Percutaneous Transhepatic Biliary Interventions In Benign Diseases of Children
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Abstract

Background/Purpose: Percutaneous transhepatic biliary interventions are performed safely and effectively in adults. There is less experience of these interventions in benign diseases of children. We aimed to evaluate the safety and efficacy of percutaneous biliary interventions in benign diseases of children.

Methods: In this retrospective study, percutaneous biliary interventions were performed in fifteen children with a mean age of 10.2 years (range 14 days-14 years). Patients presented with jaundice (n=5) and/or cholangitis (n=10). Percutaneous transhepatic biliary drainage (PTBD) performed in 10 patients, PTBD plus balloon dilation in 3, percutaneous cholecystostomy (PC) in 1, PTBD following PC in 1.

Results: All procedures were technically successful. No procedure-related mortality occurred in patients. Serum bilirubin levels returned to normal or near normal in ten of twelve cases. Preexisting cholangitis and acute cholecystitis resolved in all patients. Six patients underwent surgery following percutaneous management. Nine patients cured primarily with percutaneous interventions with no further treatment.

Conclusion: Percutaneous biliary interventions can be performed effectively in benign diseases of children. It can be performed either as a primary treatment modality or as a bridge prior to surgery. In most of cases, percutaneous treatment is sufficient and unnecessary surgery is prevented.

Keywords: Percutaneous; Catheter; Biliary; Drainage; Children; Benign Diseases

Keypoints:
* Malignant biliary obstruction and/or cholangitis can be treated percutaneously
* Biliary obstruction caused by a benign etiology in children can be safely cured by percutaneously
* Percutaneous biliary interventions can prevent unnecessary surgery in children with benign biliary obstruction and/or cholangitis

Abbreviations

PTBD: Percutaneous Transhepatic Biliary Drainage
PTBD-E: External Percutaneous Transhepatic Biliary Drainage
PTBD-IE: Internal-External Percutaneous Transhepatic Biliary Drainage
PC: Percutaneous Cholecystostomy
PTC: Percutaneous Transhepatic Cholangiography

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Introduction

Percutaneous transhepatic biliary interventions are performed widely in order to relieve obstructive jaundice due to benign or malignant conditions in adults since 1970’s [1]. These included percutaneous transhepatic biliary drainage (PTBD) which should be performed either as external biliary drainage (PTBD-E) or internal-external biliary drainage (PTBD-IE), insertion of metallic stents, balloon dilation, and percutaneous cholecystostomy (PC). The initial step of a percutaneous transhepatic biliary drainage procedure is percutaneous transhepatic cholangiography (PTC). Following PTC, according to the disease and patient’s condition, further intervention is performed. Metallic stents are placed permanently into bile ducts in order to palliate malignant obstructive jaundice in unresectable cases [2-4]. In surgical candidates, PTBD is performed with plastic catheters placed temporarily into bile ducts in order to decrease serum bilirubin levels preoperatively. Balloon dilation is performed either primarily in order to achieve satisfactory channel diameters in benign bile strictures, or during insertion of metallic stents in order to relieve malignant strictures [5]. PC has been shown to be a safe treatment option for patients suffering from acute choledocolitis but at high risk for emergency surgery. After clinical improvement obtained with emergent PC, elective cholecystectomy should be done with lower morbidity and mortality.

Although efficacy and safety of these biliary interventions are well established in adults [2,7] and children with malignant obstructions [8,9], there is less experience of percutaneous transhepatic biliary interventions in benign diseases of children [10]. In this article, we aimed to evaluate the safety and efficacy of percutaneous transhepatic biliary interventions in benign diseases of children.

Materials and Methods

This retrospective study is approved by the ethical committee of our faculty. Patients and/or their parents gave their informed consent prior to intervention.

Between years 1997-2009, percutaneous transhepatic biliary interventions were performed on 15 children (mean age: 10.2 years, median age 12 years) with benign biliary disease (Table 1). Five children had postoperative strictures following hepatocjejunostomy (3), cholecystectomy (1) and operation for a gunshot wound (1). Five children had choledochal cysts, and two had acute calculous cholecystitis (one related to sickle cell anemia and one to hemobilia following liver biopsy). There were single cases of hydatid cyst (Echinococcus granulosus) with rupture into the bile duct, alveolar echinococcosis (Echinococcus multilocularis infection), and ceftriaxone-associated sludge ball.

Indications for performing percutaneous biliary drainage were obstructive jaundice and/or bile infection (cholangitis or cholecystitis). Twenty-nine patients with malignant etiology causing biliary tract obstruction are excluded from the study. PTBD performed in 10 cases with placing plastic catheters. PTBD procedures performed as PTBD-E in 4 cases, PTBD-IE in 4 cases, and PTBD-E converted to PTBD-IE in 2 cases. In three cases balloon dilation and following PTBD-IE performed. If possible, we preferred to perform PTBD-IE instead of PTBD-E, because of catheter stability and establishment of bile flow to duodenum. When it was not possible to perform PTBD-IE, we avoided from extended manipulations which can cause septic complications and performed PTBD-E. Initial PTBD-E is converted to PTBD-IE after several days of gravity drainage in two of our cases. PC performed in one case, and PTBD-IE following PC in one. In both cases, PC performed via transhepatic approach in order to establish catheter stability.

Technique

Percutaneous transhepatic access into the intrahepatic bile ducts (during PTC) or gallbladder (during PC) was performed via the right intercostal approach with a 21-G needle with a stylet (Accustick II Introducer System, Boston Scientific, Natick, Mass., USA) under US guidance. PC performed via transhepatic route in both cases. All PTBD procedures were performed following PTC. After the entrance of needle into bile duct or gallbladder, fluoroscopy was used in further manipulations. Contrast material is injected in order to perform a PTC or cholecystogram. Through the 21-G needle, a 0.018-inch guidewire is placed into the bile duct or gallbladder. Over this wire, the AcuStick coaxial dilator is advanced into the bile duct or gallbladder. If the case is PC, a 0.035-inch stiff guidewire (Amplatz Super Stiff Wire, Boston Scientific, Natick, Mass., USA) is advanced into the gallbladder lumen. Then a 8.5 Fr pigtail cathe-
ator (Ultrathane; William Cook Europe, Bjaeverskov, Denmark) is placed into the gallbladder lumen over this stiff wire. If the case is PTBD, a 0.035-inch angled tip hydrophilic guidewire (Terumo Corp., Tokyo, Japan) is inserted through the Accustick dilator in order to cross the biliary stricture. After crossing the stricture with the hydrophilic guidewire over a 5 Fr end-hole angiography catheter, the hydrophilic guidewire is exchanged for a stiff guidewire. Over this stiff guidewire, biliary drainage catheter ranging in size from 8.5- to 10 Fr (Ultrathane Biliary Drainage Catheter; William Cook Europe, Bjaeverskov, Denmark) is placed either as external or internal-external biliary drainage. After checking the correct location of the drainage catheter with a transcatheter cholangiogram or cholecystogram, catheter is sutured to skin and left to external gravity drainage. Balloon dilation is performed with a 8-10 mm diameter balloon (Bluemax 20, Boston Scientific, Galway, Ireland). Balloon is inflated up to 18 atm pressure until the stricture is fully dilated. Following balloon dilation, in order to establish persistent biliary patency, a larger diameter (preferably 10 Fr) biliary drainage catheter is placed as PTBD-IE for 7-24 days. Biochemical tests and imaging findings are checked 2-33 days after the intervention in order to assess the success and safety of the procedure. Following clinical and laboratory improvement, patient is either operated or catheter is removed after transcatheter cholangiogram if no further treatment is needed. Clinical follow-up of patients done in order to detect possible late complications and to determine the safety and efficacy of the treatment.

Results

Clinical features of patients at presentation and the kind of interventions performed are summarized in Table 1. Patients presented with jaundice (n=5), and/or cholangitis (n=10). Percutaneous transhepatic biliary interventions were successfully performed in all patients. The technical success rate was 100%. No procedure-related deaths or major complications occurred. No patient died following the 30 days of intervention.

Twelve patients had bilirubinemia prior to procedure. In these 12 patients, mean serum total bilirubin levels were 285 µmol/L (range 70-1368 µmol/L) before the procedure. Laboratory and clinical improvement occurred in all 12 patients at 2-33 days following the intervention. Mean serum total bilirubin levels decreased to 39 µmol/L (range 17-147 µmol/L) following percutaneous drainage procedures. Findings of cholangitis such as abdominal pain, fever, and leucocytosis were resolved in all 8 patients following the intervention. Symptoms and signs of acute cholecystitis resolved in both of two cases. In case 13 (calculus cholecystitis) improvement occurred 4 days after PC. But, in case 14 (acalculous cholecystitis), initially performed PC did not supplied satisfactory clinical improvement. For this reason, PTBD-IE performed following PC and clinical improvement occurred following 3 days after PTBD-IE.

In 2 cases (Cases 2 and 12) initial PTBD-E were converted to PTBD-IE in order to prevent catheter dislodgement and external salt loss with bile.

Minor (transient) hemobilia occured in 3 of 15 cases (20%) (Cases 6, 7, 10). The procedure performed was balloon dilation plus PTBD-IE with a large-bore (10 Fr diameter) catheter in all of 3 cases. Hemobilia was self-limiting in all 3 cases and none of them required transfusion or further treatment. In one case (case 3), PTBD-E was dislodged spontaneously even though it had been tightly sutured to the skin. In this case, catheter was redirected distally and placed again just proximal to the obstruction.

Nine of fifteen (60%) cases treated primarily (definitive treatment) with percutaneous transhepatic biliary interventions and no further treatment is needed. In case 3 (Carol's disease presented with cholangitis) cholangitis resolved following PTBD-E and patient placed in liver transplantation waiting list. In five cases of postoperative biliary strictures (cases 6,7,8,9,10), percutaneous interventions (Balloon dilation plus PTBD-IE in 3 cases and only PTBD-IE in 2 cases) successfully solved the problem without any need for further surgery (Figures 1 and 2). Also, in case 11 (Echinococcus granulosus cyst communicated with the bile duct) (Figure 3), case 14 (acute acalculous cholecystitis due to hemobilia) (Figure 4), and case 15 (Figure 5) (ceftriaxone-related sludge ball), PTBD-IE alone successfully solved the problem. In all of these 9 cases, biliary catheters removed after clinical, laboratory and radiological improvement. Mean duration of catheter stay was 14.5 days (range: 2-33 days).

In six of fifteen (40%) cases, patients gone to surgery following PTBD-E (3 cases), PTBD-IE (2 cases) (Figure 6), and PC (1 case). In these 6 cases, percutaneous transhepatic biliary interventions performed as a bridge prior to surgery and liver function tests improved preoperatively following percutaneous interventions. In four of six cases (cases 1,2,4,5) diagnosis were choledochal cysts, one (case 12) was Echinococcus multilocularis (alveolaris), and one (case 13) was acute calculus cholecystitis. In all of these 6 cases, patients gone to surgery with their biliary drainage catheters and catheters removed during or after surgical procedure. Mean duration of catheter stay was 16.3 days (range: 12-27 days).

Fourteen patients are followed up clinically at a mean time of 52.4 months (range 9 to 144 months). One case (case 3) was lost during follow up.
Figure 1a: (Case 8) HJ anastomosis stricture. PTC shows stricture (white arrow) at HJ anastomosis.

Figure 1b: After 33 days of PTBD-IE, transcatheter cholangiogram showed that anastomosis stricture is resolved.
Figure 2a: (Case 9) Operated gun shot wound (3 months ago). Postoperative stricture (black arrow) at proximal choledochus.

Figure 2b: Transcatheter cholangiogram performed at day 13 of PTBD-IE shows that stricture is resolved.

Figure 3a: (Case 11) Hydatid cyst ruptured into the biliary tree. PTC shows contrast material filling into the hydatid cyst (black arrow) from the bile duct (white arrow).

Figure 3b: Fistulous passage from the bile duct to into the cyst is shown in detail.
Figure 3c: Transcatheter cholangiogram performed at day 12 of PTBD-IE. Cyst cavity became smaller and no filling defect seen in bile ducts.

Figure 3d: Transcatheter cholangiogram performed at day 29 of PTBD-IE shows that cyst cavity became smaller and no filling defect seen in bile ducts.

Figure 4a: (Case 14) Hemobilia due to prior liver biopsy. Cholangiogram performed via percutaneous cholecystostomy catheter shows multiple filling defects in gallbladder and bile ducts (white arrows) representing blood clots.

Figure 4b: Transcatheter cholangiogram performed at day 4 of PC shows there are still small filling defects in peripheral intrahepatic ducts.
bile ducts (white arrows). PC catheter removed and PTBD-IE catheter placed following this cholangiogram.

Figure 4c: Transcatheter cholangiogram performed at day 7 of PTBD-IE shows that bile ducts are clear with no filling defects.

Figure 5a: (Case 15) Sludge ball due to ceftriaxon use in a 14 day old baby. PTC shows filling defect in distal choledochus (white arrow) causing biliary obstruction.

Figure 5b: Transcatheter cholangiogram performed at day 2 of PTBD-IE shows that bile ducts are decompressed and clear.

Figure 6a: (Case 1) Type I choledochal cyst. Hilar strictures probably due to cholangitis involving the right and left hepatic ducts. PTBD-E
catheter placed.

Figure 6b: Cholangiogram performed via the PTBD-E catheter shows decompression of the intrahepatic bile ducts and the cystic dilation of the choledochus. Hilar strictures resolved 8 days following PTBD-E.

Discussion

Biliary atresia, postoperative stricture, choledochal cysts and cholelithiasis are the main causes of benign biliary tract obstruction in neonates and children. Cholelithiasis in children is most commonly due to metabolic or hematologic disorders such as hemolytic anemia or sickle cell disease [12,13]. In this study, the most common causes of biliary obstruction were choledochal cysts (n=5) and postoperative strictures (n=5).

Optimal treatment of choledochal cyst is complete surgical excision, cholecystectomy, and Roux-en-Y hepaticojejunostomy (HJ) because there is increased risk of arising carcinoma. When the cyst extends into or involves the intrahepatic biliary tree (type IV-A and type V) complete cyst excision may not be possible [14]. In this study, 4 of 5 choledochal cysts went to surgery following PTBD-E (cases 1, 4, 5) or PTBD-IE (case 2). Indications for preoperative PTBD were jaundice (one case), cholangitis (one case) and jaundice plus cholangitis (two cases). In type I cysts (cases 1, 4, 5), choledochal excision with hepaticojejunostomy is performed. In type IV-A cyst (case 2) extended left hepatectomy and HJ is performed. In type V cyst (Caroli disease) (Case 3), patient placed in liver transplant waiting list following PTBD-E performed to treat cholangitis.

In this study, all cases of postoperative bile duct strictures (cases 6, 7, 8, 9, 10) are treated successfully alone with percutaneous transhepatic biliary interventions. In 3 of 5 bile duct stricture cases (Cases 6, 7 and 10) balloon dilation plus PTBD-IE was performed with a large bore (10 Fr) catheter in order to prevent establish persistent biliary patency. In the remaining two (cases 8 and 9), PTBD-IE alone was performed. Although there is some experience in balloon dilation of biliary-enteric strictures in children following liver transplantation [10,15], to our knowledge, there is no recent study reporting balloon dilation of postoperative biliary strictures in children. Successful percutaneous treatment prevented re-operation in these 5 cases.

In case 11, having Echinococcus granulosus cyst communicated with the bile duct, because of the complicated nature of the cyst (Type III according to Gharbi classification) [16], percutaneous cyst treatment would not have been effective and probably the cyst contents obstructing the bile ducts would not have been removed with direct puncture of the cyst itself. For this reason we preferred to perform PTBD-IE. As it was previously reported in a case of transbiliary drainage in an adult case [17], biliary obstruction caused by the cyst content is successfully cleared with repeating saline irrigations via the PTBD-IE catheter in our case. Biliary obstruction successfully relieved and the cyst diameter reduced (Figure 3).

In case 12, having hepatic Echinococcus multilocularis (alveolaris) mass covering the left liver lobe, diagnosis of alveolar hydatid disease was based on clinical, imaging and serological findings [18]. In this case, total serum bilirubin level was 632 µmol/L at presentation. After 14 days of PTBD-IE performed preoperatively, total serum bilirubin level decreased to 147 µmol/L. Although histopathologically a benign disease, alveolar hydatid disease shows malignant behaviour and the only curative treatment is surgical en-bloc resection. Extended left hepatectomy and HJ anastomosis was performed in this case. In case 13 having acute calculous cholecystitis due to sickle cell anemia, serum total and direct bilirubin levels were 1368 µmol/L and 1026 µmol/L, respectively. PC performed urgently because the patient was critically ill. After 6-8 hours of PC, approximately 500 ml pus drained via the PC catheter. Following both the clinical and laboratory improvement, patient gone to elective cholecystectomy after 15 days of PC. Emergent PC was life-saving in this critically ill patient.

Ceftriaxone, a third-generation cephalosporin, is known to induce reversible precipitations in the gallbladder of children which is known as biliary pseudolithiasis. Biliary sludge due to precipitation of the calcium salt of ceftriaxone is responsible for the phenomenon [19]. In case 15, sludge ball was thought to be due to ceftriaxone use. Initial US diagnosis was choledocholithiasis in this case, so we planned to extract stone percutaneously or push it into the duodenum with a balloon catheter.
But, during PTC and biliary catheterization, filling defects in the choledochal duct dissolved/disintegrated and flowed into duodenum. Our diagnosis of “sludge ball” was obtained during the intervention, not before the intervention. If we were aware of sludge ball before the intervention, we should wait for a possible spontaneous dissolution of the ball. Both the sludge ball in case 15 and blood clots in case 14 were successfully pushed into duodenum with the repeated saline irrigations via the PTBD-IE catheters. Bile ducts were free of filling defects at the time of PTBD-IE catheter removals in both of these two cases. The procedure was safely performed and effective even in a 14 day-old baby (case 15).

No major complication occurred in our series. Minor (transient) hemobilia occurred in our 3 of 15 (20%) cases. In all of these 3 cases, transient hemobilia occurred following balloon dilation and placing large-bore (10 Fr instead of 8.5 Fr) biliary drainage catheter. In order to maintain/stabilize the bile duct stricture dilated with balloon, large-bore (10 Fr) catheters were placed for a longer time interval (7-24 days). In all of three cases, hemobilia was self-limiting and no transfusion or further treatment needed. No serious/major complication occurred related with the diameter of the catheters placed. Another minor complication, dislodgement of the drainage catheter in case 3, is usually expected when PTBD-E is performed instead of PTBD-IE. It is difficult to stabilize the PTBD-E catheter in place especially in children.

These results showed that percutaneous transhepatic biliary interventions are effectively and safely performed in benign diseases of children causing biliary obstruction and/or cholangitis. These procedures can be performed safely either as a definitive treatment method or preoperatively as a bridge to surgery in benign biliary diseases of children.

**Conclusion**

In children having benign biliary obstruction with or without cholangitis, percutaneous intervention should be performed first, in order to prevent unnecessary surgery.

**References**


