



# The impact of pre-operative cholecystostomy on laparoscopic excision of choledochal cyst in paediatric patients

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## Abstract

**Purpose** This aim of this study was to identify the pre-operative risk factors for conversion during laparoscopic excision of choledochal cyst in paediatric patients.

**Methods** A retrospective single-centre study was carried out. All paediatric patients (< 18 years) who had undergone laparoscopic excision of choledochal cyst between 2004 and 2021 were reviewed. The outcome was conversion to open surgery and pre-operative factors that affected the conversion rate were analyzed.

**Results** Sixty-one patients were included. Conversion was required in 24 cases (39.3%). There was no difference in the conversion rate between the first (before 2012,  $n=30$ ) and second (after 2012,  $n=31$ ) half of the series (36.7% vs. 42.0%,  $p=0.674$ ). Majority was type 1 cyst (86.8%) and the median cyst size was 4.6 cm (IQR: 2.2–6.4 cm). Antenatal diagnosis was available in 18 patients (29.5%). The median age at operation was 23.0 months (IQR: 8.0–72.0 months). Pre-operatively, 19 patients (31.1%) suffered from cholangitis and 5 (8.2%) of them required cholecystostomy. Comparing patients with successful laparoscopic surgery (L) and converted cases (C), there were no differences in the age at operation ( $p=0.74$ ), cyst size ( $p=0.35$ ), availability of antenatal diagnosis ( $p=0.23$ ) and cholangitic episodes ( $p=0.40$ ). However, a higher percentage of patients required cholecystostomy in the converted group (L vs. C = 2.7% vs. 16.7%,  $p=0.05$ ). Using logistic regression analysis, it was also a risk factor for conversion (OR = 3.5 [1.37–5.21],  $p=0.05$ ).

**Conclusion** Pre-operative cholecystostomy is a potential risk factor for conversion during laparoscopic excision of choledochal cyst in children.

**Keywords** Choledochal cyst · Cholangitis · Laparoscopy · Biliary drainage · Cholecystostomy

## Introduction

Choledochal cyst (CC) is a congenital disorder characterized by dilations of the biliary tree. This disorder is more prevalent in Asian populations especially among Japanese with an estimated incidence around 1 in 1000 [1]. In contrast, it is rare in Western countries [2]. CC can be divided into five subtypes according to the morphology and level of ductal dilatation [3]. Type I cyst refers to a fusiform dilatation of the extra-hepatic bile duct and is the most common type. Although CC can present at all ages, most cases are

diagnosed in early childhood. The introduction of fetal ultrasound has resulted in more CC being picked up antenatally. While symptomatic CC warrants surgical treatment, excision is also recommended for asymptomatic cases because of the long-term risks including obstructive jaundice, cholangitis, and cholangiocarcinoma.

Laparoscopic excision of CC in a paediatric patient was first reported by Farello et al. in 1995 and has recently become the state-of-art approach [4]. Although a longer operative time is sometimes encountered, patients generally recover faster with a shorter hospital stay. Both early and long-term outcomes of laparoscopic surgery are comparable to conventional open surgery [5]. However, there are still occasions when conversion is required even under the hands of skillful surgeons. The aim of this study was to identify the pre-operative factors that may increase the chance of conversion during laparoscopic excision of CC in paediatric patients.

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## Method

### Subjects and data

This was a retrospective single-centre study. All paediatric patients (< 18 years) who underwent laparoscopic excision of CC between 2004 and 2021 were reviewed. All the operations were performed by three consultant surgeons with standardized technique. CCs were classified according to Todani classification [3]. Cholangitis was defined as the presence of increased body temperature to above 38.5 degree Celsius and total serum bilirubin level above 20  $\mu\text{mol/L}$ . All cholangitic episodes were treated with intravenous antibiotics for at least 1 week. If there was no sign of clinical improvement, external biliary drainage by cholecystostomy would be considered.

Demographic data including gender, age at operation, antenatal diagnosis, type of cyst, surgical approach, operative time, and peri-operative events were collected. The primary outcome was conversion to open surgery. Pre-operative factors that potentially affected the conversion rate were analyzed. Patients with incomplete record and scheduled open surgery were excluded. This study was approved by the Institutional Review Board of the authors' centre (UW 21-648).

### Operative technique

#### External biliary drainage by cholecystostomy

The procedure was performed under general anaesthesia. A 5 mm sub-umbilical camera port and two additional 3 mm working ports were inserted at right and left side of the abdomen via open technique. The gallbladder was identified under direct visualization and dissected away from the liver until the fundus could be brought to the abdominal surface. The pneumoperitoneum was then released and the gallbladder was exteriorized by enlarging the right side port wound. A cholecystostomy was created and a balloon catheter was inserted as the conduit to provide drainage. The catheter was connected to a drainage bag and secured by sutures.

#### Laparoscopic excision of choledochal cyst

The operations were attended by at least two surgeons who were competent in choledochal cyst surgery and followed the same operative approach that was described in our previous publication [6]. In brief, patient was placed in a supine position and 4 to 5 working ports including the camera port were inserted. The original port sites created during previous cholecystostomy were re-used. The liver and gallbladders

were hitched up to reveal the cyst. Excision was performed with monopolar hook, bipolar Maryland dissector and ultrasonic dissector as appropriate. Distally, the bile duct was transected at the retroduodenal region where tapering was encountered. The proximal dissection was extended to the hepatic hilum. The cyst was dissected circumferentially away from the hepatic artery and the portal vein. It was transected just above the confluence of common hepatic duct and cystic duct, leaving a small cuff for anastomosis. For biliary reconstruction, we used to perform Roux-en-Y hepaticojejunostomy (HJ) before 2010 and have switched to hepaticoduodenostomy (HD) since 2010 [6]. In HJ reconstruction, a Roux loop of 30 to 40 cm was brought to the liver hilum in retrocolic manner for anastomosis. In HD, an opening was made in the second part of the duodenum at around 3 cm from the pylorus for anastomosis. The decision to convert to open surgery was based on the judgement of the operating surgeon.

### Statistical analysis

Values were presented as median (interquartile range). Continuous variables were compared with Kruskal–Wallis test. Categorical variables were compared using Pearson chi-squared test. Logistic regression analysis was carried out to determine the association between different factors and conversion. Scientific analysis was performed with a commercial statistical package (Windows, version 26.0; SPSS Inc., Armonk, NY, USA). In all analyses, a *p* value of less than or equal to 0.05 was considered to be statistically significant.

## Results

During the study period, 76 patients received surgery for choledochal cyst and 61 patients met the inclusion criteria (Table 1). There were more female patients (female:male = 49:12). Twenty-four operations (39.3%) required conversion to open surgery with the following reasons: bleeding ( $n = 5$ ), severe adhesion ( $n = 11$ ), variant of usual anatomy ( $n = 4$ ) and others ( $n = 4$ ). There was no difference in the conversion rate between the first (before 2012,  $n = 30$ ) and second (after 2012,  $n = 31$ ) half of the series (36.7% vs. 42.0%,  $p = 0.674$ ). The majority of the cyst was type 1 ( $n = 53$ ) and the median cyst size was 4.6 cm (2.2–6.4 cm). Antenatal diagnosis was available in 18 patients (29.5%). The median age at operation was 23.0 months (8.0–72.0 months). Pre-operatively, 19 patients (31.1%) suffered from cholangitis. Five patients (8.2%) did not respond to medical treatment and required external biliary drainage.

When patients with successful laparoscopic surgery (L) were compared with converted cases (C), there were no differences in the age at operation (L vs. C 18.0 [8.0–36.0] vs. 22.0 [10.0–56.0] months,  $p=0.74$ ), cyst size (L vs. C: 3.8 [3.2–5.4] vs. 4.3 [2.9–6.1] cm,  $p=0.35$ ), presence of antenatal diagnosis (L vs. C = 35.1% vs. 20.8%,  $p=0.23$ ), and pre-operative cholangitis (L vs. C: 35.1% vs 25.0%,  $p=0.40$ ). However, there was a higher percentage of patient requiring external drainage by cholecystostomy in the converted group (L vs. C = 2.7% vs. 16.7%,  $p=0.05$ ) (Table 2).

Next, we evaluated the association between above factors and conversion rate. With logistic regression analysis, only pre-operative cholecystostomy was a risk factor for conversion (OR = 3.51 [1.37–5.21],  $p=0.05$ ). The other factors including operative age over 23.0 months (OR = 0.97 [0.56–1.87],  $p=0.67$ ), cyst size larger than 4.6 cm (OR = 1.45 [0.89–2.11],  $p=0.51$ ), presence of antenatal diagnosis (OR = 0.48 [0.14–1.60],  $p=0.31$ ), and pre-operative cholangitis (OR = 0.61 [0.19–1.93],  $p=0.61$ ) were not risk factors for conversion (Table 2).

For the five patients requiring pre-operative cholecystostomy, they all suffered from cholangitis and did not show any clinical improvement after 1 week of antibiotics therapy. The median serum bilirubin level before drainage was 89  $\mu\text{mol/L}$  (76–121  $\mu\text{mol/L}$ ). Antenatal diagnosis was available in two patients while the other presented at the age between 2 and 23 months. The median cyst size was 7.6 cm (5.3–10.0 cm) and the median time from drainage to definitive cyst excision was 49 days (23.5–57.5 days). None of them suffered from drainage related complications and they were allowed to stay home before cyst excision. Three patients required conversion due to severe intra-peritoneal adhesions. The median length of stay after cyst excision was 8.0 days (5.5–11.0 days). There were no surgical complications reported (Table 3).

## Discussion

Laparoscopic excision of CC has been developed for almost 20 years and excellent outcomes were reported. The reported conversion rate ranged from 10 to 37% [7, 8]. Although there is no consensus regarding the optimal timing of surgery, most surgeons prefer to operate under an elective situation when the patient's condition is stable. Jaundice and acute cholangitis are adverse factors that can potentially impair the outcomes. These should be treated before cyst excision. Antibiotics therapy is the first line management for cholangitis but this is often ineffective. When this happens, biliary drainage is the next step to consider.

Various techniques for biliary drainage have been proposed and are broadly classified into internal or external

[9]. Each approach has its own merits and limitations. Internal drainage is usually achieved by endoscopic retrograde cholangiopancreatography (ERCP). This is less invasive compared to external drainage as a skin incision could be avoided. The preservation of enterohepatic circulation of bile is another advantage. However, performing ERCP in paediatric patients could be limited by the size of the endoscope. The paediatric duodenoscope scope has a diameter of 7.5 mm and difficulty maybe encountered during oral intubation in infants. Infant ERCP has been reported but cannulation failure can occur in up to 10% [10, 11]. In addition, sphincterotomy is sometimes needed and this procedure is associated with the risk of bleeding, duodenal perforation, and pancreatitis. Percutaneous transhepatic biliary drainage (PTBD) is the alternative when ERCP is not feasible. The prerequisite for a successful drainage by PTBD is the presence of dilated ducts for cannulation but this may not be present. Besides, PTBD in a small-sized baby is technically challenging and can result in serious complications including pseudoaneurysm and fistula formation [12]. Dislodgment of PTBD catheter is common and require urgent revision. As a result, many patients opt to stay in the hospital until definitive surgery. The insertion of drainage catheter into the cyst has been proposed but the safety of puncturing the acutely inflamed bile duct is the major concern [8].

Laparoscopic cholecystostomy was introduced as a surgical treatment to provide temporary relief of jaundice in children with CC around 10 years ago. In 2015, Yamoto et al. reported a series of seven patients with choledochal cyst undergoing cholecystostomy before cyst excision [9]. Their indications were slightly different from ours as the majority of them suffered from ruptured or impending ruptured cyst. Together with the data from our current series, it is obvious that cholecystostomy does have a role in managing complicated CC. It can enhance the recovery of the patients to receive definitive surgery. There is no guideline to when cyst excision should be carried out. In our opinion, this interval is flexible and largely depends on the recovery of the patient. We have demonstrated that patients with cholecystostomy tube did not need to stay in the hospital while waiting for definitive surgery.

Although cholecystostomy can be a bridging therapy to ameliorate jaundice in obstructive CC, our results suggested it is a risk factor for conversion during subsequent cyst excision by laparoscopic approach. Indeed, the patients requiring drainage were all suffering from the more severe form of cholangitis that was refractory to antibiotics therapy. We postulated that severe inflammation could have resulted in adhesions and hence increased the complexity of the surgery. Moreover, cholecystostomy itself inevitably created more adhesions at the operative site. However, whether the adhesions were severe enough to warrant conversion was mostly

**Table 1** Demographic information of 61 patients undergoing laparoscopic excision of choledochal cyst between 2004 and 2021

Total number of patients	61
Female to male	49:12
Antenatal diagnosis	18 (29.5%)
Age of operation (months)	23.0 (8.0–72.0)
Number of patients with pre-operative cholangitis	19 (31.3%)
Number of patients requiring pre-operative cholecystostomy	5 (8.2%)
Conversion to open surgery	24 (39.3%)
Before 2012	11 (36.7%)
After 2012	13 (42.0%)
Cyst type	
Type 1	53 (86.8%)
Type 4	8 (13.1%)
Cyst size (cm)	4.6 (2.2–6.4)

a judgement call. There have been no standard criteria for conversion but we always adopted the safest approach and avoided heroic actions. This may explain a higher conversion rate in our series when compared to other studies. We are pleased to report that none of our patients who have undergone drainage procedure suffered from surgical complications after cyst excision. Since all patients with drainage were not responsive to antibiotics therapy, the necessity for conversion could be related to the more severe cholangitis rather than the drainage procedure itself. We could not prove or disprove this assumption. However, given the higher

chance of conversion after cholecystostomy, we postulate that in selected cases, ERCP or a longer period of antibiotics therapy maybe considered before proceeding to external drainage. A large-scaled prospective study maybe able to test our postulation. In this study, as the sample size was small, we could not perform further sub-group analysis to identify any additional factors that might affect the conversion rate among those drained cysts. Nonetheless, from the data presented in Table 3, the severity of jaundice, cyst size and timing to definitive surgery did not appear to have a significant impact on this.

Besides the small sample size, our study was also limited by the retrospective study design. As many other operations, the decision to convert was based on the surgeon's subjective assessment. This bias has been minimized by the number of surgeons ( $n=3$ ) performing this procedure but it can never become negligible. Another major factor that would influence the conversion rate is the experience of the surgeon especially this study covered a long period of time and our technique has become more mature in the recent years.

In conclusion, external biliary drainage by cholecystostomy is an effective treatment for refractory cholangitis in CC. It enhances the recovery of the patient to undergo cyst excision. However, the operating surgeons should also anticipate a higher chance of conversion during subsequent cyst excision by laparoscopic approach. Alternative drainage methods can be attempted but the availability of equipment and surgical expertise should be taken into consideration.

**Table 2** The associations between different factors and conversion to open surgery

	Without conversion ( $n=37$ )	With conversion ( $n=24$ )	<i>p</i> value	Odds ratio	<i>p</i> value
Median age at operation (months)	18.0 (8.0–36.0)	22.0 (10.0–56.0)	0.74	0.97 (0.56–1.87)*	0.67
Median cyst size (cm)	3.8 (3.2–5.4)	4.3 (2.9–6.1)	0.35	1.45 (0.89–2.11)#	0.51
Antenatal diagnosis available (%)	35.1	20.8	0.23	0.48 (0.14–1.60)	0.31
Cholangitis (%)	35.1	25.0	0.40	0.61 (0.19–1.93)	0.61
Pre-operative cholecystostomy (%)	2.7	16.7	0.05	3.51 (1.37–5.21)	0.05

\*Age at operation over 23.0 months

#Cyst size larger than 4.6 cm, respectively

**Table 3** Characteristics of the five patients who have undergone pre-operative external biliary drainage by cholecystostomy before cyst excision

Sex	Antenatal diagnosis	Current age (years)	Age at excision (months)	Transverse diameter of the cyst (cm)	Serum bilirubin before drainage ( $\mu\text{mol/L}$ )	Interval between drainage at cyst excision (days)	Conversion
M	Yes	4	3	7.6	127	14	Yes
F	No	7	25	8.2	89	49	No
F	No	10	7	7.3	115	60	Yes
F	Yes	10	5	11.8	64	55	Yes
F	No	12	4	3.2	88	33	No

**Author contributions** PHYC and KKYW designed the study; PHYC, FY and MKIM collected and analyzed the data; PHYC wrote the manuscript; KKYW performed critical review.

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## Declarations

**Conflict of interest** This abstract was presented at the BAPS congress 2023.

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