Percutaneous Transhepatic Cyst Drainage as a “Bridge Procedure” to Definitive Treatment of Perforated Choledochal Cysts

A Case Report

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Abstract: Perforated choledochal cysts are rare. Traditionally, they have been managed by primary T-tube choledochostomy and interval cystectomy with hepaticojejunostomy. We present here our recent experience with a case of perforated choledochal cyst, which was managed with preoperative percutaneous transhepatic cyst drainage, followed by definite surgery for choledochal cyst, cyst excision, and reconstruction with Roux-en-y hepaticojejunostomy. Preoperative percutaneous cyst drainage seems to be useful and effective in managing complicated choledochal cysts. The procedure garners time for patients’ general condition and laboratory findings to improve to the extent that general surgery can be performed.

Key Words: choledochal cyst, percutaneous transhepatic cyst drainage, perforation

(C Surg Laparosc Endosc Percutan Tech 2008;18:598–600)

Choledochal cyst is a relatively uncommon disease characterized by congenital dilatation of the intrahepatic and/or extrahepatic portion of the biliary tree. Todani et al’s classification system,1 a modification of the Alonso-Lej et al’s classification scheme,2 is widely accepted for the identification of choledochal cyst. Most cases of choledochal cyst present with abdominal pain, jaundice, or an associated abdominal mass, but this symptom triad is rarely seen together, and patients usually present with some combination of symptoms and signs. Historically, only cyst drainage procedures were employed to treat the condition, but such procedures have recently been avoided because of an elevated risk of cholangiocarcinoma during follow-up.3,4 Complete cyst excision and Roux-en-y hepaticojejunostomy are regarded as the standard treatment.5

Spontaneous perforation is a rare complication of the choledochal cyst. Its frequency has been reported from 1.8% up to 7% in the literature.6–10 Therefore, many surgeons are unfamiliar with this clinical presentation and face particular difficulty in treating the condition. Often, inflammation and spilled bile around the choledochal cyst and peritoneum may make it even more challenging for the surgeon to find the exact anatomic planes for surgical dissection. Further complicating matters, the general condition of a patient with a ruptured choledochal cyst may not allow for safe surgical correction under general anesthesia because of bile peritonitis. In cases of “huge” choledochal cysts, handling of the huge cyst in the operative field is expected to be especially difficult because the anatomic relations of the cyst and other surrounding structures could be obscured by the cyst itself. Despite all this, there have been reports of single-stage surgical reconstruction of ruptured choledochal cysts,11 even though the traditional management of perforated choledochal cysts is T-tube drainage and interval cystectomy with hepaticojejunostomy.

Here, we present our recent experience with a perforated choledochal cyst that was managed by initial percutaneous transhepatic cyst drainage followed by definite correction of the choledochal cyst. We focus our discussion on the usefulness of the preoperative cyst drainage procedure.

CASE

A 24-year-old woman, with a history of open heart surgery in 1997 to repair an atrial septal defect, was referred to the emergency department with a diagnosis of choledochal cyst. She presented with acute abdominal pain, nausea, and vomiting, but no fever. Her pulse rate was over 120/min and her blood pressure was 110/80 mm Hg. On physical examination, the patient seemed acutely ill and had a distended abdomen with diffuse direct and indirect tenderness. Mild-to-moderate abdominal rigidity was detected. Although her sclera did not seem icteric, her lips and tongue were dry. Initial laboratory tests revealed moderate leukocytosis with the neutrophil proportion greater than 85%; hemoconcentration (Hb:Hct, 17.3 g/dL: 49.8%), hypoalbuminemia (2.6 mg/dL), azotemia (blood urea nitrogen:Cr, 30.4 mg/dL:1.4 mg/dL), hyponatremia (Na, 125mmol/L), amylase (455 U/L), and lipase (169 U/L). The level of transaminases was also mildly elevated, and the initial...
total bilirubin level was 2.2 mg/dL. A computed tomography scan of the abdomen and pelvis showed significant peritoneal fluid in the peritoneal space and a cystic structure with a ruptured anterior wall (Fig. 1). Clinically, the patient’s preoperative condition was thought not to be appropriate to perform emergent major operation and her symptom of abdominal pain was very severe. In addition, it was necessary for the patient to wait several times till emergency operation was available in the middle of so many other elective surgeries. Therefore, we decided to perform the percutaneous approach first to gain more time to stabilize the patient’s condition and reduce the intensity of severe abdominal pain.

Normal healthy persons usually have a hard time receiving elective endoscopic retrograde cholangiopancreatography. Therefore, we thought the endoscopic procedure might not be appropriate in this critically ill patient. If the purpose of the procedure was similar, the percutaneous approach seemed to be the simple and more appropriate procedure for this patient. In addition, she had concomitant pancreatitis at presentation; so, the endoscopic transpapillary procedure might have the risk of inducing more severe pancreatitis after endoscopic retrograde cholangiopancreatography. Management began with the insertion of a peritoneal pig tail catheter and intermittent lavage with sterile isotonic saline for symptomatic relief because she apparently needed time for preparation for emergency laparotomy. However, unexpectedly, we could notice her symptoms were markedly alleviated by this extemporaneous maneuver. Two days later, percutaneous transhepatic cyst drainage was performed to divert bile flow, with the expectation that the amount of bile in the peritoneal space would be reduced. The usual technique of percutaneous transhepatic biliary drainage was employed. At supine position, the patient’s right upper quadrant was draped in a sterile manner. Under the guidance of an ultrasound scan, a transhepatic puncture of choledochal cyst was performed. The right position of the tip of the puncture needle was ensured by a cystogram and introduced guide wire. Pig-tail catheter (8.5 Fr) was inserted along the previously introduced guide wire and an external fixation to the skin was made. After ensuring symptomatic improvement and only minor drainage was sustained through the peritoneal pig-tail catheter, a cholangiography was obtained from a percutaneous cyst-draining catheter. The patient was found to have a type I choledochal cyst with spontaneous sealing of the perforation site and an anomalous pancreatico-biliary junction (Fig. 2). Therefore, considering cost effectiveness, additional magnetic resonance cholangiopancreatography was not performed because it was not thought to be critical to decide the surgical management plan in this patient. Seven days after cholangiography, the patient underwent definite correction of choledochal cyst: cholecystectomy, excision of the choledochal cyst, Roux-en-y hepaticojejunostomy, and incidental appendectomy because secondary inflammatory change of the appendix was suspected owing to previous bile peritonitis during exploratory laparotomy. The patient was discharged from the hospital without complications on the seventh postoperative day. The patient has been followed up regularly for about 2 years without any clinical problems.

**DISCUSSION**

Several theories propose mechanisms for spontaneous perforation of choledochal cysts. One theory suggests that perforation of choledochal cysts is related to an abrupt increase in intraluminal pressure in the common bile duct owing to obstruction by a protein plug at the common channel. Another, based on 13 cases of spontaneous choledochal cyst perforation, relates the etiology of a perforation to epithelial irritation of the biliary tract caused by a protein plug at the common channel.
by refluxed pancreatic juice in the pancreatico-biliary malunion and to mura immaturity owing to infancy. As most perforations reported in the literature involve the anterior or posterior wall of the choledochal cyst, ischemic insult associated with increased intraluminal pressure is also proposed to explain spontaneous perforation. However, it is generally accepted that no single factor is responsible for cyst perforation; rather, several mechanisms are likely acting concurrently in choledochal cyst perforations.

As has been mentioned previously, perforation of choledochal cysts is a particularly difficult clinical issue. In addition to surgeons’ lack of clinical experience with perforated choledochal cysts, the situation is complicated by free perforation into the peritoneal cavity, which can cause bile peritonitis and then require emergency exploratory laparotomy regardless of a patient’s general condition. Inflammation and bile staining around the choledochal cyst and peritoneum make it even more difficult for the surgeon to identify the appropriate anatomic planes for surgical dissection. Consequently, perforations of choledochal cysts have traditionally been managed conservatively with T-tube drainage and internal cystectomy with hepaticojejunostomy. But even this approach requires two surgeries each under general anesthesia. Although single-stage cystectomy and reconstruction of hepaticojejunostomy are available, it can only be used in a select few patients. Another approach to this clinical problem is needed, which must have the same therapeutic effects as the current treatment while also being minimally invasive.

To the best of our knowledge, our report is the first of a preoperative percutaneous cyst drainage procedure for treating cases of perforated choledochal cyst. Our recent experience of percutaneous cyst drainage in complicated choledochal cyst apparently has several advantages. First, it reduces the amount of bile spillage into the peritoneum in case of perforated choledochal cyst. This occurred because spontaneous sealing of the perforation site happens when bile flow is diverted from the choledochal cyst to the external drainage tube. In the present case, we initially inserted a pig-tail catheter into the peritoneal space to remove the large amount of bile that had collected there because she needed to wait for some time for urgent operative preparation. Symptomatic relief was achieved as more than 800 mL of bile was drained in a single day. This unexpected clinical improvement prompted us to have a sustaining conservative strategy in managing this case, which resulted in subsequently inserting a percutaneous cyst drainage catheter to divert the bile flow. Follow-up cholangiography several days later showed spontaneous sealing of the choledochal cyst perforation site. We were able to remove the intraperitoneal pig-tail catheter before definite surgery because the daily drainage amount decreased to almost nothing. The second advantage of our preoperative cyst drainage procedure is that, even though we could detect the perforated choledochal cyst by abdominal pelvic computed tomography, we could not determine which type of choledochal cyst the patient had or what kind of anomalous pancreatico-biliary junction existed. Before definite correction of choledochal cyst, a cholangiograph taken by the percutaneous cyst drainage tube answered these questions for us.

However, there are important considerations to bear in mind when applying this percutaneous drainage procedure before definitive correction of choledochal cysts. In the first case presented here, external cyst drainage and peritoneal irrigation through the intraperitoneal pig-tail catheter allowed us to confirm lessened bile staining in the peritoneal organ with only mild-to-moderate inflammation around the pericholedochal cyst. Definite surgery was complicated by residual pancreatitis, however, especially when we were dissecting the choledochal cyst from the inflamed pancreatic tissue. Therefore, we think the proper time for definite surgery must be chosen carefully. It could be chosen on the basis of both clinical symptoms and laboratory findings. We performed definite surgery when the level of amylase/lipase was between 100 and 150 IU/mL, but dissection between the choledochal cyst and pancreas would likely have been easier if we had performed definite surgery after confirming resolution of pancreatitis.

Our case suggests that percutaneous transhepatic cyst drainage preceding definite surgery can be considered when patients have perforated choledochal cysts associated with bile peritonitis or other risk factors for emergent operative morbidity. However, we need more experience with this procedure to address the safety and effectiveness of this approach to complicating choledochal cyst.

REFERENCES


