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Hemobilia Due to Cystic Artery Pseudoaneurysm: A Rare Late Complication of Laparoscopic Cholecystectomy

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ABSTRACT
We discuss a patient with late presentation of hemobilia following cholecystectomy, which is unusual because pseudoaneurysm caused by vascular injury during surgery typically presents soon after surgery. Endoscopic retrograde cholangiopancreatography revealed a large blood clot arising from the biliary orifice with subsequent computed tomography angiography diagnosing a large pseudoaneurysm in the region of the cystic artery adjacent to the cholecystectomy clips. Embolization was performed via direct percutaneous puncture of the pseudoaneurysm.

INTRODUCTION
Hemobilia as a consequence of laparoscopic cholecystectomy is a rare and potentially life-threatening condition. It is a diagnostic challenge due to its rarity and various clinical presentations. Therefore, the clinician must have a high index of suspicion to quickly identify and treat the complication. A laparoscopic approach to cholecystectomy provides a safe and efficient method for gallbladder removal, but compared to open abdominal surgery, the incidence of biliary and vascular injuries are higher. These vascular injuries may result in pseudoaneurysm formation with a potential to hemorrhage. Bleeding typically presents soon after surgery; however, the onset of hemorrhage may be delayed several weeks in rare cases.

CASE REPORT
A 79-year-old man presented to the hospital with acute onset epigastric pain, nausea, and jaundice. His medical history was significant for hypertension, prostate cancer, and peptic ulcer disease. Surgical history was significant for laparoscopic repair of gastrointestinal (GI) bleeding thought to originate from gastric ulcers or a Mallory-Weiss tear complicated by an incisional hernia. He also had a history of cholelithiasis with cholecystitis and had undergone a laparoscopic cholecystectomy (LC) 15 months earlier. The surgery was prolonged due to adhesiolysis and concurrent repair of incisional ventral hernia and urethral stricture dilation. It was completed within 3 hours. The cystic duct was reported to be long and narrow and was doubly clipped and cut along with the cystic artery. No complications were reported.

Review of systems on admission was significant for no fever, vomiting, or weight loss. The patient had no changes in bowel habits or rectal bleeding. He denied using aspirin, non-steroidal anti-inflammatory drugs, or anti-coagulants. On physical examination, his vital signs were stable, sclerae were icteric, and he had mild epigastric pain without signs of peritonitis. Laboratory evaluation revealed leukocytosis (white blood cell count, 16,000/mm³) and normal hemoglobin (13.8 gm/dL) on admission that trended down (10.7 gm/dL) in a few days. Liver chemistries were elevated (direct bilirubin 5.3 mg/dL, alkaline phosphatase 337 IU/L, aspartate aminotransferase 379 IU/L, and
Alanine aminotransferase 798 IU/L. Acute viral hepatitis serologies were negative. Abdominal magnetic resonance imaging revealed moderate biliary ductal dilation and hyper-enhancing filling defects in the distal bile duct. Endoscopic retrograde cholangiopancreatography (ERCP) revealed a large blood clot emerging from the biliary orifice (Figure 1). Cholangiography revealed a dilated biliary tree with multiple distal filling defects. These findings were consistent with hemobilia. Two stents were placed for biliary decompression: one 10-Fr x 10-cm plastic stent with internal/external pigtail and one 10-Fr x 5-cm plastic stent with single internal/external flap. Bile flow was noted after stent placement. Sphincterotomy was not performed. A computed tomography (CT) angiogram revealed a 13 x 6-mm pseudoaneurysm arising in the region of the cystic artery adjacent to the cholecystectomy clips (Figure 2). This was confirmed on visceral angiography (Figure 3): however, the artery feeding the pseudoaneurysm could not be identified for embolization. Embolization was performed via direct percutaneous puncture of the pseudoaneurysm. The patient recovered uneventfully with resolution of his symptoms. He was discharged the day after his embolization. Follow-up CT 2 weeks later revealed no residual filling of the pseudoaneurysm, and ERCP 3 months later revealed a normal cholangiogram. The stents were subsequently removed.

DISCUSSION

Vascular injury is an uncommon complication of LC, occurring in 0.25–0.7% of patients. Pseudoaneurysm of the hepatic or cystic artery is a rare manifestation of LC-related vascular injury, with fewer than 100 cases in the literature. The most common presentation of pseudoaneurysm rupture into the biliary tree is GI hemorrhage (GIH). However, the classic Quincke’s triad presentation of hemobilia (obstructive jaundice, abdominal pain, and GIH) is seen in fewer than 50% of cases. The average time between LC and onset of bleeding is 13–21 days, with a range between 5 and 120 days reported in the literature. Our patient presented with a significantly delayed hemorrhage 15 months after his surgery.

Pseudoaneurysm formation results from a direct vessel wall injury resulting in a periarterial hematoma. This direct injury is typically iatrogenic and created during a surgical, endoscopic, or vascular interventional procedure. In the setting of LC, the most common etiologies include direct vascular injury during resection of the cystic duct or initial clip placement, cholecystectomy clip erosion, or thermal injury. During
laparoscopic surgery, normal variations in the anatomic location or course of the right hepatic artery can lead to misidentification as the cystic artery and inadvertent damage.9,12 The exact pathogenesis of pseudoaneurysms is unknown, but the toxicity of bile acids from associated leaks and secondary infections are thought to contribute to weakening suture lines and surgical clips used for hemostasis during surgery.3,11

The initial evaluation of patients with hemobilia generally begins with GI endoscopy or ERCP to exclude more common causes of GIH and relieve the biliary obstruction. Subsequent angiography (CT or conventional) is required to identify the source of bleeding. Transarterial or percutaneous embolization is the treatment of choice for iatrogenic pseudoaneurysms of the hepatic artery and its branches. Surgery is an option of last resort due to high morbidity and mortality associated with operative intervention in this setting.1,5,9

There needs to be a high index of suspicion for patients with prior gallbladder and biliary surgeries presenting with GIH. These patients should have a prompt upper endoscopy performed with close evaluation of the papilla for signs of hemobilia, and follow-up cross-sectional imaging is called for if a pseudoaneurysm is suspected.

DISCLOSURES

Author contributions: R. Badillo, D. Darcy, and VM Kushnir wrote and revised the manuscript. R. Badillo is the author guarantor.

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Informed consent was obtained for this case report.

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