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## Case report of non-traumatic spontaneous intrahepatic bile duct rupture in an adult



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## ABSTRACT

**INTRODUCTION:** Spontaneous rupture of the biliary duct, a rare condition in adults, is difficult to diagnose preoperatively and presents with acute abdominal symptoms. The treatment of this rare condition should be based on the individual's clinical status. We present peripheric biliary duct rupture (segment three) treated with external segment III drainage and postoperative endoscopic removal of the stones.

**PRESENTATION OF CASE:** An 82-year-old male patient presented with abdominal pain and fever. An ultrasound (US) revealed a solid gall stone lesion, 3 cm in diameter, in liver segments three and four with additional intra-abdominal fluid accumulation without coexisting free air. A diagnostic laparotomy was then performed because the patient had signs of peritonitis. Exploration revealed a biliary leakage from the posterior surface of segment three. An external biliary drainage catheter was inserted to the perforated segment III duct via a 6 French (6F) feeding catheter. He was discharged after 10 days and his intracholedocal stent was removed postoperative after three months. The patient continues to be monitored.

**DISCUSSION:** Spontaneous rupture of the intrahepatic biliary duct is a rare condition. Although occurrence is frequently reported as spontaneous, the majority of cases are related to choledocholithiasis. The role of surgical treatment in cases of spontaneous bile duct rupture is unclear. When biliary peritonitis is present, drainage of contaminated biliary fluid, T-tube drainage, closure of the biliary duct, as well as primary disease conditions, should be reviewed prior to treatment.

**CONCLUSION:** Surgical treatment of spontaneous biliary duct rupture should be indicated only after careful consideration of the patient's clinical and comorbidity status.

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## 1. Introduction

Spontaneous bile duct rupture is a rare condition in adults, which may have its origins in the extra or intra hepatic biliary ducts. Choledocal cysts and pancreatobiliary ductal system abnormalities in a newborn are responsible for the relatively higher incidence associated with children [1]. Extrahepatic biliary duct rupture may arise in the ductus choledochus or common biliary duct; intrahepatic biliary duct rupture is a relatively less common condition. Rupture may result from cholelithiasis, choledocholithiasis, hepatolithiasis, or tumor obstruction. Increases in pressure, caused by obstructions, erosion of gallstones, or necrosis of the biliary duct wall, may lead to rupture and localized biliary peritonitis [2]. Peri-

tonitis often masks this condition in patients and thus ductal issues are frequently underdiagnosed. We herein present a case of segment three biliary duct rupture and suggest that this condition should be assessed during evaluation of acute abdominal symptoms and before considering treatment options.

## 2. Presentation of case

An 82-year-old male patient presented with abdominal pain and fever. Upon physical examination, rebound tenderness, also known as Blumberg's sign, was present in all of the abdominal quadrants. The patient's body temperature was 38 °C, blood pressure was 80–40 mmHg, heart rate was 118 bpm, white blood count was 22,000/mL, total bilirubin level was 3.2 mg/dL, and direct bilirubin level was 2.4 mg/dL. Abdominal ultrasonography (USG) examination revealed fluid collection in all abdominal compartments. A chest X-ray did not reveal air accumulation below the diaphragm. Macroscopic fluid, with biliary content, was aspirated using USG-guided paracentesis, with a resulting bilirubin level of 28.9 mg/dL. Culture samples were collected from the fluid. An US also revealed

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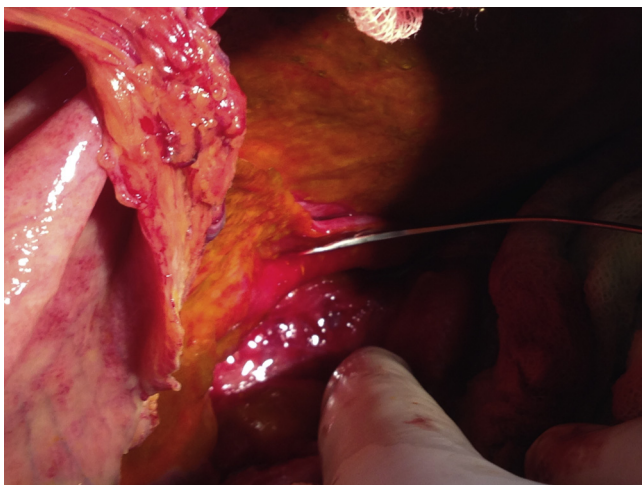


Fig. 1. Ruptured peripheric bile duct.

a solid gall stone lesion, 3 cm in diameter, in liver segments three and four and additional intra-abdominal fluid accumulation without coexisting free air. Diagnostic laparotomy was then performed. During abdominal exploration, approximately 2.000 ml. of purulent fluid, with bile content, was discovered in all of the abdominal quadrants, but principally in the perihepatic region and Morrison's pouch. The abdominal cavity was irrigated and aspirated. Upon visual examination, the gall bladder and extrahepatic biliary structures were of normal condition. A dilated biliary structure, 3–4 cm in diameter, was observed in the left lobe of the liver. Further exploration revealed a biliary leakage from the posterior surface of segment three, adjacent to the minor curvature of the stomach, which became more apparent following debridement of the surrounding tissues and irrigation. Intrahepatic bile ducts were checked via an examination probe through the ruptured orifice of segment III bile duct (Fig. 1). Intrahepatic stones were not removed during surgery. The patient's advanced age (82 year-old), the existence of severe abdominal biliary peritonitis, and the possibility of a biliary obstruction at the level of the left major branch prompted us to perform an external biliary drainage of the perforated segment III duct by a 6F feeding catheter. Three abdominal drainage tubes were placed. During the postoperative course, abdominal serous fluid was observed when the bile drainage volume increased to approximately 100 mL/day. The severity of fever and other clinical symptoms related to systemic inflammation decreased. The preoperative abdominal fluid sample culture was positive for *Escherichia coli*. On postoperative day 10, abdominal drainage fluid changed to biliary in content. Cholangiography revealed that the feeding catheter had been dislodged from the segment III bile duct. ERCP provided evidence of a peripheral biliary leak from the left lobe of segment three (Fig. 2). Following debridement of gallstones inside the left major bile duct, a nasobiliary stent was inserted. During follow-up, the biliary content of the abdominal drainage fluid disappeared. Control nasobiliary-grap did not reveal any leakage. A 10F plastic stent was placed in the ductus choledochus so that it extended into the left major bile duct. The nasobiliary and abdominal drainage tubes were removed. He was discharged after 10 days and the intracholedocal stent was postoperatively removed after three months. The patient continues to be monitored.

### 3. Discussion

Spontaneous rupture of the intrahepatic biliary duct is a rare condition. Although occurrence is frequently reported as spontaneous, the majority of cases are related to choledocholithiasis [3].

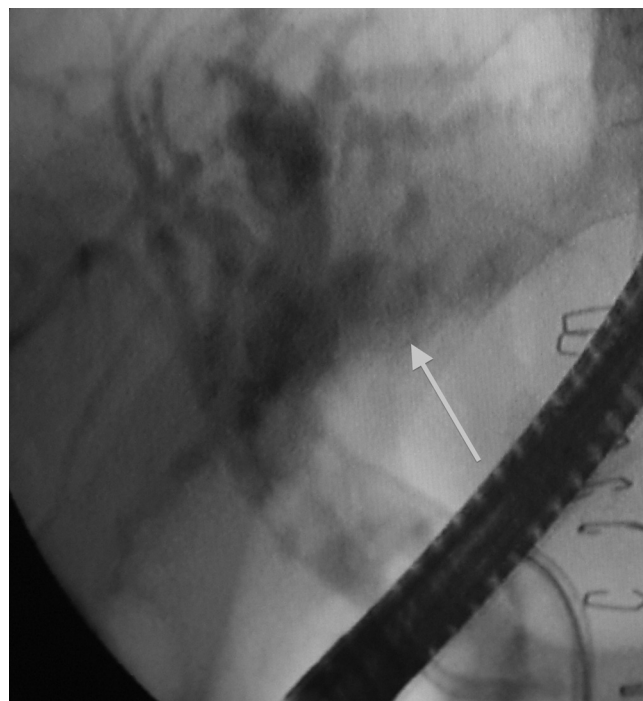


Fig. 2. ERCP provided evidence of a peripheral biliary leak.

A literature search for intrahepatic bile duct ruptures of the left hepatic lobe yield more results since the right hepatic duct might be less likely to rupture. Superficially located left lobe bile ducts are particularly prone to rupture due to increased intra-canalicular pressure.

Preoperative diagnosis of spontaneous biliary duct ruptures, which present with acute abdominal pain due to localized or generalized biliary peritonitis, is problematic [4]. Abdominal US and computed tomography scans could aid in the diagnosis of primary lesions or identify intra-abdominal fluid collection [5]. Biliary leakage can be detected through nuclear medicine studies or via intraoperative cholangiography [6].

As delineated previously, preoperative diagnosis of spontaneous bile duct rupture is challenging, despite advances in diagnostic procedures [7,8]. Biliary fluid collection might indicate a duodenal or gastric rupture, as in the present case. Failure to localize any such rupture might necessitate gall bladder and extrahepatic bile duct exploration. If no rupture is observed, the anterior and posterior surfaces of the left hepatic lobe should be inspected for biliary leakage. When leakage is not visualized macroscopically, intraoperative cholangiography following cholecystectomy is advised. However, the patient would not benefit from cholangiography if a gall bladder stone or tumor were among the aetiologic factors.

The role of surgical treatment in patients with spontaneous bile duct rupture is unclear. When biliary peritonitis is present, drainage of contaminated biliary fluid, T-tube drainage, closure of the biliary duct, as well as primary disease conditions, should be reviewed prior to treatment. Major surgery in patients with generalized biliary disease might increase mortality and morbidity.

### 4. Conclusion

Surgical treatment of a spontaneous biliary duct rupture should be indicated only following careful consideration of the patient's clinical state and comorbidity status.

**Conflict of interest**

There is no conflict of interest.

**Ethical approval**

None.

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None.

**Author contribution**

Fatih Sumer performed the operation and drafted the manuscript. Servet Karagul, Asim Onur, Ismail Ertugrul, Mehmet Ali Yağci participated in the care of the patient. Cuneyt Kayaalp was involved in revising it critically for important intellectual content.

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