



### Percutaneous Transhepatic Biliary Interventions in Benign Diseases of Children

Çocukluk Çağı Benign Hastalıklarında Perkütan Transhepatik Biliyer Girişimler

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#### ABSTRACT

**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.

**Materials and 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.

**Key words:** percutaneous, catheter, biliary, drainage, children, benign diseases

#### ÖZET

**Amaç:** Perkütan transhepatik biliyer girişimler erişkinlerde güvenli ve etkili bir biçimde uygulanmaktadır. Çocukluk çağı benign hastalıklarında kullanımı konusundaki deneyimler daha azdır. Çalışmamızda çocukluk çağı benign hastalıklarında yöntemin güvenilirliği ve etkinliği araştırılmıştır.

**Materyal ve Metod:** Bu retrospektif çalışmada, ortalama yaşı 10.2 (ranj 14 gün-14 yaş) olan 15 çocuğa perkütan biliyer girişim uygulanmıştır. Beş hasta sarılık, 10 hasta kolanjit yakınmaları ile başvurmuştur. Perkütan transhepatik biliyer drenaj (PTBD) 10 hastaya, PTBD ve balon dilatasyon 3 hastaya, perkütan kolesistomi (PK) 1 hastaya, PTBD yi takiben PK bir hastaya uygulanmıştır.

**Bulgular:** Uygulanan bütün işlemler teknik olarak başarıyla sonuçlandırılmıştır. İşleme bağlı mortalite olmamıştır. Oniki hastada serum bilirubin seviyeleri normal veya normale yakın seviyelere dönmüştür. İşlemden önce mevcut kolanjit ve akut kolesistit iyileşmiştir. Altı hastaya perkütan işlem sonrası cerrahi uygulanması gerekmiştir. Dokuz hasta ileri tedaviye ihtiyaç duyulmaksızın primer olarak perkütan girişimle iyileşmiştir.

**Sonuç:** Perkütan biliyer girişimler çocukluk çağı benign hastalıklarında etkili bir biçimde uygulanabilir. Primer tedavi yöntemi olarak veya cerrahi öncesi kullanılabilir. Birçok olguda perkütan tedavi yeterli olmuş ve gereksiz cerrahi önlenmiştir.

**Anahtar Kelimeler:** Perkütan, katater, biliyer, drenaj, çocuklar, benign hastalıklar

## 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<sup>1</sup>. 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<sup>2-4</sup>. 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 biliary strictures, or during insertion of metallic stents in order to relieve malignant strictures<sup>5</sup>. PC has been shown to be a safe treatment option for patients suffering from acute cholecystitis 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<sup>2,7</sup> and children with malignant obstructions<sup>8,9</sup>, there is less experience of percutaneous transhepatic biliary interventions in benign diseases of children<sup>10</sup>. 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 hepaticojejunostomy<sup>3</sup>, cholecystectomy<sup>1</sup> and operation for a gunshot wound<sup>1</sup>. Five children had choledochal cysts, and two had acute acalculous 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.

Choledochal cysts are classified according to Todani classification<sup>11</sup>.

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.

### Patient Preparation

Diagnostic imaging work up with abdominal US only (2 cases), US plus CT (4 cases) and US plus MRI (9 cases) were performed before percutaneous biliary interventions. Serum bilirubin levels, complete blood count, Prothrombin Time (PT), Partial Thromboplastin Time (PTT) and International Normalized Ratio (INR) were checked and any coagulopathy was corrected before the intervention. No prophylactic antibiotic was administered as a routine before the procedure, if the patient had no cholangitis. After 4-6 hours of fasting, procedure was performed in angiography suite under US and fluoroscopic guidance. Intravenous sedation and analgesia was performed by an anesthesiologist while patient is under cardiac and respiratory monitorization.

### 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 Accustick 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 catheter (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 large 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  $\mu\text{mol/L}$  (range 70-1368  $\mu\text{mol/L}$ ) 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  $\mu\text{mol/L}$  (range 17-147  $\mu\text{mol/L}$ ) 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 (calculous cholecystitis) improvement occurred 4 days after PC. But, in case 14 (acalculous cholecystitis), initially performed PC did not supply 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 occurred 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 catheter 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 (Caroli'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. Also, in case 11 (*Echinococcus granulosus* cyst communicated with the bile duct), case 14 (acute acalculous cholecystitis due to hemobilia) and case 15 (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) 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 calculous 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.

**Table 1. Clinical features of patients at presentation and the type of interventions performed**

Case No	Sex	Age	Diagnosis	Findings at Presentation		Percutaneous Intervention	Post-Interv. Serum Bilirubin	Surgery
				Symptom	Serum Bilirubin ( $\mu\text{mol/L}$ )			
1	M	12	Choledochal cyst type I	Jaundice, cholangitis	205	PTBD-E	37	Choledochal excision, HJ
2	F	14	Choledochal cyst type IV-A	Cholangitis	47	PTBD-E converted to PTBD-IE	17	Extended L hepatectomy, HJ
3	F	12	Choledochal cyst type V	Cholangitis	22	PTBD-E	17	-
4	F	3	Choledochal cyst type I	Jaundice, cholangitis	133	PTBD-IE	23	Choledochal excision, HJ
5	F	1.5	Choledochal cyst type I	Jaundice	145	PTBD-E	18	Choledochal excision, HJ
6	M	14	Postop. stricture	Jaundice, cholangitis	393	Balloon dilation and PTBD-IE	30	-
7	F	13	Postop. stricture	Cholangitis	29	Balloon dilation and PTBD-IE	17	-
8	M	12	Postop. stricture	Jaundice, cholangitis	73	PBD-IE	17	-
9	F	11	Postop. stricture	Jaundice, cholangitis	119	PTBD-IE	17	-
10	F	14	Postop. stricture	Jaundice	71	Balloon dilation and PTBD-IE	20	-
11	M	13	Hydatid cyst rupture	Jaundice	70	PTBD-IE	17	-
12	M	12	<i>E.alveolaris</i> invasion	Jaundice	632	PTBD-E converted to PTBD-IE	147	Extended L hepatectomy, HJ
13	M	12	Acute calculous cholecystitis	RUQ pain jaundice, fever	1368	PC	109	Cholecystectomy
14	M	9	Acute acalculous cholecystitis	RUQ pain jaundice, fever	123	PC and PTBD-IE	20	-
15	M	1/12	Sludge ball	Jaundice	88	PTBD-IE	17	-

RUQ: Right Upper Quadrant

PTBD-E: Percutaneous Transhepatic Biliary Drainage-External

PTBD-IE: Percutaneous Transhepatic Biliary Drainage-Internal/External

PC: Percutaneous Cholecystostomy

HJ: Hepaticojejunostomy

## 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<sup>12,13</sup>. 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<sup>14</sup>. In this study, 4 of 5 choledochal cysts gone 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<sup>10,15</sup>, 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)<sup>16</sup>, 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<sup>17</sup>, 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.

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<sup>18</sup>. In this case, total serum bilirubin level was 632  $\mu\text{mol/L}$  at presentation. After 14 days of PTBD-IE performed preoperatively, total serum bilirubin level decreased to 147  $\mu\text{mol/L}$ . 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  $\mu\text{mol/L}$  and 1026  $\mu\text{mol/L}$ , 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<sup>19</sup>. 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.

### 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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