive and non-erosive forms. According to a study by Feng et al. [8], non-erosive late haemorrhages typically commence around two weeks postoperatively, whereas erosive haemorrhages tend to occur between postoperative days 5 and 14. The same study identified the hepatic artery as the most common bleeding source in cases of non-erosive haemorrhage, accounting for 83.33% of cases, with none originating from the pancreaticojejunostomy. A plausible pathophysiological mechanism for non-erosive PPH involves pseudoaneurysm formation following surgical injury to blood vessels, with subsequent rupture precipitated by sudden elevations in blood pressure. None of the patients with non-erosive bleeding suffered from rebleeding after complete haemostasis. In the group with erosive haemorrhage 44% of the patients underwent a second bleeding after initial haemostasis [8].

Another important concept when we talk about late postpancreatectomy bleeding is the sentinel bleeding. It's often characterized by minor blood loss via abdominal drains or the gastrointestinal tract. A "sentinel bleed" can precede a major hemorrhage and is a critical warning sign of a potential vascular defect. The observation of sentinel bleeding should lead to emergency angiography [10,11].

Regarding the management of postoperative pancreatic haemorrhage (PPH), Yekebas and colleagues, along with Izbicki et al., have established widely accepted guidelines and proposed a therapeutic algorithm based on their study of 1,669 consecutive pancreatic resections. Their findings indicate that early extraluminal bleeding—primarily due to inadequate haemostasis—generally occurs within 24 to 48 hours postoperatively and necessitates prompt reoperation without diagnostic delay [2].

For intraluminal PPH, endoscopy may be appropriate in the first few days, but late-onset cases, especially those with pancreatic fistula, warrant angiography [2].

Angiography is the initial intervention for late PPH, regardless of whether the bleeding is intraluminal or extraluminal. Re-angiography can be considered if the patient is stable, and the bleeding source is not found. Re-angiography can be performed within 6 to 24 hours [2].

Surgical intervention is reserved for cases where angiography fails or is not feasible [2,8].

The primary causes of hemobilia are iatrogenic, traumatic, and neoplastic. Haemobilia presents as bleeding within the upper gastrointestinal tract. The common iatrogenic causes of haemobilia include percutaneous procedures and endoscopic interventions involving the hepatopancreatobiliary system. A less common iatrogenic cause of haemobilia involves surgical procedures. Both laparoscopic and open surgeries conducted near the cystic and right

hepatic arteries may result in injury to these vessels or adjacent structures, potentially leading to bleeding into the biliary system, through pseudoaneurysm formation. Bile itself can damage the endothelial lining of blood vessels, and surgical injury to both arterial and biliary tissues can impair healing and increase the risk of pseudoaneurysm formation. The right hepatic artery is the most affected vessel. Over the years, the primary cause of haemobilia has transitioned from trauma-related origins to iatrogenic factors. Consequently, radiologic procedures have emerged as the preferred method for both diagnosing and treating ongoing or unstable haemobilia, establishing them as the gold standard in managing this condition [3].

A review of the literature did not identify any other cases of late PPH presenting as hemobilia due to a fistula between a right hepatic artery aneurysm and the biliodigestive anastomosis. However, similar complex cases have been reported.

Zeyara et al. described a late PPH from a gastroduodenal artery stump into an insufficient hepaticojejunostomy [12]. The bleeding source was through an angiography detected and treated.

Gachabayov et al. reported a case of recurrent haemobilia secondary to a right hepatic artery pseudoaneurysm. The patient, who had previously undergone laparoscopic cholecystectomy, was readmitted on the 15th postoperative day presenting with Quincke's triad—gastrointestinal bleeding, right upper quadrant pain, and jaundice. Contrast-enhanced computed tomography (CT) imaging identified a pseudoaneurysm of the right hepatic artery. The patient was subsequently treated with transarterial embolization of the right hepatic artery. However, on the sixth postoperative day following embolization, the patient developed recurrent abdominal pain, and on the seventh postoperative day, melena recurred. Repeat contrast-enhanced CT imaging revealed enlargement and recurrence of the right hepatic artery pseudoaneurysm. The patient then underwent urgent laparotomy with ligation of the right hepatic artery [13].

We identified two cases of arterio-biliary fistula complicating endoscopic retrograde cholangiopancreatography (ERCP). One such case was documented by Mathur et al., who described haemobilia resulting from an arterio-biliary fistula following ERCP performed for residual cystic duct stones in a patient with Mirizzi syndrome. The management of this case was surgical. Intraoperatively, a dense adhesion was observed between the posterior surface of the common hepatic duct and the anterior surface of the right hepatic artery. Consequently, the right hepatic artery was ligated to control the bleeding. The biliary continuity was restored via a hepaticojejunostomy [14].

The second case was documented by Welsch et al. and involved a 76-year-old patient who developed significant haemobilia due to intrahepatic bleeding originating from the right hepatic artery segment 8. Notably, there was no evidence of a true aneurysm, abscess, or metastatic disease. This complication occurred four weeks after a pylorus-preserving total pancreatectomy performed for pancreatic cancer. Initial gastroduodenoscopy suggested haemorrhage from the duodenojejunostomy site; however, haemostasis was not achieved endoscopically, necessitating exploratory laparotomy. Intraoperative findings revealed that the source of bleeding was within the intrahepatic biliary tract. Haemorrhage was controlled by occluding the right hepatic bile duct with a Fogarty catheter, followed by transarterial embolization. Computed tomography imaging did

not demonstrate any localized hepatic or vascular pathology. Upon retrospective analysis, it is most probable that the delayed, profuse haemobilia resulted from a traumatic intrahepatic pseudoaneurysm, likely induced by the endoscopic bile duct stenting performed three weeks prior to the pancreatectomy [1].

Conclusion

Our case highlights the complex and often elusive nature of PPH, especially when it presents as hemobilia due to an arterio-biliary fistula. The interplay between vascular injury, anastomotic integrity, and postoperative complications such as pseudoaneurysm formation creates a diagnostic challenge that requires a high index of suspicion and a multidisciplinary approach. Vigilant postoperative monitoring, early recognition of sentinel bleeding, and the use of advanced imaging like CT angiography and interventional radiology are crucial for diagnosis and treatment.

In conclusion, awareness of potential vascular-biliary fistula formation after pancreatic surgery is essential for surgeons, radiologists, and gastroenterologists. A comprehensive, multidisciplinary strategy combining clinical assessment, advanced imaging, and interventional therapies is fundamental for effective management and improved patient outcomes.

This report underscores the rarity of such a complication, highlighting it as a significant diagnostic and therapeutic challenge.

Long-Term Outcomes

The long-term follow-up of this patient expands our understanding of the postoperative course after complex pancreatic surgery. The patient's continued stability without recurrence of malignancy or need for further surgery highlights the importance of prolonged surveillance. The decision for conservative management, particularly in a patient with significant comorbidities, illuminates the complexities of treatment planning in these challenging cases.

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