Spontaneous choledoco-enteric fistula formation after bile duct injury due to laparoscopic cholecystectomy: A case report

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**Abstract**

In situations like peritonitis due to bile-duct injuries drains insertion give an opportunity for healing. However in presented case the bile leakage from drains spontaneously stopped. Only constipation leads to icterus. A spontaneous choledoco-enteric fistula explains these conditions, discovered later. It could be the result of inflammation causing adhesion.

**Keywords:** bile duct injury; choledoco-enteric fistula; laparoscopic cholecystectomy.

**Abbreviations:** MRCP: Magnetic Resonance Cholangio Pancreateography; ERCP: Endoscopic Retrograde Cholangio Pancreatography; BDI: Bile Duct Injury; CBD: Common Bile Duct; MR: Magnetic Resonance.

**Background**

Cholelithiasis is a common disease (10-20% in Americans) and only less than 20% of patients get to be symptomatic [1,2]. Gold standard for treatment is laparoscopic cholecystectomy [2] and it is because of low mortality (0.23% vs 1.90%, P<.0001) and low complication in comparison to open cholecystectomy [3]. However with increasing of laparoscopic cholecystectomy, rate of Bile Duct Injury (BDI) has been increased (0.06% to 0.3%).

Some complications of laparoscopic cholecystectomy are: Bile duct injury and bile leakage, haemorrhage, infection, respiratory problems, Trocar injury to intra-abdominal viscera and great vascular damage [2]. Surgeries on gallbladder, pancreas and stomach could lead to BDI and laparoscopic cholecystectomy is cause of 80-85% of them. In one study, not significantly, BDIs after laparoscopic cholecystectomy are twice in comparison with open surgeries [4].

0.3-2.7% of patients, after cholecystectomy, have bile leakage. In one study most common sites of bile leak were: Cystic duct stump, ducts of Luschka, the T-tube site and other sites [5].

We have a case with Strasberg type E3 (bismuth type 3) injury which we placed 2 drains and didn’t make an anastomosis because of peritonitis; but the bile leakage from drains spontaneously decreased (that it was almost impossible [6]) and stopped. Only a spontaneous choledoco-enteric fistula formation can explain it.

**Case presentation**

A 27-years old women with chief compliant of abdominal pain in right upper quadrant, radiated between two scapulas, had been admitted. Other symptoms were nausea and vomiting. Evaluation of liver function tests, alkaline phosphatase and Bilirubin were normal. In ultrasonography of the abdomen, thickness of gallbladder wall was increased and cholelithiasis were detected. Common Bile Duct (CBD) and intrahepatic ducts were normal. The patient with probable diagnosis of acute cholecystitis had undergone a laparoscopic cholecystectomy.

After six days, she complicated with colic form epigastric pain and dark brown colour of urine. In laboratories evaluations, total bilirubin of 5.6 mg/dl, direct bilirubin of 3.5 mg/dl, Aspartate
Transaminase (AST) of 18 u/L, Alanine Aminotransferase (ALT) of 48 u/L and alkaline phosphatase of 695 u/L were detected.

Ultrasonography revealed dilation of CBD (9 mm) and intrahepatic duets. Then, MRCP or ERCP was recommended. In this way, she was referred to our hospital. By ERCP, complete cut-off of proximal part of CBD was seen which neither guide wire nor contrast liquid could not pass. Consequently, there was not possibility of stent insertion. In addition, severe deodenitis was detected.

During these procedures, the patient was get complicated by fever, bile leakage from laparoscopic holes and acute abdomen condition. In term of liver function tests, AST, ALT and alkaline phosphatase were 64 u/L, 59 u/L and 560 u/L, respectively. Total bilirubin was 9.2 mg/dl with direct bilirubin of 6.2 mg/dl. Emergent laparotomy was done.

Within the operation, the peritoneum cavity consisted 3-4 ml of bile which was suctioned. In exploration of the cavity, firm adhesion of omentum to liver hilum and recent region of cholecystectomy was found. Thus, the adhesion slightly was detached. Additionally, complete cut-off of CBD and hepatic bile duct was seen. The hilum was widely burned due to excess bile leakage from choleductus. Regarding to this condition, hilum of liver was irreparable. Therefore, after washing the peritoneum and placing two drains, the abdomen was closed. Her signs and symptoms were improved in ward and after 2 weeks, the patient discharged with 2 drains. After 2 months, her bile leakage from the drains decreased spontaneously and ended up finally. The drains, also, extinguished spontaneously.

She was followed up without having any problem. In ultrasonography of abdomen, the patient did not have any collection. In MRCP that was done, dilation of intrahepatic bile ducts and continuities of them up to portohepatic area was seen. Additionally, common hepatic duct and proximal portion of the CBD were not appeared (Figure 1). These events just can be explained by spontaneous choledoco-enteric fistula formation.

After 3 months, the patient came back with complications of icterus and right upper quadrant tenderness. Again, liver function tests increased up to about 1.5 times and total bilirubin was 5.8 mg/dl. According to previous approaches, MRCP or ERCP would be useless. Additionally, there is no gold standard for assessment and treatment in conditions like this. As a result, laparotomy with the aim of anastomosis of ducts to the duodenum was done. In laparotomy, Strasberg type E3 (bismuth type 3) injury of bile ducts and spontaneous fistula (length of 2 cm and diameter of 1-2 mm) from Duodenum (D2) to hilum of liver with stenosis were seen (Figure 2). Unfortunately, due to wide burning status of the hilum, the surgery of Hepaticojejunostomy Roux en Y could not be done. A temporary drain was again inserted, waiting to improvement of the area for liver transplantation. After a while, again, jaundice spontaneously improved and bile leakage from drains decreased and then stopped.

In the following up, according to the patient’s claim, when she has constipation, she gets icterus and with good function of intestinal tract her icterus improves. A good explanation to this condition was not found. Additionally, she is candidate for liver transplantation because of progressive liver ducts damages and impossibility of makinganastomosisfrom ducts to duodenum.

Discussion & conclusion

Bile Duct Injuries (BDIs) mostly occur in healthy young people and can affect their quality of life and overall survival. These injuries cause to bile leakage (with frequency of less than 2%). Bile leakage can appear first week after operation [4,6].

For approach and management of BDIs there are classifications like Bismuth, Strasberg, Mc Mahon, Stewart-Way, Hannover and Mattex. Bismuth and Strasberg classification are most used classifications. Strasberg classification is modified type of Bismuth classification. Small to serious damages in Strasberg classification considered as type A to D and for Bismuth classification it is type E (E1 to E5). Most important disadvantages of Bismuth classification is no including the wide spectrum of possible biliary injuries. And Strasberg classification doesn’t consider right and left partial injuries and vascular involvement [3,4].
In the presented case, the injury could be classified as Strasberg type E3 (bismuth type 3).

In term of evaluation of BDIs, ultrasonography, percutaneous cholangiography and nowadays MR (Magnetic Resonance) cholangiography are used [4]. 20% of the surgeons can make a tension free biliary-enteric anastomosis by laparoscopy [6] and Roux en Y Hepaticojejunostomy is the main surgery for bile leaks. In good conditions, anastomosis would be done; but if there is chemical (bile leakage) or infectious peritonitis, the surgeon should insert the drain to give the opportunity of improvement of the inflammatory statements and makes a good anastomosis chance for the next operation [4] as it was done in this patient for two times.

BDIs can appear with diffuse abdominal pain, nausea, fever, ileus, bile collections, peritonitis, leukocytosis and rising of bilirubin (direct and indirect) and mostly with Strasberg type E injury. Delaying in BDIs diagnosis and intervention will cause to chronic liver disease, cirrhosis and portal hypertension [4].

In such cases requirement to multiple repair attempts will lead to candidacy of patient to liver transplantation. Because of complete cut offin Strasberg type E injuries, surgeon have to make a hepaticojejunal anastomosis with specifications of “low tension” and “well-perfused”. Choledoco-choledoco or hepatico-duodenum anastomosis in these cases mostly lead to high tension anastomosis [4].

According to Strasburg’s et al study about the best post-operative outcome condition, by making wide and high quality hepaticojejunal anastomosis beside enough vascularization, hepatic injuries will be protected [18].

Mercado et al about good results of BDIs repair operation recommended to use the best vascularized duct with large diameter for decreasing the tension; and sutures with lesser reaction to epithelium to mucosa apposition [4].

But in this case, because of wide injury and inflammation, making an anastomosis was almost impossible. Spontaneously formed fistula with unsuitable conditions leads the requirement of liver transplantation.

Spontaneous fistula can be result of inflammatory state in the area that cause to adhesion of intestine and proximal part of choleductus and formation a fistula between them [6].

According to Sezgin’s et al case report [6] this is 2nd case of spontaneously choledoco-enteric fistula formation, after bile duct injury, due to laparoscopic cholecystectomy.

Spontaneous choledoco-enteric fistula formation could happen rarely. Management of these patients is complicated and needs an appropriate approach. We decided to utilize this fistula until it works and only in relapse of signs such as icterus do an intervention. But the good function of intestine should be considered.

Declarations

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethics approval: Not applicable.

Availability of data and material: All data and material is available for review by the Editor-in-Chief of this journal on request.

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