Bile Duct Injury during Cholecystectomy Requiring Delayed Liver Transplantation: A Case Report and Literature Review

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 ÖNCEL, D., ÖZDEN, İ, BİLGÉ, O., TÉKANT, Y., ACARLI, K., ALPER, A., EMRE, A. and ARIOGUL, O. Bile Duct Injury during Cholecystectomy Requiring Delayed Liver Transplantation: A Case Report and Literature Review. Tohoku J. Exp. Med., 2006, 209 (4), 355-359 —— Major bile duct injury during cholecystectomy represents potentially severe complications with unpredictable long-term results. If these lesions are not treated adequately, they can lead to hepatic failure or secondary biliary cirrhosis therefore requiring liver transplantation. We report a patient who required liver transplantation 15 years after open cholecystectomy. A 10-year old girl underwent open cholecystectomy and duodenal repair for cholelithiasis and cholecystoduodenal fistula. She required two surgical interventions, hepticojejunostomy which was performed in another center and portoenterostomy for biliary stricture at our institution seven years after the cholecystectomy. Eight years after the third operation, she required recurrent hospitalization for treatment of hepatic abscesses. The extremely short intervals between the three life threatening episodes and the rapid progression to severe sepsis were taken into consideration and liver transplantation was performed at the age of 25. She is leading a healthy life at 4 years post transplantation. Although iatrogenic biliary injury can usually be treated successfully by a combination of surgery, radiological and endoscopic techniques, patients with severe injuries develop irreversible liver disease. This case report and review of the literature suggest that liver transplantation is a treatment modality for a selected group of patients with end-stage liver disease secondary to bile duct injury. ——— bile duct injury; secondary biliary cirrhosis; liver transplantation

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Biliary and hepatic vascular injuries during cholecystectomy are complex lesions, associated with high rates of morbidity and mortality (Lillemoe et al. 1997; Buell et al. 2002). The incidence of major bile duct injury has remained constant at the rate of 0.1-0.3% during open surgery, and 0.3-1% during laparoscopic surgery. Although some centers report successful results in almost all patients with a combination of surgery and radiological techniques (Lillemoe et al. 1997;
Johnson et al. 2000), biliary cirrhosis cannot always be prevented (Nordin et al. 2001). An associated vascular injury may occur in 10-30% of these patients, and although it generally remains “silent,” in rare cases it may lead to extensive hepatic necrosis (Bacha et al. 1994; Erkan et al. 2001; Nordin et al. 2001). These are the two serious complications of a benign disease that may eventually require liver transplantation. In this paper, we present a case of bile duct injury following open cholecystectomy, which resulted in secondary biliary cirrhosis requiring liver transplantation and also review the world literature.

CASE REPORT

A 10-year old girl underwent open cholecystectomy at another institution in 1987. At laparotomy, the gallbladder was found to be fistulized to the duodenum, and cholecystectomy and duodenal repair were performed. The postoperative period was complicated with both biliary and duodenal fistulae. The biliary fistula was treated by a Roux-en-Y hepaticojejunostomy (HJS), whereas the duodenal fistula was treated conservatively. This second operation showed a definite biliary tract injury, with no signs of any congenital abnormality.

Seven years later, she was referred to our institution, for recurrent cholangitis. She was followed conservatively for one year but her clinical course warranted an intervention. Her percutaneous transhepatic cholangiography (PTC) revealed a Bismuth III stricture. Preoperative color Doppler ultrasound showed normal flow pattern in both the portal vein and hepatic arteries. During exploratory laparotomy, which was done seven years after her first operation, the HJS was found to be stenosed, and surrounded by dense, fibrotic tissues. Although the preoperative surgical strategy was to perform a HJS alone or following extended hepatectomy, attempts to find healthy bile ducts suitable for anastomosis were unsuccessful and a portoenterostomy (similar to

Fig. 1. Percutaneous transhepatic cholangiography. A: Complex biliary strictures in the hepatic hilus. B: Balloon dilation of the strictures. The strictured and dilated segments are marked with arrows.

Fig. 2. T2-weighted Magnetic Resonance Imaging showing hepatic abscess in segment 7. The abscess is marked with arrows.
the Kasai procedure) was performed. An intraoperative liver biopsy confirmed presence of biliary cirrhosis. She was started on ursodeoxycholic acid treatment. Except for self-limiting or mild episodes of cholangitis, her bilirubin level remained below 2 mg/dl for six years until June 2000, when the total bilirubin level rose to 4.2 mg/dl and did not become lower than 2.7 mg/dl until October 2000. Her ultrasonographic evaluation showed dilation of the intrahepatic biliary tree. PTC and balloon dilation of the hilar strictures were performed (Fig. 1A and B). Her bilirubin level gradually decreased to 1.4 mg/dl by May 2001. However, between October 2001 and February 2002, she was hospitalized three times for severe sepsis due to hepatic abscesses in the right lobe (Fig. 2). She underwent percutaneous drainage and required admission to the intensive care unit. Radiological examinations performed at that time did not show significant dilation of the biliary tree, therefore PTC and balloon dilation were not attempted. Given the extremely short intervals between the three life threatening episodes and the rapid progression from prodromal symptoms to severe sepsis, it was decided to perform a liver transplantation. Her preoperative liver function tests were as follows: total bilirubin, 4.7 mg/dl; aspartate aminotransferase 47 U/L; alanine aminotransferase 42 U/L; alkaline phosphatase 478 U/L; albumin 3.0 g/dl; and prothrombin time 16 seconds. The Magnetic Resonance Imaging showed signs of cirrhotic liver, left hepatic lobe atrophy and dilation of the intrahepatic bile ducts. After obtaining informed consent, orthotopic liver transplantation from a blood group identical donor was performed. At operation, the hepatic artery and its branches were found to be intact. Her postoperative course was uneventful. The pathologic findings were consistent with inactive cirrhosis. She is leading a healthy life at 4 years post transplantation.

**DISCUSSION**

The hepatic vasculature and the biliary tree may be injured during cholecystectomy (Lillemoe et al. 1997; Nishio et al. 1999; Erkan et al. 2001; Koffron et al. 2001; Buell et al. 2002; Bilge et al. 2003). Although bile duct injury is more common, detailed studies show that “silent” concomitant arterial injury may be detected in patients with bile duct injury (Erkan et al. 2001; Koffron et al. 2001; Nordin et al. 2001). In rare cases, vascular injury occurs alone and causes dramatic tissue damage that initiates a series of events that lead to liver transplantation. The first report on such a patient was published in 1994. Inadvertent clipping of the right hepatic artery resulted in the development of a hepatic abscess in the right lobe and “necrosis in the central bile ducts” (Bacha et al. 1994). In the absence of extensive hepatic infarction due to vascular injury, the liver can usually be salvaged by treatment of the biliary lesion. However, since 1994, reports of patients who required liver transplantation for secondary biliary cirrhosis due to biliary tract injury (with and without concomitant vascular injury) have been published (Bacha et al. 1994; Robertson et al. 1998; Loinaz et al. 2001; Nordin et al. 2001, 2002; de Santibanes et al. 2002; Fernandez et al. 2004) (Table1).

Surgery is the main treatment method for complex biliary injuries. Unfortunately, a subgroup of patients (0-19%) has less than satisfactory outcome during a mean follow-up of only 12-36 months (Ahrendt and Pitt 2001). The data on the long-term outcome of these patients are sparse. Although some centers report successful results in almost all patients with a combination of surgery and radiological techniques (Lillemoe et al. 1997; Johnson et al. 2000), biliary cirrhosis cannot always be prevented (Nordin et al. 2001). It must be noted that the patient reported in this paper had to undergo transplantation 15 years after cholecystectomy.

Roux-en-Y HJS is the technique of choice in the management of bile duct injuries occurring during open or laparoscopic surgery. Nevertheless, it is a technically demanding operation, which can ultimately lead to secondary biliary cirrhosis, especially if it fails in the first attempt (Mirza et al. 1997; Nordin et al. 2002; Fernandez et al. 2004). One possible cause of reconstruction failure may be concomitant vascular injury, which is clinically silent in the majority of the patients.
Although some reports accept concomitant vascular injury as a significant prognostic indicator, autopsy findings and anatomical studies do not support this idea. The authors do not recommend routine angiography since an association between vascular injuries and clinical outcome has not been shown (Bilge et al. 2003).

It must also be stressed that in many papers, outcome is rated as excellent “if there were no symptoms attributable to the biliary tract injury or reconstruction” or good “if mild symptoms not requiring invasive investigation and treatment” (Lillemoe et al. 1997; Ahrendt et al. 2001). In other words, these “success” definitions include patients with residual cholestasis that has been asymptomatic during follow up. The long-term fate of the patients without excellent results according to the criteria proposed by Schweizer et al. (1991) (normal alkaline phosphatase, freedom from cholangitis, no obstruction and/or calculi on radiological investigations) has not received adequate attention. This issue is particularly important because the mean age of the patients with biliary injury is in the 40-50 ranges, i.e., a patient group with long life expectancy (Lillemoe et al. 1997; Johnson et al. 2000; Loinaz et al. 2001; Buell et al. 2002). This case report and review of the literature suggests that liver transplantation will be a treatment modality for a selected group of patients with end-stage liver disease secondary to bile duct injury.

References


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<th>Author</th>
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<td>OC</td>
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<td>2002</td>
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<td>LC</td>
<td>None</td>
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<td>De Santibanes et al.</td>
<td>2002</td>
<td>5</td>
<td>3 LC, 2 OC</td>
<td>2 HJS, 3 EEA-T tube</td>
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<td>LC</td>
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<td>OC</td>
<td>HJS</td>
<td>Biliary: 1; Vascular: 0</td>
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</table>

LC, laparoscopic cholecystectomy; OC, open cholecystectomy; HJS, hepaticojejunostomy; EEA, end-to-end anastomosis.

* This report also contained the three patients that were published in 2001, which were excluded from this table, to prevent double count.


