Non-traumatic Biliary Perforation: Lessons for Diagnosing a Rare but Deadly Condition

Matthew S. Lee, MS[1] and Kevin M. Lowe, MD, PhD, FACS[1, 2]
[1]Carle Illinois College of Medicine [2]Department of General Surgery, Carle Foundation Hospital

Introduction

Non-traumatic perforation of the biliary tree is exceedingly uncommon. While it is occasionally seen in infants in association with congenital malformations, it is rarer in adults[1]. The pathogenesis remains unclear, but most cases are associated with choledocholithiasis or another obstructive biliary pathology[1,2]. Thus, increased biliary pressure, stasis, and dilatation may play a role.

Due to its rarity, the index of suspicion for this condition may be low and diagnosis difficult. Nonetheless, mortality is high in the absence of timely management. Here, we report a case of spontaneous hepatic duct perforation in an elderly patient presenting with acute abdomen secondary to gallstone pancreatitis.

Case Presentation

HPI: A 78-year-old female smoker with no pertinent medical history presented with a one-day history of nausea, vomiting, and sudden onset abdominal pain.

Physical Exam: afebrile, notable for hypotension, tachycardia, ascites, abdominal distension, and diffuse abdominal tenderness

Labs: AST 550 U/L; ALT 570 U/L; ALKP 240 U/L; T-bili 3.8 mg/dL; lipase 3645 U/L

Hospital Course:

- Abdominal CT and RUQ ultrasound **(Figure 1)** showed cholelithiasis and normal CBD diameter
- KUB and repeat abdominal CT showed free air **(Figure 2),** so she was taken for emergent exploratory laparotomy
- No bowel or biliary injury was found; drains were placed and the patient was admitted to the ICU
- Despite initial improvement, on POD 3 the patient developed signs of sepsis and grossly bilious drainage was noted from the right JP drain
- ERCP was attempted on POD 5, but CBD cannulation was not possible
- Repeat ex-lap showed significant bowel necrosis and a contained bile leak in the RUQ
- Further exploration revealed a perforation at the confluence of the common hepatic and right hepatic ducts **(Figure 3)**
- Cholecystectomy was performed, and a T-tube was placed through the biliary perforation
- Multisystem organ failure occurred; the power of attorney chose to withdraw care, and the patient expired

Key Takeaways: Clinicians should be aware of possible bile duct perforation in patients with biliary obstruction. Liver hilum exploration should be a key part of laparotomy for abdominal sepsis of unknown source.

Images



Figure 1: Right upper quadrant ultrasound showing stones in the gallbladder, as well as trace gallbladder wall thickening to 5mm and pericholecystic fluid.

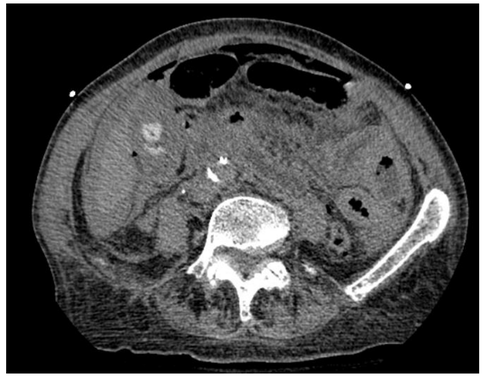


Figure 2: Non-contrast CT with free abdominal air, stones in the gallbladder, and inflammatory changes around the gallbladder and pancreas.

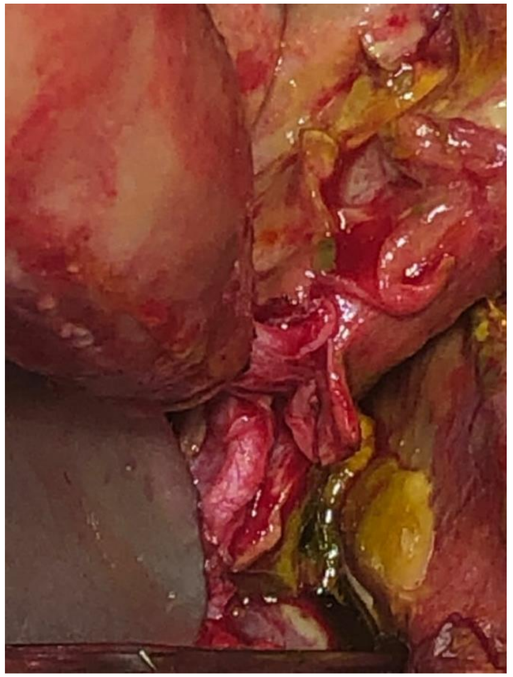


Figure 3: Intra-operative photograph demonstrating a defect at the confluence of the common hepatic and right hepatic ducts.

Discussion

Spontaneous bile duct perforation is rare, but dangerous.

- Kang et. al. found 70 reported cases in the literature in 2004[2]; our own Scopus search found 34 additional cases since then. Mortality is reported to be >20%[3,4].

The pathogenesis is unclear, but biliary obstruction is thought to play a key role.

- The most widely accepted theory is that biliary obstruction increases intraductal pressure, leading to biliary dilatation, stasis, and ischemia, ultimately resulting in necrosis and duct rupture[2]. In one review, biliary obstruction was due to gallstones in 20/26 cases (77%)[3].

Thorough exploration of the liver hilum should be performed during ex-lap in cases of abdominal sepsis of unknown source.

- Initial ex-lap in our patient did not reveal any biliary defects. The perforation may have developed later, or it may have been missed due to its location high at the biliary confluence.
- Another case of biliary injury missed on initial ex-lap also noted that the liver hilum was not explored[5].

The mainstay of treatment is biliary drainage with T-tube placement; surgical repair is usually infeasible and unnecessary.

- Consistent with the literature[1,2,6], our patient's biliary perforation was too necrosed to repair.
- When other comorbidities are well-controlled, clinicians have reported good outcomes with T-tube placement alone[1,2,7,8]; the biliary defect often closes spontaneously[8,9].

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