

SPONTANEOUS EXTRA-HEPATIC BILE DUCT PERFORATION POSTPARTUM

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SUMMARY

Spontaneous bile duct perforation is an unusual cause of acute abdomen. It is an extremely rare condition and rarely suspected or correctly diagnosed pre-operatively. A case of a 29-year-old adult female, presenting with peritonitis, 2 days post partum is presented. Exploratory laparotomy showed biliary peritonitis secondary to a perforated common bile duct. She had a cholecystectomy and closure of the perforation over a T-tube. She recovered well and was discharged home. Awareness of spontaneous common bile duct perforation as a rare cause of biliary peritonitis, avoids undue delay in the diagnosis and thus improve prognosis. Cholecystectomy and drainage of bile duct using a T-tube is emphasized.

Keywords: Pregnancy, Pre-eclampsia, Peritonitis, Bile duct perforation, Cholangiogram

INTRODUCTION

Spontaneous non-traumatic perforation of the extrahepatic or intrahepatic bile ducts was first described in 1882 by Freeland at autopsy.^{1,2} Spontaneous bile duct perforation is an unusual cause of acute abdomen and is rarely diagnosed preoperatively and is, even more so, an extremely rare entity in pregnancy.³ It is commoner in the extremes of life, children and the aged, older than 60 years.^{4,5} We report a successfully managed case of spontaneous extrahepatic bile duct perforation in a postpartum woman at the Korle Bu Teaching Hospital (KBTH).

CASE REPORT

A 29-year-old woman was referred to the surgical team two days after delivery with complaints of epigastric pain of four days duration. The pain had initially been localized in the epigastrium, radiating to the right hypochondrium and somewhat relieved by leaning forwards. It later became generalized. The abdomen became distended and compromised breathing. Two litres of bile stained fluid was, therefore, tapped from the abdomen by the obstetrician and referred with a diagnosis of peritonitis from a perforated peptic ulcer disease.

The patient had developed imminent eclampsia at 38 weeks gestation and was managed on admission with oral nifedipine and intramuscular magnesium sulphate (MgSO₄). She had also been diagnosed with hepatitis B viral infection during the index pregnancy.

Examination revealed an acutely ill woman with a tinge of jaundice and bilateral pedal edema. She was afebrile (T° 37°C) and not pale (Hb 11.2g/dl). Her pulse was 100bpm with a BP of 130/100mmHg. She was dyspnoeic (44 cpm) with reduced air entry over the right lung field, but there were no crepitations. The abdomen was distended and generally tender, more marked in the epigastrium with guarding. There was, however, no rebound tenderness. The bowel sounds were present but infrequent and rectum was empty.

The white cell count was 15.85 X 10⁹/L with 85% neutrophilia. Her blood urea and electrolytes were normal. She had raised serum total bilirubin-22.0umol/L, unconjugated bilirubin-15.3umol/L, AST- 64U/L, ALT-76U/L, GGT-87U/L, alkaline phosphatase- 198U/L; normal total protein- 52g/L, low serum albumin-23g/L and a normal serum uric acid-341g/L.

She was presumptively diagnosed with peritonitis secondary to a perforated peptic ulcer disease, even though a chest x-ray did not reveal gas under the diaphragm. She was adequately resuscitated and prepared for exploratory laparotomy with intravenous fluids, potassium replacement, antibiotics and a proton pump inhibitor (Esomeprazole).

The findings at laparotomy were 3.5L of bilious peritoneal fluid, normal non-perforated bowel, a thickened distended gall bladder and two necrotic areas on the bile duct: one on the common hepatic duct, (Figure 1), and the other, on the left lateral aspect of the common bile duct, extending from the distal part of the supraduodenal portion to the proximal half of the retroduodenal portion, with perforation (Figure 2).

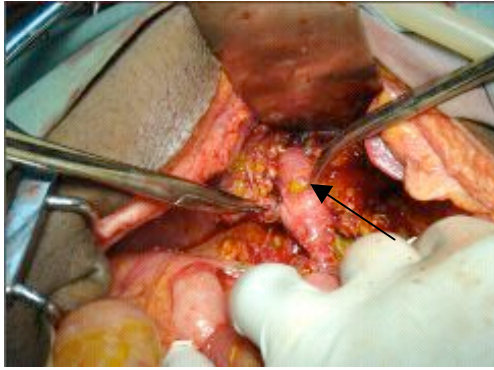


Figure 1 Image showing necrotic patch on the common hepatic duct (Arrowed).

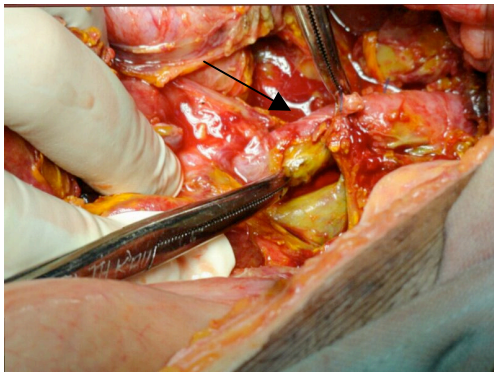


Figure 2 Image showing perforated site on the common bile duct where there was leakage, (Arrowed)

Cholecystectomy was performed and the common bile duct explored for stones. The necrotic area on the common hepatic duct was over sewn with vicryl 2/0, drawing healthy tissue to cover it. The perforation on the common bile duct was closed over a T-tube. An intra-operative cholangiogram could not be done.

The T-tube drained 450mls on the first postoperative day and the drainage subsequently reduced. Her haemogram, blood urea and electrolytes, and liver function tests improved dramatically and she was discharged on postoperative day 15 with the T-tube in situ. The T-tube drainage reduced by 50mls weekly after Week 2. A T-tube cholangiogram done at Week 4 showed leakage around the site of the T-tube insertion, (Figure 3). A repeat cholangiogram two weeks later showed no leakage, (Figure 4), and therefore, the T-tube was removed and the patient observed in hospital for 24 hours before being discharged home.

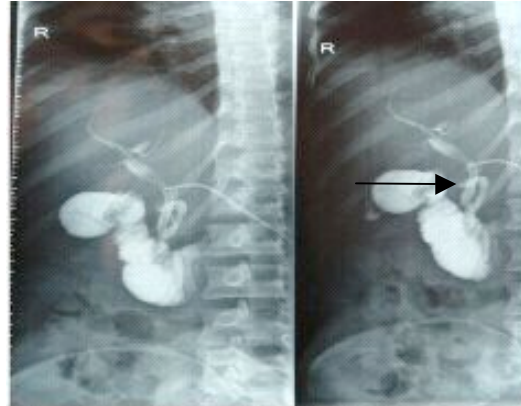


Figure 3 Postoperative cholangiogram at Week 3 showing leakage of contrast around T-tube. (Arrowed)



Figure 4 T-Tube cholangiogram 6 weeks post-operatively showing free flow of contrast into the duodenum with no leakage.

Histopathology of the gallbladder indicated acute cholecystitis with outpouching of the gallbladder mucosa. Its lamina propria had acute inflammatory cell infiltrates extending to the serosal surface.

DISCUSSION

Spontaneous perforation of the extrahepatic bile duct without a traumatic or iatrogenic injury has been described in children from congenital anomalies of the common bile duct including choledochal cysts, biliary atresia and diverticulae.³ Extrahepatic biliary rupture is an extremely rare condition in adults.⁶⁻⁹ It was first described in pregnancy by Piotrowski et al in 1990¹ and he related it to cholelithiasis.¹ The condition is associated with ductal stones in 70% of cases.¹⁰ In adults it is associated with acute pancreatitis, acalculous cholecystitis,¹⁰ HIV infections, Hodgkin's lymphoma¹⁰, tuberculosis¹¹ and severe necrotizing enterocolitis.¹² Extrahepatic biliary rupture could be also be due to cholecystitis or could be idiopathic in origin.¹³

The cause in idiopathic cases could be microcirculatory failure as a result of hypoperfusion and microthrombi formation in the end arteries supplying the biliary duct.¹⁴ Ischaemia and necrosis cause perforation.¹⁴ This patient had preeclampsia, a condition that is characterized by global arteriolar spasm and impaired microcirculation that could precipitate spontaneous necrosis and perforation. She also had acute acalculous cholecystitis probably from the preeclampsia which could equally be the predisposing factor for the necrosis.

Gallbladder disease could mimic mild preeclampsia, presenting with hypertension, epigastric pain or deranged liver function test,³ hence causing a diagnostic delay. However, our patient had signs suggestive of peritonitis, as well as a bile stained abdominal aspirate, warranting emergency surgery. Clinical presentation of extrahepatic bile duct perforation is varied: acute or insidious presentation. In insidious presentation there is painless abdominal distension, increasing jaundice and pale stools. An acute presentation may have symptoms and signs suggestive of infective peritonitis.

The optimal management of extrahepatic duct bile perforation is cholecystectomy and a choledochotomy with exploration of the common bile duct. An operative cholangiogram, which excludes distal bile duct obstruction, is an intelligent addition. The perforation is then closed over a T-tube. Primary closure of a choledochotomy has been suggested to be as safe as closure of the choledochotomy over a T-tube.^{15,16} Closure over a T-Tube in this case was a better choice since it afforded an opportunity to do a cholangiogram to rule out missed stones in the biliary tract and also diagnose any leak into the peritoneal cavity.

The T-tube is typically removed after three weeks¹⁷ postoperatively following T-tube cholangiogram demonstrating bile duct normalcy with no dye leakage. Leaving the T-tube in-situ for a longer period allows maturation of the temporary biliary cutaneous fistula and, reduces the risk of bile leakage into the peritoneal cavity.¹⁸ In this case, the T-tube was kept in situ for 6weeks to allow healing of the perforated site and it was safe to manage it on outpatient care.¹⁹

Delayed fibrosis and stricture of the bile duct could occur as a complication of the bile duct perforation. A Magnetic Resonance Cholangiopancreatography is employed to diagnose strictures when they occur and will be used in this patient during surveillance to diagnose any strictures formed.

CONCLUSION

Spontaneous perforation of bile duct is very rare, more so in pregnancy. It is difficult to diagnose preoperatively. Awareness of it as a cause of biliary peritonitis avoids undue delay in the diagnosis and management.

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