Acute Suppurative Necrotizing Pancholangitis
A Case Report

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Key Words
Biliary system · Endoscopic retrograde cholangiogram · Cholangitis · Common bile duct · [A1]

Abstract
Objective: To present a case of ascending cholangitis with resulting necrosis of the biliary system with perforation.
Clinical Presentation and Intervention: A 40-year-old male patient presented with upper abdominal pain, fever and jaundice assessed clinically and investigated by laboratory and radiological tests. Endoscopic retrograde cholangiogram and surgery were performed. However, because of extensive suppurative pancholangitis involving most of the intrahepatic radicles, sound surgical drainage could not be accomplished. Unfortunately, the patient died 2 days after surgery. Conclusion: In this case of severe cholangitis, endoscopic and surgical interventions were not successful and might have contributed to the worsening of the condition.

Introduction
Biliary tract infection remains a cause of morbidity and at times mortality, despite major advances in management such as the use of new antibiotics [1]. Cholangitis is an infection of the biliary duct that occurs especially when there is an obstruction and usually improves with the use of appropriate antibiotics and after definitive surgery [2]. Occasionally, surgeons face an unusual case that deteriorates despite all interventions and may rapidly lead to a fatal outcome. We present such a case of acute necrotizing pancholangitis involving perforation of the biliary system.

Case Report
A 40-year-old male patient was admitted to Jahra Hospital, Kuwait, with upper abdominal pain and vomiting, and a history of such recurrent attacks in the last 4 years. He had a low-grade fever, and physical examination showed tenderness and guarding of the epigastrium and right hypochondrium. Clinical diagnosis was acute cholecystitis. Laboratory investigations showed a total leukocyte count (TLC) of $10.3 \times 10^9/l$, serum amylase 696 U/l (normal 25–130), total bilirubin 28 $\mu$mol/l (normal 3–26), direct bilirubin 5 $\mu$mol/l (normal 0–4), and normal alkaline phosphatase and other liver enzymes.

Abdominal ultrasound revealed acalculous cholecystitis with normal common bile duct (CBD). Conservative treatment was started by nasogastric tube and intravenous fluids, and broad spectrum antibiotics (Clavofran, amikacin and Flagyl, which was later replaced with Tazocin). Two days later, the clinical condition remained the same, but the total bilirubin increased to 96 $\mu$mol/l, direct bilirubin to 72 $\mu$mol/l, alkaline phosphatase 140 IU/l (normal range 26–121), alanine aminotransferase 239 IU/l (normal range 10–60), aspartate aminotransferase 159 IU/l (normal range 10–42). Therefore endoscopic retrograde cholangiogram (ERC) was performed and showed slight dilatation of the CBD with biliary sludge, but no stones. Papillotomy was done to clear the sludge from the CBD. The patient’s tempera-
ture reached 38.5 °C, accompanied by rigors with a picture of cholangitis. TLC was 19.6 × 10^9/l, and total bilirubin, direct bilirubin and alkaline phosphatase were further elevated to 152 μmol/l, 98 μmol/l and 581 IU/l, respectively.

Since the patient did not show any substantial clinical improvement, a repeat ERC was done, and indicated severe suppurative cholangitis. There was irregular filling of the intrahepatic biliary system with haziness and gas bubble mixed with contrast. The papillotomy was extended and the CBD was cleared of thick sludge. Nasobiliary drainage tubing was removed because there was no obstruction.

The patient did not improve and developed a picture of septic shock, with a temperature of 38.7 °C and TLC of 23.2 × 10^9/l. Total bilirubin rose to 198 μmol/l, direct bilirubin to 133 μmol/l, and alkaline phosphatase to 640 IU/l. The patient was resuscitated and laparotomy was performed to drain the CBD. During the operation a collection of infected bile in relation to Calot’s triangle was found with disruption of the common hepatic duct (CHD) at its bifurcation. Both the CBD and CHD were necrotic with multiple perforations. There was no healthy duct up to or beyond the confluence of the hepatic duct. To get access to normal duct, both right and left hepatic ducts were dissected and followed into the liver parenchyma to the sectoral branches. Unfortunately the main bile duct and all dissected radicles were necrotic and sloughy. It was decided that no segment of bowel could be anastomosed to these necrotic ducts. Therefore the right and left hepatic ducts were cathe- terized and drained externally. The patient continued to deteriorate and died on the 3rd postoperative day.

Swabs from the bile showed Pseudomonas infection. A specimen from the CBD wall showed histological features of acute necrotizing and partly hemorrhagic cholangitis.

Discussion

The most common cause of acute cholangitis is cholecystolithiasis. Less common causes include malignant obstruction of the bile duct, strictures, biliary instrumentation, congenital abnormalities, parasitic infestation of the bile duct and acquired immune deficiency syndrome [3].

Bile is normally sterile in the absence of obstruction, but organisms can be cultured from more than 75% of patients who have bile duct stones [4]. The most common organisms are enteric in origin and include Escherichia coli, Klebsiella and Pseudomonas. Less common are gram-positive cocci and anaerobes [5]. The pathway by which bacteria gain access to the bile duct is uncertain. Translocation from the gastrointestinal tract is one proposed pathway especially in obstructive jaundice, leading to portal vein bacteremia [6]. In cases with obstructive jaundice portal endotoxemia may be related to the absence of bile acids in the gut lumen, which are normally responsible for binding endotoxins in the gut [7]. Another pathway is probably ascending Aeromonas infection from the gastrointestinal tract, which may follow retrograde instrumentation of the bile duct [8].

Bacterial cholangitis is linked to intermittent obstruction due to biliary sludge in the CBD; recurrent passage of biliary sludge may precipitate obstructive inflammation and even fibrosis of Vater’s ampulla and thus precipitate attacks of acute cholangitis [9]. Medical therapy with supportive measures and appropriate antibiotics are successful in 85% of patients with acute cholangitis [10]. For patients in whom initial management is not successful, biliary drainage is a life-saving procedure [11] and can be achieved either endoscopically, percutaneously or surgically. Endoscopic sphincterotomy was found to be associated with a complication rate of 28%. Surgical intervention, however, was associated with a complication rate of 58%. In addition, the mortality rate following endoscopic drainage is 5%, compared to 21% after surgical drainage [12]. Lai et al. [13] found a mortality rate of 10% in the endoscopic drainage group, compared to 32% in the operative group. Accordingly it is generally accepted that emergency operative decompression of the biliary tract should be reserved for patients who do not respond to adequate medical therapy or nonoperative drainage [14].

The case presented here is unique because of the magnitude of necrosis as well as the multiple perforations in the CBD and CHD, which involved all of the intrahepatic and extrahepatic biliary radicles with complete slough and gangrene. The multiple perforations in the biliary system might have contributed to the infection. It is also possible that in addition to Pseudomonas, there might have been other virulent organisms that failed to grow in culture. Medical and endoscopic therapy failed to improve the symptoms of cholangitis, and in fact it is probable that endoscopy contributed to the worsening of the condition due to excessive manipulation and possibly the introduction of resistant nosocomial organisms in the already infected and compromised biliary tree, thereby leading to death as previously reported [12, 13]. This case taught us a hard lesson not to push the patient for repeat ERC if there is no demonstrable obstruction from the first investigation.

Conclusion

The magnitude of the necrotizing pancholangitis that involved perforation of the biliary system was so great that neither endoscopic nor surgical decompression was successful. The timing of open surgical intervention in such a case of suppurative pancholangitis remains a difficult decision.
References