

Two Distinct Episodes of Life-Threatening Hemobilia Due to a Lesion of Common Bile Duct and Delayed Intrapancreatic Arterio biliary Fistula Managed by Emergency Pancreatoduodenal Resection

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Abstract

Hemobilia is an extremely rare cause of upper gastrointestinal bleeding. It often has intermittent manifestation, which may lead to significant diagnostic delay. In 65% of the cases, the causes are iatrogenic, in 7% the cause is malignancy, in 5% - gallstones, in 8% it is inflammation (cholecystitis, parasites, reflux cholangitis), vascular abnormality is the cause in 7% (most commonly pseudoaneurysm of the hepatic artery), and pancreatic pseudocyst causes hemobilia in 1%. In almost all cases, the bleeding originates from intrahepatic or extrahepatic bile ducts, and rarely from the pancreas.

PUBMED search with keywords “hemobilia” and “arterio biliary fistula” found a total of 44 papers. No case with intrapancreatic arterio biliary fistula was found. To the best of our knowledge, we present a unique case of delayed life-threatening hemobilia caused by intrapancreatic arterio-biliary fistula. It was diagnosed at the fourth admission and managed successfully by emergency Traverso-Longmire pancreatoduodenal resection. We briefly discuss the keys to a timely diagnosis and the cornerstones of the treatment.

The timely diagnosis of hemobilia depends on a high index of suspicion and careful interpretation of the symptoms. Hemodynamic stability has a crucial role in the decision-making process. Angioembolization is the cornerstone of the treatment, whereas surgery is reserved only for cases with an unstable hemodynamic or unsuccessful embolization. Surgical approach depends on the bleeding site. Although an emergency pancreatic head resection is a procedure of last resort, it can be life-saving in cases with intractable bleeding due to intrapancreatic arterio biliary fistula.

Keywords

delayed manifestation, emergency Traverso-Longmire procedure, hemobilia, intrapancreatic arterio-biliary fistula

INTRODUCTION

Hemobilia is a rare cause of upper gastrointestinal bleeding. It has often intermittent manifestation which could lead to a significant diagnostic and therapeutic challenge. In contrast to the past, nowadays, the trauma accounts for only 6% of all cases, whereas 65% are iatrogenic causes.^[1] Rarely, hemobilia is a result of malignancy (7%), gallstones (5%), inflammation (8%, cholecystitis, parasites, reflux cholangitis), vascular abnormalities (7%, most commonly pseudoaneurysm of the hepatic artery, extremely rare of the cystic artery), and pancreatic pseudocyst in 1%.^[1] In almost all cases, bleeding originates from the intrahepatic or extrahepatic bile ducts, and rarely from the pancreas.^[2,3] The PUBMED search with keywords “hemobilia” and “arteriobiliary fistula” yielded a total of 44 papers. No case reporting two distinct episodes of hemobilia or intra-pancreatic arterio-biliary fistula was found.

Herein we report a unique case of two distinct episodes of iatrogenic hemobilia, the first due to an iatrogenic lesion during laparoscopic cholecystectomy. The second life-threatening episode was caused by intrapancreatic arterio-biliary fistula diagnosed during the fourth readmission, which was managed successfully by emergency Traverso-Longmire pancreatoduodenal resection.

CASE REPORT

A 75-year-old man was admitted to the clinic for uncomplicated chronic calculous cholecystitis. He underwent an elective laparoscopic cholecystectomy and was discharged on the next day. The patient did not report receiving anticoagulant therapy. Two weeks later, he was readmitted due to a fever up to 38°C and right upper abdominal pain. The abdominal examination revealed a distended abdomen with painful

right upper quadrant without rebound tenderness. The blood test was normal except for Hb 9.9 g/dL (16 g/dL at the first admission) and CRP 54 mg/dL. The contrast CT showed hematoma in the gallbladder fossa and dilated common bile duct filled with high-density content (Figs 1, 2). ERCP and stenting were not attempted because of the unstable condition and the signs of acute abdomen so the emergency laparotomy was undertaken. Intraoperatively, a large number of blood clots around the liver and in the gallbladder fossa was found. The common bile duct was dilated with a 3×5-mm tangential lesion. It was filled with blood clots which were removed and the duct was lavaged with saline. Because there was no active bleeding, the intervention was completed by Kehr drain. Fourteen days later, the patient was reoperated due to dislocation of the Kehr with subhepatic and subdiaphragmatic abscesses. Because of severe fibrotic changes at porta hepatis and hepatoduodenal ligament no additional intervention was undertaken. On the next day, the patient underwent stent placement, which was removed one month later.

The third admission was ten months later because of melena and slight upper abdominal pain. The physical examination was unremarkable. The blood test was normal except for Hb 7.8 g/dL, total and direct bilirubin 31.4 and 20.6 mmol/l, respectively, ASAT 282 U/l, and ALAT 190 U/l. The upper endoscopy could not find a source of bleeding, MRCP revealed dilated common bile and left hepatic duct, a cystic transformation of the accessory pancreatic duct, but otherwise normal biliary tree without evidence of hemobilia or obstruction (Figs 3, 4). The capsule endoscopy could not find a bleeding source. The patient received blood transfusions and was discharged without signs of bleeding. Two weeks later a new episode of massive bleeding occurred. The endoscopy showed a clot at the major duodenal papilla. The angiography revealed bleeding at the communication of superior and inferior pancreatoduodenal arteries. Because of unstable he-

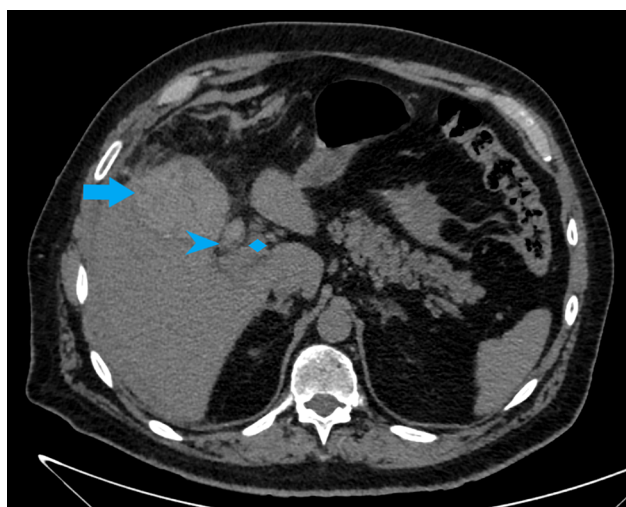


Figure 1. The abdominal CT on the second admission – hematoma in the gallbladder fossa (arrow), dilated common bile duct filled with blood (arrowhead) and portal vein (rhombus).

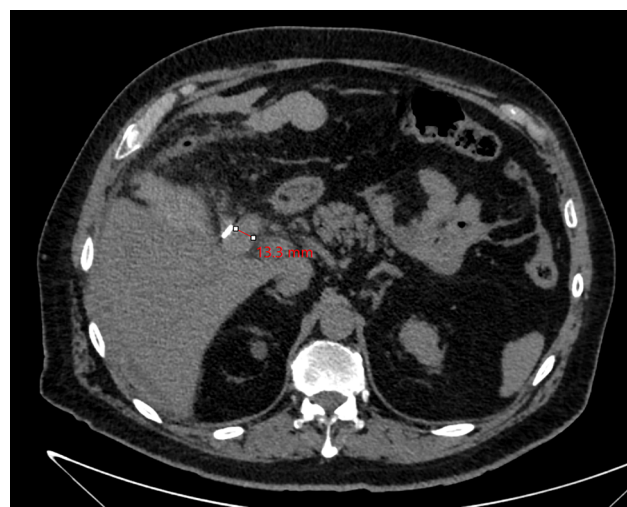


Figure 2. Common bile duct dilated to 13.3 mm with clips from the previous intervention.



Figure 3. MRCP at the third admission, ten months after the index operation – dilated common bile duct and left hepatic duct, a cystic transformation of the accessory pancreatic duct. No evidence of hemobilia or obstruction.

modynamics, the patient was taken to the operation room.

The common bile duct was filled with clots, which were evacuated. The cholangioscopy found bleeding arterial vessel at the intra-pancreatic part of the common bile duct so Traverso-Longmire pancreatoduodenal resection was performed. The patient had a complicated postoperative course with several reinterventions because of leaks from the biliary and pancreatic anastomosis. He was discharged with low debit pancreatic fistula after 41 days of hospital stay. One year after the intervention he is free of bleeding. The macroscopic examination of the specimen showed diffuse mucosal ulcerations of the distal common bile duct and thickened surrounding tissues with haemorrhages. The histological examination revealed a finding compatible with intrapancreatic arteriovenous fistula – diffuse ulcerations of the distal common bile duct with zones of fresh bleeding, haemorrhage in the surrounding pancreatic tissue and arterial vessels with a thickened wall filled with hyaline thrombi (Figs 5-7).

DISCUSSION

Francis Glisson described bleeding in the biliary tract in 1654, but the term “hemobilia” was introduced in 1948 by Sandblom. The iatrogenic hemobilia most commonly results from arteriovenous fistula after an inadvertent puncture of the vessels during a variety of mini-invasive procedures, cholecystectomy or other procedures.^[1] In the review by Green et al., 13% of the cases occurred after cholecystectomy.^[1] In the present case, there was an iatrogenic lesion of the common bile duct so the first episode of hemobilia was attributed to an injury of the 3 and 9 o'clock arteries, so no additional diagnostic work-up was performed. This assumption was supported by the absence of pseudoaneurysm on the angiography performed at later stage.

The subsequent bleeding episodes and the intrapancreatic arteriovenous fistula can also be an iatrogenic lesion during the two ERCP procedures for placement and removal of the stent. We could not find papers describing

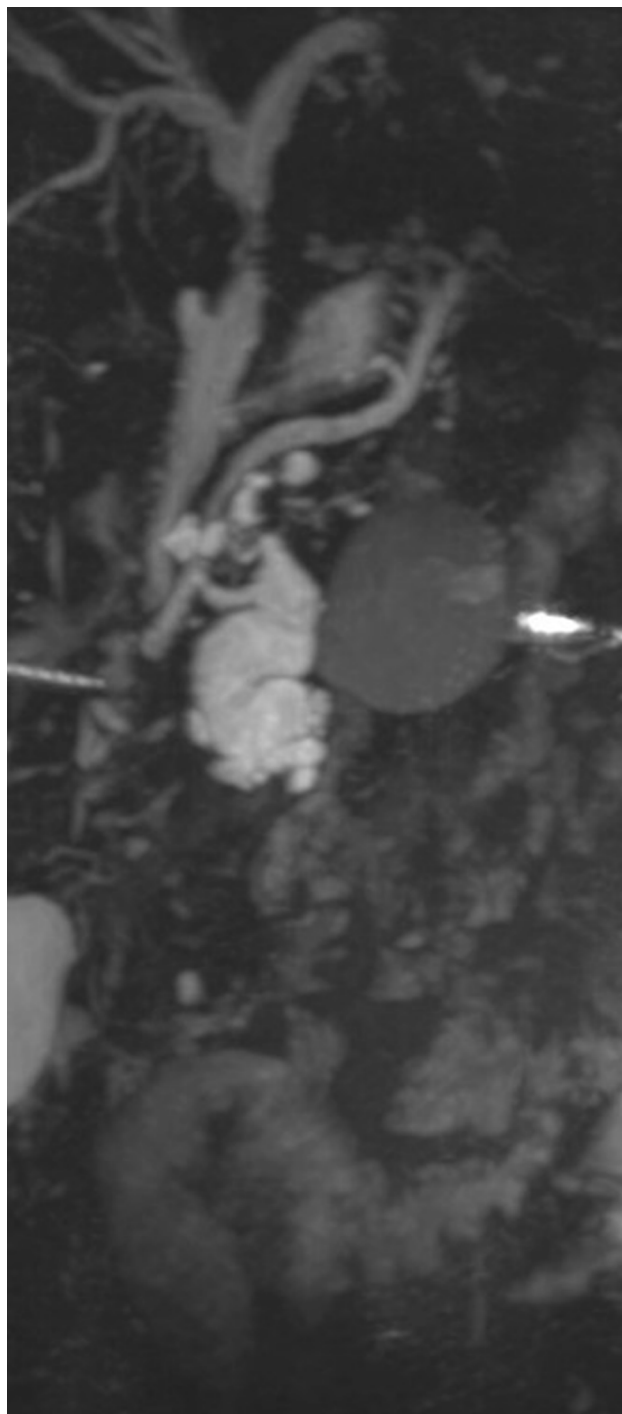
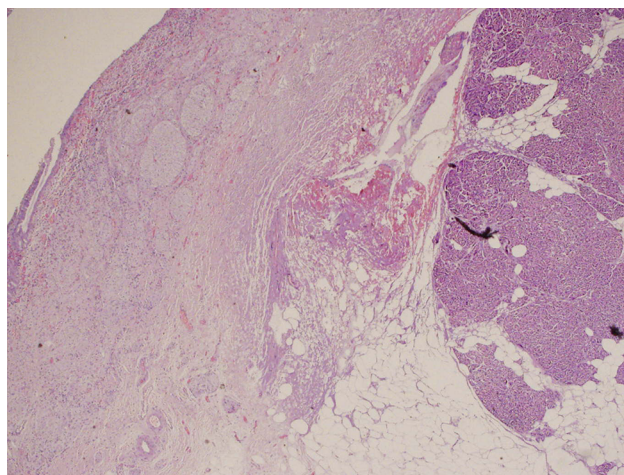
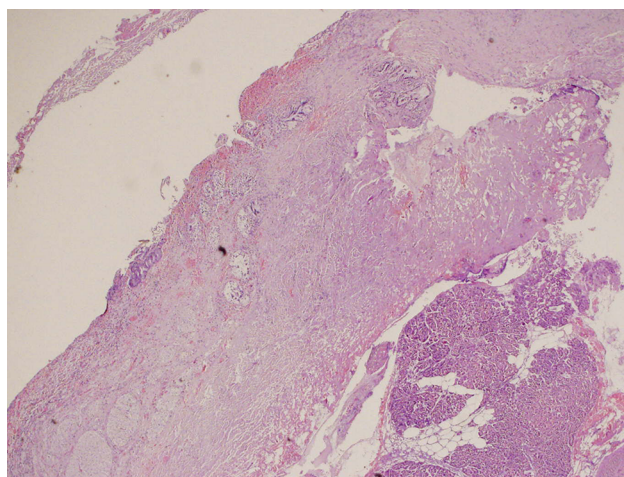
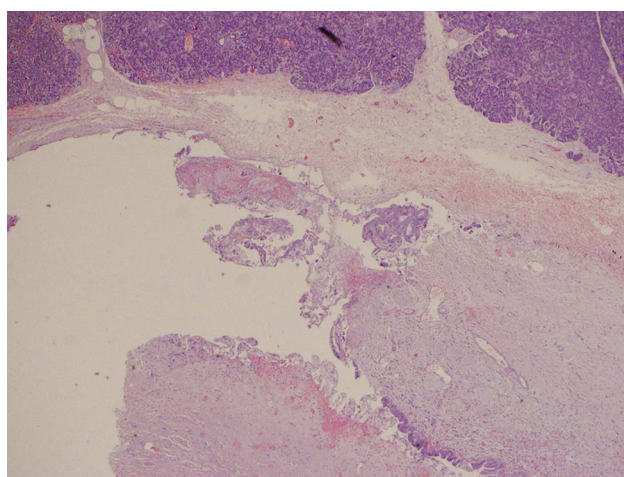


Figure 4. MRCP at the third admission, ten months after the index operation – a cystic transformation of the accessory pancreatic duct.

intrapaneatic fistula.

Quincke's classic triad consists of gastrointestinal bleeding, upper quadrant pain, and jaundice, but it occurs in only 22% of the cases. The onset can be acute (immediately after procedures or trauma) or delayed.^[1] The most common manifestation is upper gastrointestinal bleeding. In the vast majority of cases, the bleeding is self-limiting and does not compromise the hemodynamics. It is frequently intermit-

**Figure 5.****Figure 6.****Figure 7.**

Figures 5-7. Microscopic view of the intrapancreatic common bile duct – diffuse ulcerations of the distal common bile duct with zones of fresh bleeding, confluent haemorrhage in the surrounding pancreatic tissue and arterial vessels with a thickened wall filled with hyaline thrombi (Hematoxylin-eosin, 4×).

tent and leads to a significant diagnostic delay, especially in absence of trauma or iatrogenic causes as in our case. In contrast, after percutaneous transhepatic procedures, the bloody output from the drain significantly facilitates the prompt diagnosis. Obstructive jaundice is a rare manifestation. In our case, only discrete elevation of bilirubin and transaminases at the fourth admission was observed. Very rarely, hemobilia manifests with acute pancreatitis.^[4]

Hemodynamic stability has a key role in the decision-making process. Upper endoscopy is usually the first step but it is diagnostic only in 12% of the cases.^[1] The next step in hemodynamically stable patients is CT or CT-angiography, which may reveal high-density content in the gallbladder and biliary tract and could assess the liver parenchyma and pancreas. In absence of bleeding, conservative treatment with blood transfusions is indicated. The active bleeding or failed conservative therapy require angiography, which is a mandatory first step in unstable hemodynamics.^[5] The risk of failure in case of intrapancreatic arterio-biliary fistula, however, is high because of the rich collateral blood supply of the pancreatic head. An extensive series reported conservative treatment in 43% (“correction of coagulopathy and adequate drainage”), while 36% of the patients were managed by angioembolisation, 12% by cholecystectomy, and 8% by other surgery.^[1]

Surgery is reserved only for cases with massive hemobilia and unstable hemodynamics or failed angioembolisation. The type of operation depends on the site of bleeding, so serious efforts for accurate preoperative localization are mandatory. Methods of choice are excision of the aneurysm (with or without reconstruction) or ligation of the hepatic artery, cholecystectomy, liver resection, Kehr drainage of the biliary obstruction, or Roux-en-Y hepaticojejunostomy.^[6,7] The pancreatic head resection is a procedure of last resort due to the extremely high morbidity and mortality. Nevertheless, it could be life-saving as in our case.^[8-11]

CONCLUSIONS

Timely diagnosis of hemobilia depends on the high index of suspicion, detailed medical history and interpretation of the symptoms. Hemodynamic stability has a crucial role in the decision-making process. Nowadays, the angioembolization has a key role, while the surgery is reserved only for the cases with an unstable hemodynamics or failed embolization. The surgical tactic depends on the site of bleeding, so all efforts for accurate preoperative localization are mandatory. Methods of choice are liver resection, cholecystectomy, ligation of the hepatic artery, or its branches, and Roux-en-Y hepaticojejunostomy. The emergency pancreatic head resection is a procedure of last resort but can be life-saving in cases with intractable bleeding due to intrapancreatic arterio-biliary fistula.

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Два отдельных случая опасной для жизни гемобилии из-за поражения общего жёлчного протока и отсроченной интрапанкреатической артерио-билиарной фистулы, купированные экстренной панкреатодуоденальной резекцией

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Резюме

Гемобилия является чрезвычайно редкой причиной кровотечения из верхних отделов желудочно-кишечного тракта. Она часто имеет интермиттирующие проявления, что может привести к значительной задержке диагностики. В 65 % случаев причины ятрогенные, в 7% – злокачественные, в 5% – желчнокаменные, в 8% – воспалительные (холециститы, паразиты, рефлюкс-холангит), в 7% – сосудистые аномалии (чаще всего псевдоаневризма печёночной артерии), а псевдокиста поджелудочной железы вызывает гемобилию в 1%. Почти во всех случаях кровотечение происходит из внутripечёночных или внепечёночных жёлчных протоков, редко из поджелудочной железы.

Поиск в PUBMED по ключевым словам «гемобилия» и «артериобилиарная фистула» нашёл в общей сложности 44 статьи. Случаев интрапанкреатической артериобилиарной фистулы не выявлено. Насколько нам известно, мы представляем уникальный случай отсроченной опасной для жизни гемобилии, вызванной внутripанкреатической артерио-билиарной фистулой. Он был диагностирован при четвёртом поступлении и успешно лечился экстренной панкреатодуоденальной резекцией Traverso-Longmire. Мы кратко обсудим ключи к своевременной диагностике и краеугольным камням лечения.

Своевременная диагностика гемобилии зависит от высокой степени подозрительности и тщательной интерпретации симптомов. Стабильность гемодинамики играет решающую роль в процессе принятия решений. Ангиоэмболизация является краеугольным камнем лечения, в то время как хирургическое вмешательство применяется только в случаях нестабильной гемодинамики или неудачной эмболизации. Хирургический доступ зависит от места кровотечения. Хотя экстренная резекция головки поджелудочной железы является крайней мерой, она может спасти жизнь в случаях некупируемого кровотечения из-за интрапанкреатической артериобилиарной фистулы.

Ключевые слова

Отсроченное проявление, экстренная операция Traverso-Longmire, гемобилия, интрапанкреатическая артерио-билиарная фистула