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# Management of complicated choledochal cyst in children: ultrasound-guided percutaneous external drainage and subsequent definitive operation

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## Abstract

**Objective** The purpose of this study was to analyze the outcomes of the combination of ultrasound (US)-guided percutaneous external drainage and subsequent definitive operation to manage complicated choledochal cyst in children.

**Methods** This retrospective study included 6 children with choledochal cyst who underwent initial US-guided percutaneous external drainage and subsequent cyst excision with Roux-en-Y hepaticojejunostomy between January 2021 and September 2022. Patient characteristics, laboratory findings, imaging data, treatment details, and postoperative outcomes were evaluated.

**Results** Mean age at presentation was  $2.7 \pm 2.2$  (0.5–6.2) years, and 2 patients (2/6) were boys. Four patients (4/6) had a giant choledochal cyst with the widest diameter of  $\geq 10$  cm and underwent US-guided percutaneous biliary drainage on admission or after conservative treatments. The other 2 patients (2/6) underwent US-guided percutaneous transhepatic cholangio-drainage and percutaneous transhepatic gallbladder drainage due to coagulopathy, respectively. Five patients (5/6) recovered well after US-guided percutaneous external drainage and underwent the definitive operation, whereas 1 patient (1/6) had liver fibrosis confirmed by Fibroskan and ultimately underwent liver transplantation 2 months after external drainage. The mean time from US-guided percutaneous external drainage to the definitive operation was  $12 \pm 9$  (3–21) days. The average length of hospital stay was  $24 \pm 9$  (16–31) days. No related complications of US-guided percutaneous external drainage occurred during hospitalization. At  $10.2 \pm 6.8$  (1.0–18.0) months follow-up, all patients had a normal liver function and US examination.

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**Conclusions** Our detailed analysis of this small cohort suggests that US-guided percutaneous external drainage is technically feasible for choledochal cyst with giant cysts or coagulopathy in children, which may provide suitable conditions for subsequent definitive operation with a good prognosis.

**Trial registration** Retrospectively registered.

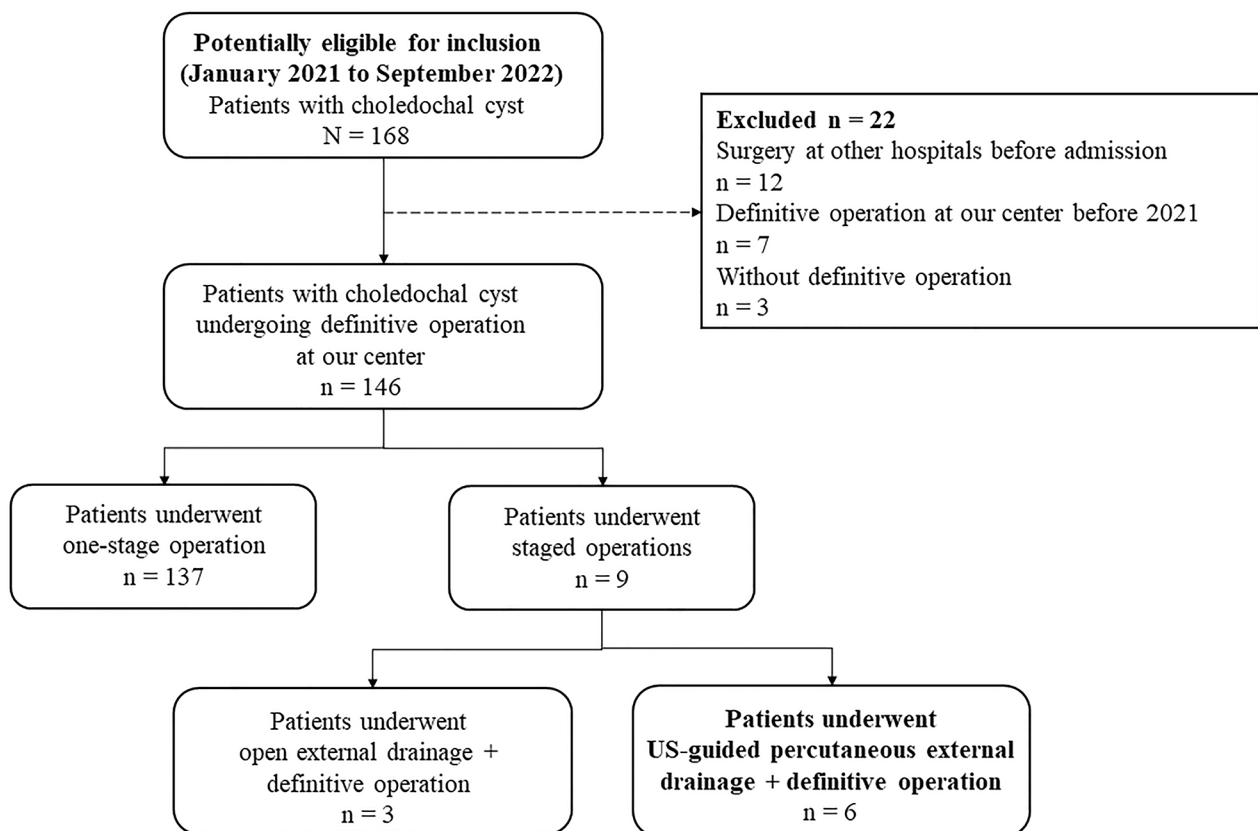
**Keywords** Choledochal cyst, Ultrasound, Percutaneous external drainage, Definitive operation

## Background

Choledochal cyst, a relatively uncommon congenital malformation characterized by biliary tract dilatation with an incidence of around 1:13,000 in Asian populations versus 1:100,000 in Western populations, often presents with abdominal pain, abdominal lump, and jaundice in pediatric patients [1–3]. The currently acknowledged etiology of choledochal cyst is the anomalous pancreaticobiliary junction with reflux of pancreatic juice into the common bile duct and the formation of protein plugs or calculi in the bile duct [4, 5]. Given that patients with choledochal cyst have a high risk of hepatobiliary complications, such as cholangitis, pancreatitis, perforation of the cyst, and potential malignant change, prompt treatment is required [1, 6, 7]. Some studies have reported that even if patients are asymptomatic, surgical intervention as soon

as possible can lead to a better outcome, such as a significantly reduced rate of liver fibrosis in patients [3, 8]. The specific approach is largely a function of the type of cyst and the preferred treatment is total cyst excision with Roux-en-Y hepaticojejunostomy [8, 9]. However, surgical intervention should be elective, some related concomitant complications, including cholangitis, pancreatitis, and coagulopathy, may require conservative treatments, delay the surgical treatments, and become the major risk for operation [10–12]. How to address complicated choledochal cyst has always been challenging in clinical management.

Patients with complicated choledochal cyst often had a distal biliary obstruction that led to bile stasis and ultimately developed severe complications, such as acute

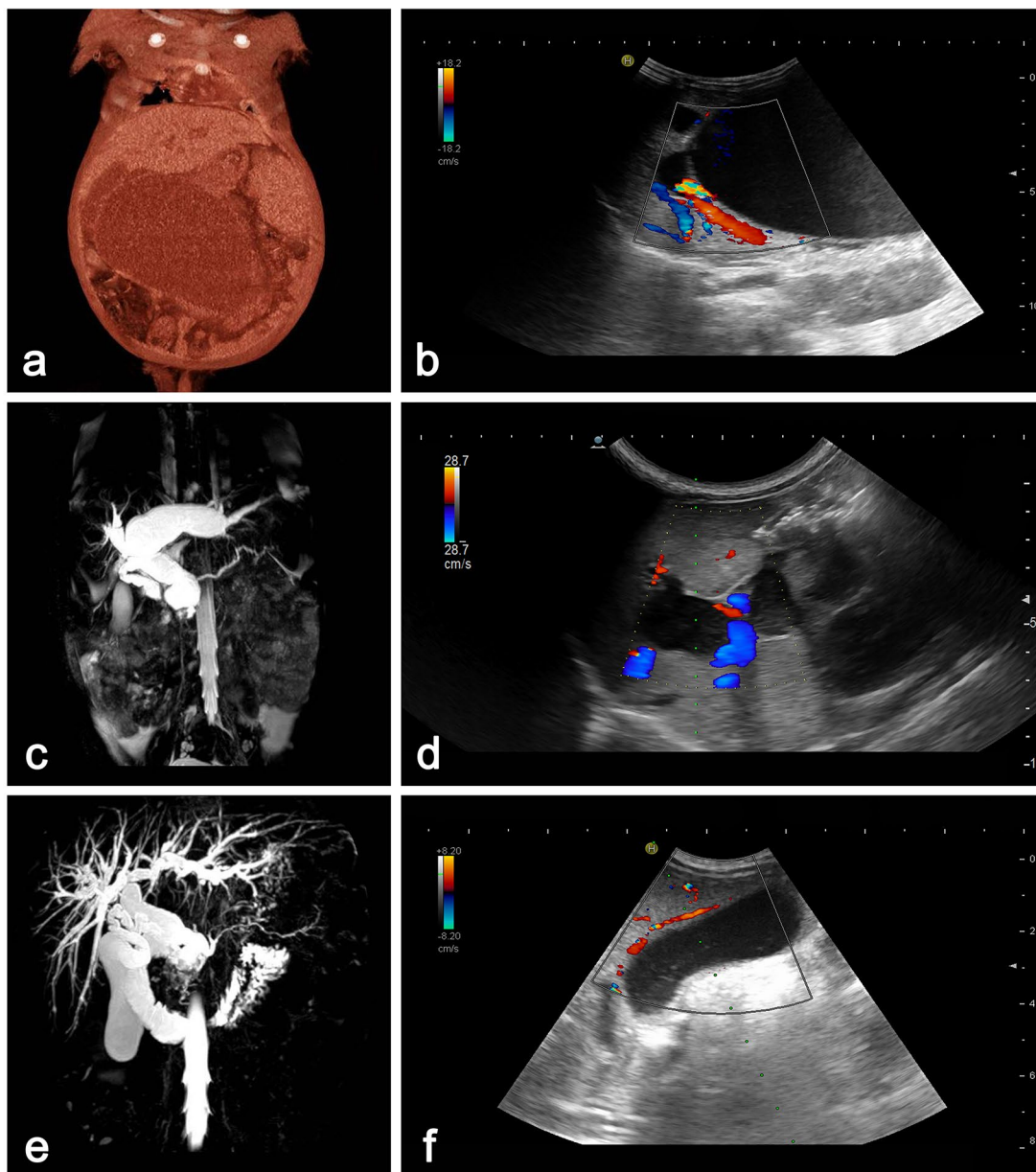


**Fig. 1** Flow diagram of the study population

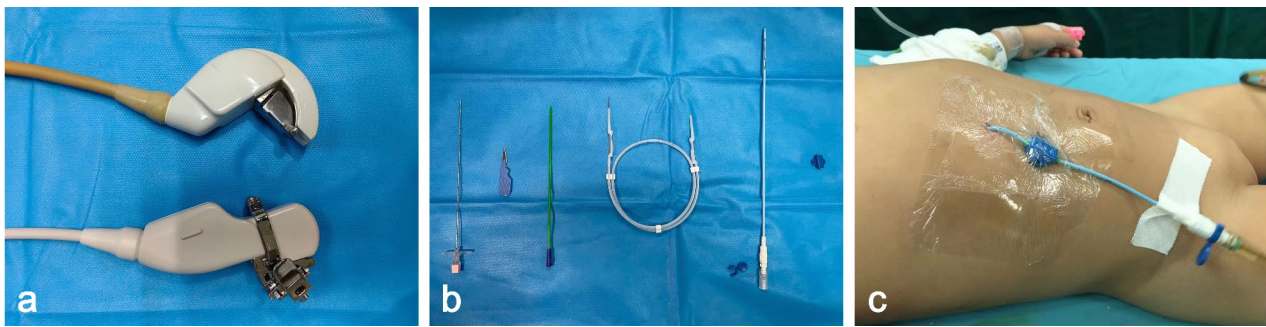
cholangitis, abnormal liver function, and even perforation of choledochal cysts and coagulopathy [11–13]. These patients commonly required two-stage operations, urgent external biliary drainage, and subsequent definitive operation [12, 14, 15]. The methods of external drainage mainly included biliary drainage of extrahepatic cyst or intrahepatic bile duct, and gallbladder drainage (cholecystostomy) [11, 15–17]. In most pediatric cases, traditional external drainage was accomplished by laparotomy or laparoscopic-assisted [11, 12, 18].

Percutaneous external drainage of choledochal cyst in children has been rarely reported in the literature, and ultrasound (US)-guided percutaneous external drainage is even rarer [11, 15].

This study aimed to report the early clinical experience of the combination of US-guided percutaneous external drainage and subsequent definitive operation to manage choledochal cyst in children in a small cohort and evaluate its safety and effect.



**Fig. 2** CT or MRCP, and US results of patients with choledochal cyst before and during US-guided percutaneous external drainage. **(A)** CT before US-guided PBD, Case 1 with a giant choledochal cyst. **(B)** US-guided PBD, Case 1 with a giant choledochal cyst. **(C)** MRCP before US-guided PTCD, Case 5 with significant dilatation of the intrahepatic bile duct. **(D)** US-guided PTCD, Case 5 with significant dilatation of the intrahepatic bile duct. **(E)** MRCP before US-guided PTGD, Case 6 with a large gallbladder. **(F)** US-guided PTGD, Case 6 with a large gallbladder



**Fig. 3** Operating instruments in US-guided percutaneous external drainage. **(A)** Ultrasound probes. **(B)** Mini-invasive kits of puncture, including puncture needle, sharp knife, tract dilator, guide wire, Pig-tail catheter, and external fixator. **(C)** The method of external fixation

**Table 1** Patients and characteristics

Cases	Sex	Age (years)	Types	Size (cm <sup>3</sup> )	Preoperative complications	Main indication for external drainage	Procedures	Definitive procedures*	Follow-up (months)
1	M	1.4	Iva	15.2*12.6*9.4	Coagulopathy, abnormal liver function, liver fibrosis	Giant cyst, coagulopathy	US-guided PBD, local anesthesia	Liver transplantation	18.0
2	F	0.5	Ia	20.0*11.2*8.2	Abnormal liver function	Giant cyst	US-guided PBD, general anesthesia	Open	16.0
3	F	6.2	Ia	13.1*5.3*4.9	Abnormal liver function	Giant cyst	US-guided PBD, general anesthesia	Robotic	8.5
4	M	0.9	Iva	10.5*6.6*4.7	Coagulopathy	Giant cyst, coagulopathy	US-guided PBD, general anesthesia	laparoscopic	4.1
5	F	3.2	Iva	5.0*2.1*2.1	Coagulopathy, abnormal liver function, acute pancreatitis	Coagulopathy	US-guided PTCD, general anesthesia	laparoscopic	13.6
6	F	3.8	Ic	1.3**	Coagulopathy, abnormal liver function, acute pancreatitis	Coagulopathy	US-guided PTGD, local anesthesia	laparoscopic	1.0

M: Male; F: Female; US: Ultrasound; PBD: Percutaneous biliary drainage; PTCD: Percutaneous transhepatic cholangio-drainage; PTGD: Percutaneous transhepatic gallbladder drainage

\* Except for liver transplantation, the procedure performed for patients with choledochal cyst in our department was cyst excision with Roux-en-Y hepaticojejunostomy

\*\* The maximum diameter of the cyst in Ic type of choledochal cyst

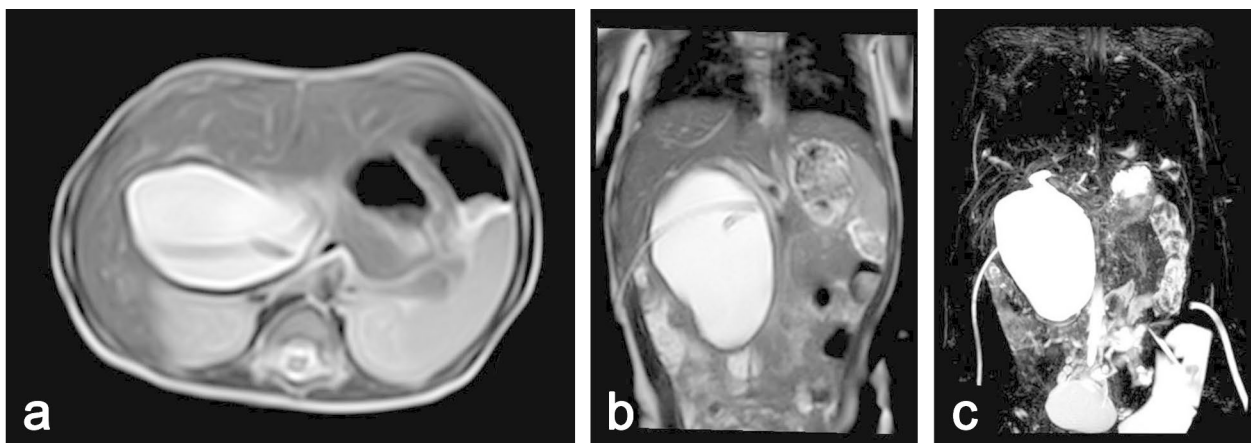
## Methods

### Patients

This retrospective study was approved by the Ethics Committee of Beijing Children's Hospital (Approval number: 2022-E-214-R) and the need for informed consent was waived. The medical records of patients diagnosed with choledochal cyst and admitted to the department of general surgery, Beijing Children's Hospital, from January 2021 to September 2022, were reviewed. The diagnosis of choledochal cyst was based on abdominal US with computed tomography (CT) or magnetic resonance cholangiopancreatography (MRCP). Patients who underwent US-guided percutaneous external drainage and subsequent definitive operation (cyst excision with Roux-en-Y hepaticojejunostomy) were included in this study. The exclusion criteria were as follows: (I) underwent surgery at other hospitals before admission; (II) underwent the definitive operation at our center before 2021; (III) no definitive operation; (IV) underwent one-stage definitive operation; and (V) underwent traditional open external drainage and definitive operation (Fig. 1).

### Clinical characteristics

Two pediatric surgeons reviewed the electronic medical records (JYY and MKZ). Patient characteristics, laboratory findings, imaging data, treatment details, and postoperative outcomes were analyzed. Patient characteristics included sex, age, presenting symptoms, features of the choledochal cyst (types and size), and preoperative hepatobiliary complications. The types of choledochal cyst were defined using Todani's classification, and the choledochal cyst with a maximum diameter of  $\geq 10$  cm was defined as a giant cyst [19, 20]. The laboratory findings included the laboratory results before US-guided percutaneous external drainage and before the definitive operation, covering blood routine, coagulation biomarkers, liver function tests and indicators of pancreatitis. The normal reference intervals for laboratory results in our study were based on the PRINCE study [21]. Fibrinogen (FIB) level  $< 2$  g/L with prolonged prothrombin time (PT) or activated partial thromboplastin time (APTT) indicated coagulopathy [12, 22]. The value of aspartate



**Fig. 4** Images of biliary drain after US-guided PBD. **(A)** Axial T2-weighted MR image after US-guided PBD, Case 2. **(B)** Coronal T2-weighted MR image after US-guided PBD, Case 2. **(C)** MRCP image after US-guided PBD, Case 2

aminotransferase (AST) and alanine aminotransferase (ALT) > 45 U/L with  $\gamma$ -glutamyl transpeptidase ( $\gamma$ -GGT) > 70 U/L indicated abnormal liver function [23]. Acute pancreatitis was characterized by abdominal pain and elevated serum amylase (AMY) and lipase (LPS) levels (> 2 times the upper limit of reference interval) [24]. In addition, for patients with features of liver parenchymal disease indicated by US, the degree of liver fibrosis would be evaluated by Fibroscan, and the value of liver stiffness measurement > 15.15 kPa indicated liver fibrosis in patients [20, 25].

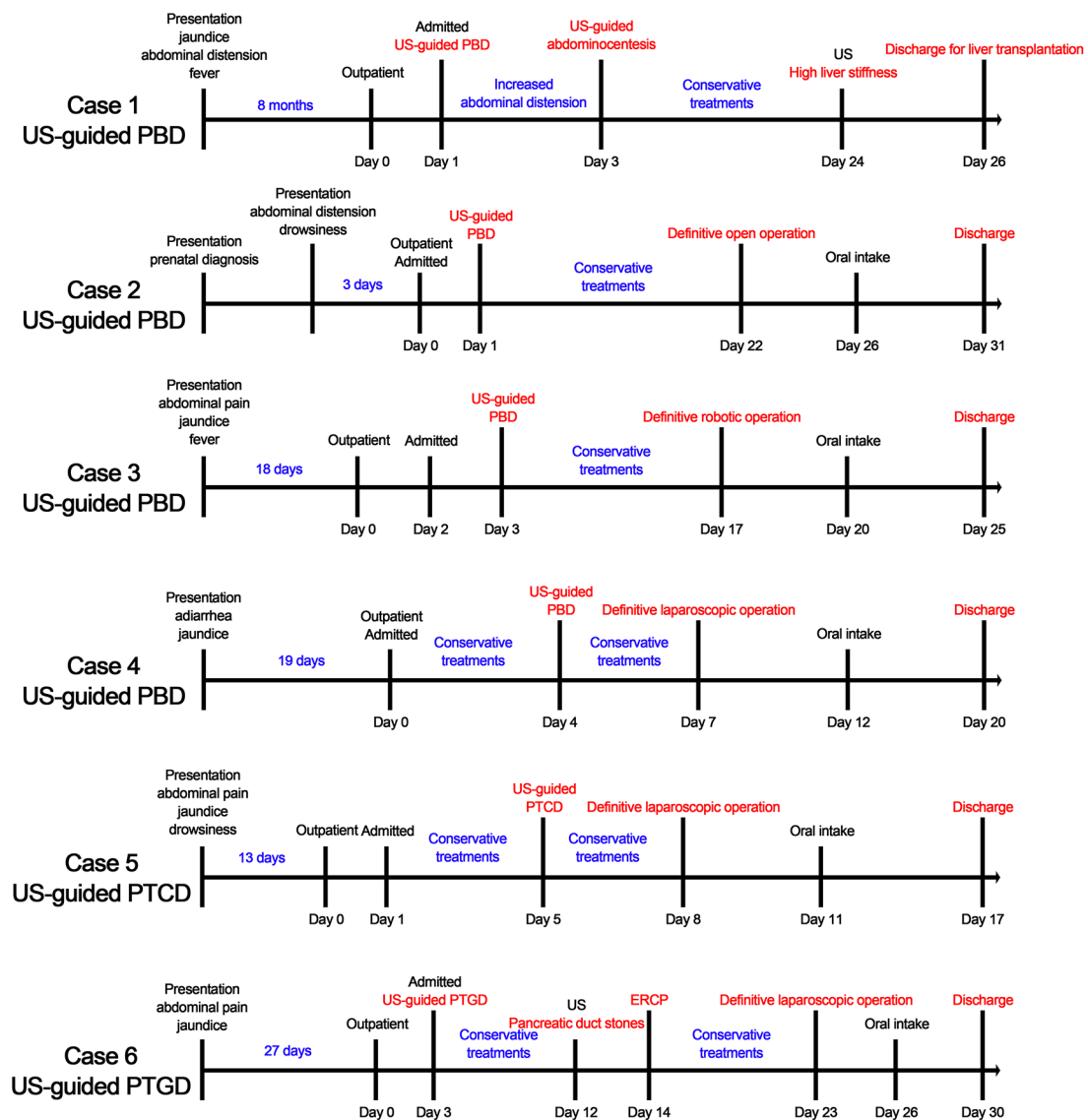
The imaging data included the images of US, CT, and MRCP performed at our center, including images during US-guided percutaneous external drainage. The treatment details had three aspects: conservative, interventional, and surgical treatments. The conservative treatments of each patient were based on the laboratory results on admission, mainly including nil-per-os, somatostatin injected with a microinfusion pump (3.5  $\mu$ g/kg/h), intravenous omeprazole (1–2 mg/kg), antibiotic treatment, and total parenteral nutrition. Telephone interviews were performed in October 2022 to ask the patients' parents about the child's prognosis and postoperative complications.

#### Ultrasound-guided percutaneous external drainage

The decision to perform external drainage was based on the patients' laboratory and imaging results (CT or MRCP). If a patient had a giant cyst with abnormal liver function or especially coagulopathy and did not improve after conservative treatments, US-guided percutaneous external drainage would be considered. Every patient fasted for a minimum of 6 h and was re-evaluated by US to determine the extent of intrahepatic bile duct dilation, the size of extrahepatic cyst and gallbladder to help select the specific methods of external drainage. The US-guided

percutaneous external drainage was performed by a pediatric radiologist (CKL) with more than 5 years of experience in interventional US techniques, after obtaining informed consent from the parents of patients. The preferred method was US-guided percutaneous biliary drainage (PBD), especially for patients with a giant extrahepatic cyst. In addition, US-guided percutaneous transhepatic cholangio-drainage (PTCD) was performed in patients with a small extrahepatic cyst, but significant dilatation of the intrahepatic bile duct, and US-guided percutaneous transhepatic gallbladder drainage (PTGD) was performed in patients with a large gallbladder (Fig. 2).

All procedures are accomplished under general anesthesia by anesthesiologists or local anesthesia with the support of oral chloral hydrate as a sedating agent by continuous close monitoring, including the preparation of emergency medications and oxygen therapy devices. At the supine position, the right upper quadrant of patients was draped in a sterile manner. Lidocaine (2%) was used as a local anesthetic. First, after the puncture point was determined by ultrasound and the skin was cut through by a sharp scalpel, the right position of the puncture needle was ensured by real-time US guidance and introduced guide wire. This process should be far away from the abdominal organs and blood vessels as possible. Second, after the puncture, tract dilation was done under real-time US guidance. Finally, a Pig-tail catheter (6–8 Fr) was inserted along the introduced guide wire to decompress the biliary system, and the external fixation to the skin was made (Fig. 3). In US-guided PBD, the point at the anterior axillary line with the smallest distance between the cyst and abdominal wall was selected as the puncture point. In US-guided PTCD and US-guided PTGD, the puncture needle was passed through the liver to penetrate the target intrahepatic bile duct or gallbladder without touching the intrahepatic blood vessels.



**Fig. 5** Clinical diagnosis and treatment timelines of patients with choledochal cyst who underwent US-guided percutaneous external drainage and subsequent definitive operation

After external drainage, conservative treatments were performed, including intravenous broad-spectrum antibiotics and hemostatic agents, and patients resumed oral intake as soon as possible to avoid electrolyte disturbances. Laboratory tests were performed on the 1st, 3rd, and 7th day after external drainage to check the improvement of the liver and coagulation function. Once needed, patients were re-examined every 7 days until the liver and coagulation function was appropriate for definite surgery. Usually in our center, CT or MRCP would be repeated prior to the definitive operation (Fig. 4).

**Data analysis**

Continuous variables were expressed as the mean and SD with ranges. Categorical variables were expressed as numbers and proportions.

**Results**

During the study period, 6 patients with choledochal cyst were treated by a combination of US-guided percutaneous external drainage and subsequent definitive operation, and their clinical characteristics were provided in Table 1. The mean age at presentation was  $2.7 \pm 2.2$  (0.5–6.2) years, and 2 patients (2/6) were boys. Their most common presenting symptoms were jaundice (5/6) and abdominal pain (3/6). Except for case 1, other patients

**Table 2** Results of laboratory tests in 6 patients\*

Case	CRP (mg/L)	WBC (10 <sup>9</sup> /L)	FIB (g/L)	PT (S)	APTT (S)	NH <sub>3</sub> (umol/L)	AST (U/L)	ALT (U/L)	γ-GGT (U/L)	DBIL (umol/L)	IBIL (umol/L)	AMY (U/L)	LPS (U/L)
Reference interval	<10.0	4.4–11.9	2.0–4.0	9.4–2.5	25.1–38.4	18.0–72.0	14.0–44.0	7.0–30.0	5.0–19.0	0.0–3.42	0.0–17.1	0.0–125.0	0.0–39.0
<b>Case 1</b>													
Before external drainage	<b>15.0</b>	9.9	<b>1.9</b>	<b>17.9</b>	34.8	<b>115.0</b>	<b>250.6</b>	<b>141.7</b>	<b>245.8</b>	<b>144.7</b>	<b>49.6</b>	81.0	<b>144.4</b>
Pre-operation**	<10.0	5.5	<b>1.7</b>	<b>17.0</b>	37.1	<b>80.0</b>	<b>181.4</b>	<b>83.0</b>	<b>90.4</b>	<b>45.1</b>	<b>25.6</b>	11.0	6.0
<b>Case 2</b>													
Before external drainage	<10.0	11.0	3.3	<b>18.7</b>	34.7	—	<b>354.5</b>	<b>278.1</b>	<b>1751.9</b>	<b>40.1</b>	12.4	31.0	<b>206.8</b>
Pre-operation	<10.0	7.16	—	—	—	—	40.1	27.2	<b>336.2</b>	2.8	9.6	—	—
<b>Case 3</b>													
Before external drainage	<10.0	9.0	3.4	9.2	35.2	—	<b>557.0</b>	<b>615.9</b>	<b>1542.0</b>	<b>55.0</b>	<b>17.3</b>	74.0	—
Pre-operation	<10.0	9.7	—	—	—	—	<b>132.0</b>	<b>133.0</b>	<b>265.2</b>	<b>9.7</b>	15.3	106	—
<b>Case 4</b>													
Before external drainage	<10.0	<b>18.4</b>	<b>1.9</b>	<b>117.4</b>	<b>172.0</b>	32.0	<b>93.1</b>	<b>42.2</b>	<b>60.2</b>	<b>100.8</b>	<b>35.9</b>	—	—
Pre-operation	<10.0	9.0	<b>1.9</b>	11.2	<b>45.0</b>	—	<b>75.7</b>	23.9	<b>28.5</b>	<b>103.9</b>	<b>43.0</b>	—	—
<b>Case 5</b>													
Before external drainage	<10.0	9.4	<b>0.3</b>	<b>21.5</b>	32.2	68.0	<b>254.0</b>	<b>284.7</b>	<b>748.9</b>	<b>96.0</b>	<b>38.2</b>	<b>1330.0</b>	<b>382.5</b>
Pre-operation	<10.0	5.7	2.0	<b>14.8</b>	35	<b>83.0</b>	33.9	<b>52.7</b>	<b>362.1</b>	<b>9.2</b>	<b>19.5</b>	115.0	27.5
<b>Case 6</b>													
Before external drainage	<10.0	11.1	<b>1.7</b>	<b>30.7</b>	<b>45.6</b>	—	<b>265.4</b>	<b>210.2</b>	<b>597.4</b>	<b>133.8</b>	<b>55.3</b>	<b>365.0</b>	<b>1929.2</b>
Pre-operation	<10.0	6.86	2.1	10.9	36.0	—	<b>60.7</b>	<b>33.3</b>	<b>74.2</b>	<b>5.9</b>	<b>21.0</b>	57.0	30.5

CRP: C-reactive protein; WBC: White blood cell; FIB: Fibrinogen; PT: Prothrombin time; APTT: Activated partial thromboplastin time; NH<sub>3</sub>: Ammonia; AST: Aspartate aminotransferase; ALT: Alanine aminotransferase; γ-GGT: γ-glutamyl transpeptidase; DBIL: Direct bilirubin; IBIL: Indirect bilirubin; AMY: Amylase; LPS: Lipase

\* Numbers in bold indicated higher than the upper limit of reference interval

\*\* The laboratory results re-examined in 21 days after ultrasound-guided percutaneous external drainage

visited our center within 1 month of presenting symptoms (Fig. 5).

According to the laboratory and imaging results, the main indications for US-guided percutaneous external drainage were giant cyst and coagulopathy. Four patients (4/6, Cases 1–4) had a giant choledochal cyst and underwent US-guided PBD on admission or after conservative treatments. The size of their cysts decreased after US-guided PBD (Fig. 6). The other 2 patients (2/6, Cases 5 and 6) underwent US-guided PTCD and US-guided PTGD due to coagulopathy, respectively (Table 2). Of these patients, 4 patients (4/6, Cases 2–5) underwent external drainage under general anesthesia, and another 2 (2/6, Cases 1 and 4) under local anesthesia. Five patients (5/6) recovered well after external drainage and subsequently underwent the definitive operation, whereas 1 patient (1/6, Case 1) was considered to have liver fibrosis with the value of liver stiffness measurement of 58.8 kPa by Fibroscan 21 days after external drainage and ultimately underwent liver transplantation in another hospital 2 months after external drainage. The mean time from US-guided percutaneous external drainage to the definitive operation was  $12 \pm 9$  (3–21) days. The average length of hospital stay was  $24 \pm 9$  (16–31) days. No infection, electrolyte disturbance, and other complications of US-guided percutaneous external drainage occurred during hospitalization. At  $10.2 \pm 6.8$  (1.0–18.0) months follow-up, all patients had a normal liver function and US examination.

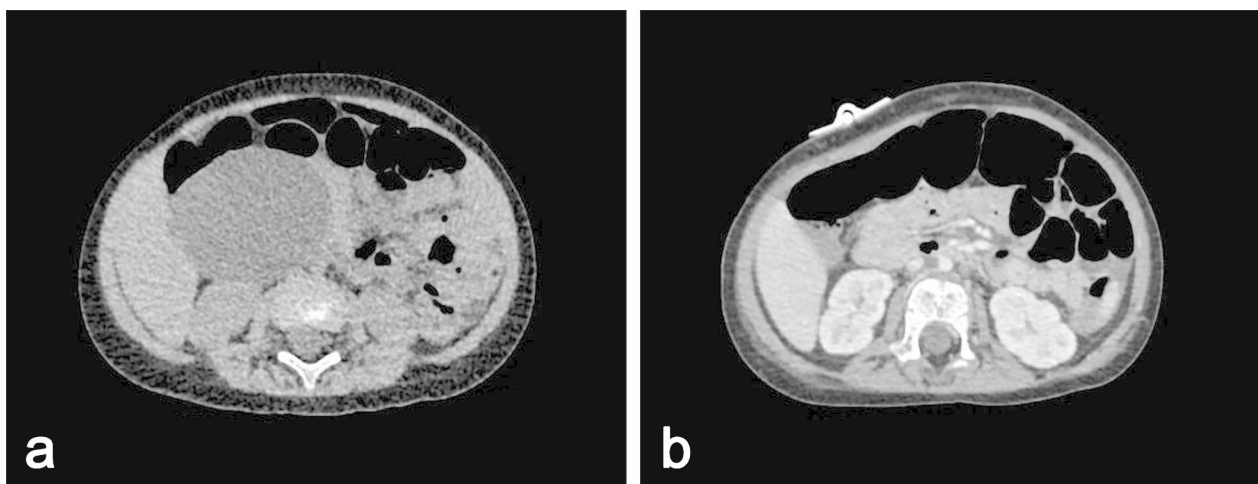
## Discussion

The study demonstrated the feasibility of the combination of US-guided percutaneous external drainage and subsequent cyst excision with Roux-en-Y hepaticojejunostomy to manage the complicated choledochal cyst

in children, especially for choledochal cyst with giant cysts or coagulopathy, although the applicable conditions should be further studied given the small sample size of patients.

Early cyst excision has been recommended for children with choledochal cyst to reduce the risk of long-term complications, such as biliary infections and cancer development [26–28]. Moreover, with the improvement of perioperative management and surgical techniques, most patients can undergo one-stage definitive operation [29, 30]. However, in some cases, severe common bile duct (CBD) dilatation, liver dysfunction, or coagulopathy due to biliary obstruction still leads to high surgical risk [12, 18]. Severe CBD dilatation often suggests the presence of CBD stenosis. It can be accompanied by bile duct inflammation, which will lead to repeated infection before the definitive operation, increase the difficulty of surgical operations, and have a high potential for anastomotic leakage during the primary closure [29, 31]. Liver dysfunction and coagulopathy also have a higher risk of anesthesia, intraoperative bleeding, and postoperative disseminated intravascular coagulation. Therefore, timely external drainage is still essential for complicated choledochal cyst [10, 11].

US is a good method to diagnose biliary diseases, including choledochal cyst, and US-guided percutaneous external drainage has been widely used in adult biliary disorders, such as acute cholecystitis, but few studies have reported its application in pediatric patients [8, 32]. Compared with fluoroscopy-guided external drainage, US-guided percutaneous external drainage can avoid unnecessary radiation exposure and reduce vascular injury during operation to prevent bleeding [15, 16]. However, visualization of the bile duct cannot be fully realized by US, so we suggest that cholangiography, CT, or MRCP should be performed consequently before the



**Fig. 6** The size of choledochal cyst before and after US-guided percutaneous external drainage. (A) CT before US-guided PBD, Case 4. (B) CT after US-guided PBD, Case 4



definitive operation to understand the morphologic features of the whole biliary tract and to check the location of the drainage catheter [16]. In addition, in this small cohort study, we found that patients could undergo appropriate external drainage (PBD, PTCD or PTGD) according to the extent of intrahepatic bile duct dilation, the size of extrahepatic cyst and gallbladder re-evaluated by US before external drainage, which had not been reported in previous studies. However, US is a very user-dependent imaging modality, and proper skills and rich experience of the radiologists are required to ensure the safety of patients, which is why US-guided percutaneous external drainage is not widely used in pediatric patients [33].

For patients with complicated choledochal cyst, US-guided percutaneous external drainage is minimally invasive and effective in relieving the symptoms of biliary obstruction, but it also has risks of electrolyte disturbance and infection [11, 16, 18]. It has been reported that the coagulation function of patients with choledochal cyst improved significantly 7 days after external drainage [12]. Cases 4–6 in our study with coagulopathy on admission were improved after US-guided percutaneous external drainage. To reduce the associated complications of US-guided percutaneous external drainage, all patients underwent the definitive operation within 21 days after external drainage during one hospitalization. If the liver and coagulation function of patients cannot be improved after US-guided percutaneous external drainage, the existence of liver fibrosis should be considered, which indicates a poor prognosis after the definitive operation; in such cases, further liver transplantation should be performed [34]. In our study, Case 1 presented symptoms in the infantile period, had already developed liver fibrosis on admission with a long-time biliary obstruction, and ultimately underwent liver transplantation. Therefore, US-guided percutaneous external drainage can help identify such cases. Similar to previous studies, early surgical intervention was recommended for all patients once a choledochal cyst was identified [1, 8, 12, 28, 34].

Many studies have shown that minimally invasive operations, including laparoscopic- or robot-assisted, have obvious advantages in treating choledochal cysts [1, 35, 36]. For complicated choledochal cyst, compared with traditional open external drainage, US-guided percutaneous external drainage can ensure minimally invasive treatment of the entire process. The giant choledochal cyst was conventionally treated by laparotomy, but in our study, except for Case 2, all subsequent patients underwent minimally invasive operations [18, 20]. This suggests that minimally invasive operation after US-guided percutaneous external drainage is safe and feasible for complicated choledochal cyst.

This study was limited by the small sample of patients, the isolated use of ultrasound, and the lack of a control group. Based on the present findings and the absence of a control group, it is not possible to prove the superiority of US-guided percutaneous external drainage over simple conservative treatments for patients with complicated choledochal cyst. Therefore, to what extent complicated choledochal cyst should be performed US-guided percutaneous external drainage before the definitive operation needs further studies. Further accumulation of cases is expected in the future.

## Conclusions

Our detailed analysis of this small cohort suggests that US-guided percutaneous external drainage is technically feasible for choledochal cyst with giant cysts or coagulopathy in children, which may provide suitable conditions for subsequent definitive operation and has a good prognosis.

## Abbreviations

US	ultrasound
CT	computed tomography
MRCP	magnetic resonance cholangiopancreatography
M	male
F	female
PBD	percutaneous biliary drainage
PTCD	percutaneous transhepatic cholangio-drainage
PTGD	percutaneous transhepatic gallbladder drainage
CRP	C-reactive protein
WBC	white blood cell
FIB	fibrinogen
PT	prothrombin time
APTT	activated partial thromboplastin time
NH <sub>3</sub>	ammonia
AST	aspartate aminotransferase
ALT	alanine aminotransferase
γ-GGT	γ-glutamyl transpeptidase
DBIL	direct bilirubin
IBIL	indirect bilirubin
AMY	amylase
LPS	lipase
CBD	common bile duct

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## Author contributions

Conceptualization: JYY, CKL. Study design: JYY, CKL, DZ, XMW, YJC. Execution: JYY, CKL, DZ, CHP, WBP, WC, SWW. Acquisition of the data: JYY, MKZ. Analysis and interpretation: JYY, CKL, DZ, MKZ. Writing: JYY, CKL, DZ, MKZ. Review and editing: JYY, CKL, DZ, MKZ, CHP, WBP, WC, SWW, XMW, YJC. The author(s) read and approved the final manuscript.

## Funding

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## Data Availability

All data generated or analysed during this study are included in this published article.

## Declarations

### Ethics approval and consent to participate

This study was approved by the Ethics Committee of Beijing Children's Hospital (Approval number: 2022-E-214-R) and the need for informed consent was waived. All methods used in this study adhere to the tenets of the Declaration of Helsinki 1967.

### Consent for publication

Not applicable.

### Competing interests

The authors declare that they have no competing interests.

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## References

- Xie X, Li Y, Li K, Wang Q, Xiang B. Total robot-assisted choledochal cyst excision using da Vinci surgical system in pediatrics: report of 10 cases. *J Pediatr Surg.* 2021;56:553–8.
- Sela-Herman S, Scharshmidt BF. Choledochal cyst, a disease for all ages. *Lancet.* 1996;347:779.
- Soares KC, Arnaoutakis DJ, Kamel I, Rastegar N, Anders S, Maithel S, et al. Choledochal Cysts: Presentation, clinical differentiation, and management. *J AM Coll Surg.* 2014;219:1167–80.
- Glenn F, McSherry CK. Congenital segmental cystic dilatation of the biliary ductal system. *Ann Surg.* 1973;177:705–13.
- Santiago I, Loureiro R, Curvo-Semedo L, Marques C, Tarda 'guila F, Matos C, et al. Congenital cystic lesions of the biliary tree. *AJR Am J Roentgenol.* 2012;198:825–35.
- Brown ZJ, Baghdadi A, Kamel I, Labiner HE, Hewitt DB, Pawlik TM. Diagnosis and management of choledochal cysts. *HPB (Oxford).* 2022;51365-182X(22)01600-8.
- Chowbey PK, Soni V, Sharma A, Khullar R, Bajjal M. Laparoscopic hepaticojejunostomy for biliary strictures: the experience of 10 patients. *Surg Endosc.* 2005;19:273–9.
- Soares KC, Goldstein SD, Ghaseb MA, Kamel I, Hackam DJ, Pawlik TM. Pediatric choledochal cysts: diagnosis and current management. *Pediatr Surg Int.* 2017;33:637–50.
- She WH, Chung HY, Lan LC, Wong KK, Saing H, Tam PK. Management of choledochal cyst: 30 years of experience and results in a single center. *J Pediatr Surg.* 2009;44:2307–11.
- Lal R, Agarwal S, Shivhare R, Kumar A, Sikora SS, Kapoor VK, et al. Management of complicated choledochal cysts. *Dig Surg.* 2007;24:456–62.
- Song G, Mou Y, Ren C, Zhou H, Wang J. Comparison and selection of three methods of minimally invasive external drainage for children with congenital Choledochal cysts. *J Laparoendosc Adv Surg Tech A.* 2021;31:462–7.
- Diao M, Li L, Cheng W. Coagulopathy in a subtype of choledochal cyst and management strategy. *World J Gastroenterol.* 2014;20:10606–12.
- Uno K, Tsuchida Y, Kawarasaki H. Development of intrahepatic cholelithiasis after primary excision of choledochal cysts. *J Am Coll Surg.* 1996;183:583–8.
- Minagawa T, Dowaki S, Kikunaga H, Fujita K, Ishikawa K, Mori K, et al. Endoscopic biliary drainage as a bridging procedure to single-stage surgery for perforated choledochal cyst: a case report and review of the literature. *Surg Case Rep.* 2015;1:117.
- Kahrman G, Ozcan N, Gorkem SB. Percutaneous management of benign biliary disorders in children. *Diagn Interv Imaging.* 2018;99:37–42.
- Shan QY, Jiang H, Chen SL, Chen HD, Guo Y, Xie XY, et al. Postsurgical Management of dilated biliary tract in children: Ultrasound-Guided Percutaneous Transhepatic Cholangial Drainage and subsequent percutaneous Ultrasound Cholangiography. *AJR Am J Roentgenol.* 2020;214:1377–83.
- Jin X, Jiang Y, Tang J. Ultrasound-Guided Percutaneous Transhepatic Gallbladder Drainage Improves the Prognosis of Patients with Severe Acute Cholecystitis. *Evid Based Complement Alternat Med.* 2022;5045869.
- Upadhyaya VD, Kumar B, Raut SK, Sthapak E. External drainage of Giant Infantile Choledochal Cyst before definitive repair: is it worth? *J Clin Diagn Res.* 2017;11:PR01–4.
- Todani T. Congenital choledochal dilatation: classification, clinical features, and long-term results. *J Hepatobiliary Pancreat Surg.* 1997;4:276–82.
- Kumar S, Singla S, Kumar S, Singh RK, Raj AK, Mandal M. Tactics, techniques, and challenges in the management of giant choledochal cyst in adolescents and adults: a retrospective cohort study. *Langenbecks Arch Surg.* 2021;406:1925–33.
- Yan R, Peng Y, Hu L, Zhang W, Li Q, Wang Y, et al. Continuous reference intervals for 21 biochemical and hematological analytes in healthy chinese children and adolescents: the PRINCE study. *Clin Biochem.* 2022;102:9–18.
- Deras P, Nouri J, Martinez O, Aubry E, Capdevila X, Charbit J. Diagnostic performance of prothrombin time point-of-care to detect acute traumatic coagulopathy on admission: experience of 522 cases in trauma center. *Transfusion.* 2018;58:1781–91.
- Radcke S, Dillon JF, Murray AL. A systematic review of the prevalence of mildly abnormal liver function tests and associated health outcomes. *Eur J Gastroenterol Hepatol.* 2015;27:1–7.
- Ismail OZ, Bhayana V. Lipase or amylase for the diagnosis of acute pancreatitis? *Clin Biochem.* 2017;50:1275–80.
- Shen QL, Chen YJ, Wang ZM, Zhang TC, Pang WB, Shu J, et al. Assessment of liver fibrosis by Fibroscan as compared to liver biopsy in biliary atresia. *World J Gastroenterol.* 2015;21:6931–6.
- Gomes C, Tivnan P, McAneny D, Tseng JF, Tkacz J, Sachs TE. Choledochal cyst or benign biliary dilation: is resection always necessary? *J Gastrointest Surg.* 2021;25:2353–7.
- Tannuri ACA, Hara LAA, Paganoti GF, Andrade WC, Tannuri U. Choledochal cysts in children: how to diagnose and operate on. *Clin (Sao Paulo).* 2020;75:e1539.
- Serin KR, Ercan LD, Ibis C, Ozden I, Tekant Y. Choledochal cysts: management and long-term follow-up. *Surgeon.* 2021;19:200–6.
- Ohba G, Yamamoto H, Nakayama M, Honda S, Taketomi A. Single-stage operation for perforated choledochal cyst. *J Pediatr Surg.* 2018;53:653–5.
- Yin T, Chen S, Li L, Diao M, Huang T, Li Q, et al. One- versus two-stage single-incision laparoscopic cyst excision and hepaticojejunostomy in patients with completely perforated choledochal cysts and good medical conditions. *Pediatr Surg Int.* 2022;38:541–5.
- Yamashita Y, Ueda K, Abe H, Tamura T, Itonaga M, Yoshida T, et al. Common bile Duct Dilatation with Stones indicates requirement for early drainage in patients with or without Cholangitis. *Gastroenterol Res.* 2013;6:219–26.
- Ahmed O, Rogers AC, Bolger JC, Mastrosimone A, Lee MJ, Keeling AN, et al. Meta-analysis of outcomes of endoscopic ultrasound-guided gallbladder drainage versus percutaneous cholecystostomy for the management of acute cholecystitis. *Surg Endosc.* 2018;32:1627–35.
- Todsen T. Surgeon-performed ultrasonography. *Dan Med J.* 2017;64:B5421.
- Fumino S, Higuchi K, Aoi S, Furukawa T, Kimura O, Tajiri T. Clinical analysis of liver fibrosis in choledochal cyst. *Pediatr Surg Int.* 2013;29:1097–102.
- Wang B, Feng Q, Mao JX, Liu L, Wong KK. Early experience with laparoscopic excision of choledochal cyst in 41 children. *J Pediatr Surg.* 2012;47:2175–8.
- Koga H, Ochi T, Seo S, Miyake Y, Lane GJ, Yamataka A. Comparison of 2K and 4K imaging systems for laparoscopic repair of choledochal cyst in children. *J Pediatr Surg.* 2022;57:235–8.

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