

# Biliary duodenostomy: a safe and easier biliary drainage procedure after choledochal cyst excision

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**Background/purpose** The treatment of choice for choledochal cyst (CC) is complete excision followed by biliary–enteric anastomosis. Roux-en-Y biliary jejunostomy has been favored by most surgeons for decades, with satisfying results. The use of biliary duodenostomy (BD) is another simple alternative for biliary drainage after CC excision. Our intermediate-term outcomes of both biliary drainage procedures after CC excision are presented.

**Methods** We carried out a retrospective analysis of the outcome of CC management in children operated at the Pediatric Surgery Department of Ain-Shams University over 5 years, from January 2010.

**Results** A total of 23 cases (16 females) were included in this study. The mean age at operation was  $4.02 \pm 2.52$  years. Twenty-one cases had type I and two cases had type III CC. Complete excision of the CC was performed in 22 cases, and partial excision with mucosectomy was performed in one case. Biliary–enteric anastomosis was performed with the duodenum in

18 patients (group I, BD) and with the jejunum in five cases (group II, Roux-en-Y biliary jejunostomy). The mean follow-up period was  $53 \pm 4.48$  months. A patient from group II suffered from ascending cholangitis 6 months postoperatively.

**Conclusion** BD is a simple technique for biliary drainage after CC excision with no major complications at intermediate-term follow-up. *Ann Pediatr Surg* 13:129–132 © 2017 Annals of Pediatric Surgery.

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**Keywords:** biliary obstruction, choledochal cyst, hepaticoduodenostomy, jaundice

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## Introduction

The treatment of choice for choledochal cyst (CC) is complete excision of the cyst followed by biliary–enteric anastomosis [1]. This would separate the biliary tree from the pancreatic duct, inhibiting the mixing of pancreatic and biliary secretions responsible for the pathogenesis of the disease, and decrease the risk of malignant transformation [2].

Biliary drainage by Roux-en-Y biliary jejunostomy (RYBJ) after CC excision has been favored by many surgeons for decades, with satisfying long-term results [3,4]. The use of biliary duodenostomy (BD) is another simple alternative for biliary drainage after CC excision. However, some authors have raised concerns about the higher rates of ascending cholangitis and biliary gastritis after BD reconstruction [5].

This study was conducted to compare the outcome of CC management with two different methods of biliary reconstructions after CC excision in children operated at a tertiary pediatric surgery center over 5 years.

## Methods

After obtaining approval of the institutional review board of Ain-Shams University hospitals in June 2015 (numbered: 23-8-5-2015), a retrospective study was initiated, including all cases below 18 years of age with a diagnosis of CC, from January 2010 to January 2015. Patients were excluded from the study if there was any associated congenital anomaly or any associated hepatobiliary condition, other than CC, and cases with inoperable CC.

For all patients, data on demographics, diagnostic modalities used, CC type according to the Todani modification of the Alonso–Lej classification [3], surgical techniques used for correction, immediate and delayed postoperative complications, and follow-up data (clinical, laboratory, and radiological) of at least 1 year were retrieved.

The procedure was carried out under general anesthesia with the patient lying in the supine position with a tilt to the right and head elevation of the operating table. Open or laparoscopic techniques were used according to the surgeon's preference.

For laparoscopy, five ports (5 mm) were used – one in the umbilicus for inserting the 30° scope and four working ports: one in the anterior axillary line for traction on the gallbladder, the second in the midline below the xiphisternum for liver retraction, and two working ports (one in the mid-clavicular line and the other in the midline between the umbilicus and the xiphisternum) [6]. A right transverse supraumbilical incision was used for open cases.

After intraoperative cholangiography through the gallbladder, complete excision of the CC was performed if possible. If not, partial excision with mucosectomy was used. BD was performed through a wide end-to-side anastomosis, beyond the first part of the duodenum and away from the pylorus, using interrupted long-term absorbable sutures. RYBJ was performed 30–40 cm after the duodenojejunal junction, the Roux loop was passed through a retrocolic window, and anastomosed end to end using long-term, absorbable, interrupted sutures. For type

III CC, dissection of the lower part of the common bile duct down to the level of the pancreatic head (avoiding the pancreatic ducts), followed by marsupialization of the CC through a transduodenal incision, excision of the CC, closure of the duodenum, and a biliary drainage procedure was carried out.

All patients remained in the high-dependency unit for 48 h, and discharged from the hospital after full tolerance for oral intake. As per the hospital policy, no postoperative long-term antibiotic prophylaxis against cholangitis was used. Liver enzymes and bilirubin levels were routinely measured after 1 month since the operation, and follow-up abdominal ultrasonography was carried out after 3 months.

Patients were divided according to the methods of biliary drainage after CC excision into the following: group I with BD and group II with RYBJ. Statistical package for social science, version 20 (IBM SPSS) was used for analysis of the results. Comparison between the two groups with qualitative data was carried out using  $\chi^2$ -test. Comparison between two groups with quantitative data was carried out using independent *t*-test. The confidence interval was set at 95%, and the margin of error accepted was set at 5%. Therefore, *P* value more than 0.05 was considered nonsignificant; *P* value less than 0.05 was considered significant, and *P* value less than 0.01 was considered highly significant.

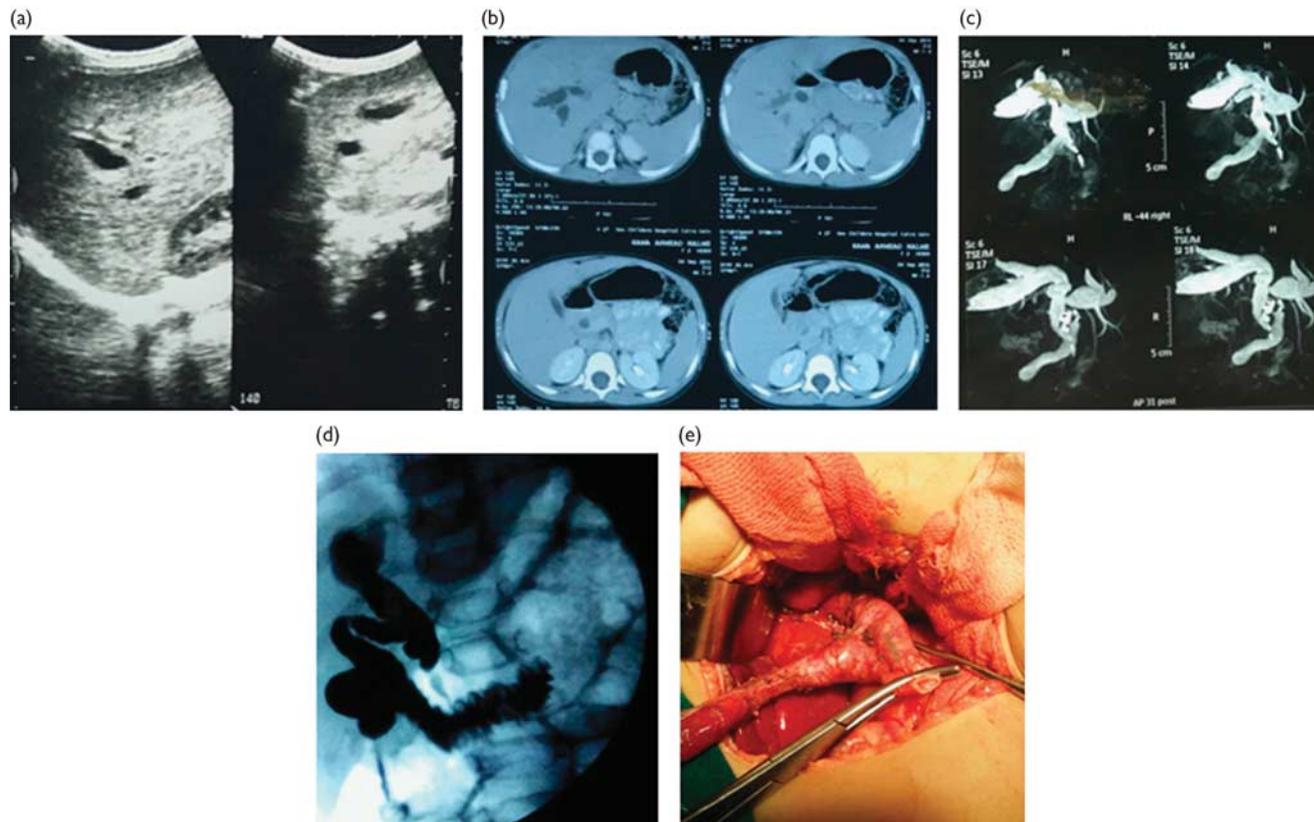
## Results

During the mentioned period, 28 cases with a diagnosis of CC were retrieved, and five were excluded: three had inoperable Caroli disease (listed for liver transplantation), one had associated duodenal duplication, and one case was falsely diagnosed as having CC, and upon exploration fasciola hepaticum infestation was found as the cause for biliary dilatation. Finally, 23 cases (16 females) were included for the study. One of them had a family history of a sibling with biliary atresia. The mean age at operation was  $4.02 \pm 2.52$  years, ranging from 0.2 to 7.5 years, with peak age of 5–6 years (35% of cases).

Obstructive jaundice was the most common presenting symptom, present alone in about 50% of the cases. The second most common symptom was right hypochondrial pain, present in about 20% of the cases. Three patients (15% of the cases) presented with the triad of jaundice, abdominal pain, and swelling. Only one patient presented with isolated abdominal swelling, without jaundice or pain. Two cases were accidentally discovered during ultrasonography, which was performed for other unrelated causes.

Ultrasonography was highly reliable for diagnosis in five (21%) cases (Fig. 1a). In 11 (48%) cases, computed tomography (CT) with intravenous contrast was used (Fig. 1b), and in 17 (74%) cases magnetic resonance cholangiopancreatography (MRCP) was used to confirm the diagnosis (Fig. 1c).

Fig. 1



A 2.5-year-old girl with CC: (a) ultrasonography showing a dilated common bile duct at the porta hepatis; (b) computed tomographic scan showing the dilated biliary radicals; (c) magnetic resonance cholangiopancreatography: dilatation reached the hepatic ducts; (d) intraoperative cholangiography showing type 1 choledochal cyst; (e) operative view showing the dilated common bile and cystic ducts before proceeding for proximal dissection.

Laparoscopy was performed for three cases (with conversion in one case due to difficulty in completing BD), and laparotomy was performed in 20 cases. After intraoperative cholangiography through the gallbladder (Fig. 1d), 21 cases were shown to have type I (fusiform) CC and two cases had type III CC (choledochocoele at the biliopancreatic junction, 'intraduodenal diverticulum') (Fig. 1e). Complete excision of the CC was possible in 22 cases, and partial excision with mucosectomy was performed in only one case because of extensive adhesions. Biliary drainage anastomoses following CC excision were performed with the duodenum in 18 patients (group I, BD) and with the jejunum in five cases (group II, RYBJ).

The mean operative time for group I was  $104 \pm 13$  min and  $119 \pm 16$  min for group II. The mean time to achieve complete oral intake was  $3.95 \pm 0.76$  days (range: 3–5 days), and the duration of hospital stay ranged from 5 to 11 days. Four patients [three after choledochoduodenostomy (group I) and one after choledochojejunostomy (group II)] suffered from superficial wound infection.

All patients had normal liver enzymes and total and direct bilirubin within 1 month after the operation. At the 3 month follow-up, abdominal ultrasonography was carried out for all patients. The mean follow-up period was  $53 \pm 4.48$  months. Only one patient from group II (RYBJ) had ascending cholangitis 6 months after surgery. Ultrasonographic examination of this patient showed no residual dilatation of the biliary system, and she was managed medically. Contrast study was planned if recurrent cholangitis occurred (followed up for 39 months after this cholangitic attack). None of our cases had anastomotic leakage, adhesive intestinal obstruction, symptoms suggestive of biliary reflux, or required reoperation.

Statistical analysis showed that the two groups were comparable with regard to age at operation and sex distribution (Table 1). No statistically significant difference was noted between the two groups with regard to the day of return to complete oral intake, postoperative wound infection, and postoperative cholangitis (Table 2).

## Discussion

Only a few reports have compared hepatojejunostomy with choledoduodenostomy after CC excision in a single institution [4], and most of these reports are from the Far East with limited data available from the Middle East area.

We reported our experience with pediatric CC in a tertiary center over a 5-year period. Jaundice was the most common symptom in our study groups; this is in agreement with

other studies on pediatric CC [7,8]. The triad of abdominal pain, jaundice, and palpable abdominal mass was recognized in three of our patients. In the present study, about 10% of cases were accidentally discovered. Dhupar *et al.* [8] found that more than 36% of patients were accidentally discovered either during a laparoscopic operation or after a CT scan for an unrelated problem.

If the patient is diagnosed with CC while acutely ill with active cholangitis or pancreatitis, external drainage should be performed before surgery [9]. However, surgery should not be delayed for long, as a greater potential for cyst rupture is present [10]. Most pediatric surgeons advocate cyst excision for prenatally diagnosed CCs, even before the onset of symptoms [11].

Ultrasonography was the test of choice for the diagnosis of CC. In addition, ultrasonography is a useful tool for follow-up and assessment of any residual biliary dilatation following CC excision [4,12]. However, if the diagnosis of CC is unclear, MRCP is now considered the gold standard for imaging [3]. All our patients underwent initial ultrasonography, with further confirmatory imaging required in 18 patients (11 CT scan, 17 MRCP).

After CC excision and biliary–enteric anastomosis, symptomatic improvement was observed in all patients. The same was observed by Gananadha *et al.* [13] as intrahepatic duct dilatations decompress and hepatic fibrosis and varices regress.

In the present study, with a mean follow-up period of  $53 \pm 4.48$  months, no complications were detected in 18 cases. Wound infection occurred in three cases from group I (BD), and in one case from group II (RYHJ). Cholangitis was detected in one case (6 months after RYHJ), and was managed medically. Cholangitis is the most common complication after CC excision [14,15]. Cholangitis or pancreatitis could result from duct stenosis or obstruction or protein plugs. Although long-term antibiotics have been reported to minimize the incidence of cholangitis [15], none of our cases were on antibiotic prophylaxis.

None of our patients developed symptoms suggestive of biliary reflux, and therefore no further investigations were carried out, such as contrast study or endoscopy, as it is often difficult to convince parents to carry out such studies if their child is asymptomatic [16]. It was believed that the reconstruction of the bile exit near the pyloric ring may interrupt continuous excretion of bile into the duodenum [17]. Therefore, an important step in DB reconstruction is to perform the anastomosis beyond the first part of the duodenum, after a good Kocher maneuver, without any tension to avoid any impact on pyloric function or gastric emptying [18].

A meta-analysis in 2013 comparing BD with RYBJ, in a total of 679 patients [among them, 412 (60.7%) underwent BD, and 267 (39.3%) had RYBJ], suggested that BD is comparable with conventional RYBJ with respect to most postoperative outcomes except for a higher rate of gastric reflux [follow-up was variable (3–204 months)] [18]. Our results showed no statistical significant difference in the outcome of both techniques of

**Table 1** Showing that both groups' demographic data were comparable

	Group I No. = 18	Group II No. = 5	Chi-square test	
			$\chi^2/t$	P-value
Sex				
Female	16 (80.0%)	4 (80.0%)	0.317	0.573
Male	2 (20.0%)	1 (20.0%)		
Age at operation				
Mean $\pm$ SD	$3.00 \pm 2.18$	$4.35 \pm 2.60$	-1.043	0.311
Range	1–6.5	0.2–7.5		

Table 2 Showing outcome analysis of the 2 groups

	Group I No.=18	Group II No.=5	Independent t-test		OR (95% CI)
			t/X <sup>2</sup>	P-value	
Day of full feeding					
Mean ± SD	4.13 ± 0.74	3.40 ± 0.55	-2.016	0.059	9.29 (0.84–102.77)
Range	3–5	3–4			
Wound infection					
No	15 (83.3%)	4 (80.0%)	0.000	1.000*	0.596 (0.031–11.45)
Yes	3 (16.6%)	1 (20.0%)			
Ascending Cholangitis					
No	18 (100.0%)	4 (80.0%)	3.158	0.076*	0.000 (0.000–1.000)
Yes	0 (0.0%)	1 (20.0%)			

biliary–enteric anastomosis with a mean of  $53 \pm 4.48$  months.

In light of previous data, we are in favor of BD reconstruction if feasible for the following reasons. BD is a physiological procedure compared with RYBJ; it is simpler to perform; and there are less possibilities of complications such as adhesive bowel obstruction, anastomotic leakage, and entails a shorter surgical procedure. In contrast, RYBJ requires the creation of a Roux-en-Y jejunal limb with two anastomoses [19]. In addition, regarding laparoscopic intervention, it is better and easier to perform BD rather than RYHJ [20]. The occurrence of postoperative cholangitis is highly related to the presence of anastomotic or supra-anastomotic strictures [19], which is more dependent on the surgeon's experience rather than the technique used for biliary–enteric anastomosis. Some surgeons preferred a hilar anastomosis at the bifurcation of hepatic ducts rather than a distal anastomosis in type 1 cysts to ensure wider anastomosis [19]. Surveillance or intervention later on are easier with BD (no Roux loop as in RYBJ) if a stricture or stone occurs during contrast studies or endoscopies [17,18]. There are higher incidences of both intrahepatic and extrahepatic stone formation in the elongated pouch of RYBJ, and the duodenal obstruction caused by compression by a high RYBJ vascular arch [16].

However, conventional hepatobiliary surgeons still prefer the Roux-en-Y reconstruction because of the following issues: RYBJ has a very long history of safety and is the gold standard for biliary reconstruction [18]; contrary of our findings, some authors have reported high incidences of endoscopy-proven bilious gastritis due to duodenogastric bile reflux, adhesive bowel obstruction, and cholangitis in the BD group [5]; and Todani *et al.* [21] reported a case with hilar bile duct carcinoma 19 years after CC excision and BD. They thought that reflux of duodenal contents (including activated pancreatic enzymes) into the intrahepatic bile ducts through the BD anastomosis may be hazardous to the bile duct mucosa [21]. However, the development of cholangiocarcinoma after excision of CC requires a longer-term follow-up before justifying any association with the BD type of biliary–enteric reconstruction [17].

Our study had the inherent limitations of a retrospective study and a small sample size, therefore, further studies should be carried out to detect the long term complications of biliary–enteric anastomosis, especially the possible malignant transformation.

## Conflicts of interest

There are no conflicts of interest.

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