

## Mid-term Outcomes of Laparoscopic Versus Open Choledochal Cyst Excision in a Tertiary Paediatric Hospital

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### Abstract

**Introduction:** Paediatric laparoscopic choledochal cyst excision has increasingly gained acceptance as an alternative to open excision. Laparoscopic excision is feasible and safe in the short term, but long-term outcomes are not as well established. KK Women's and Children's Hospital started performing laparoscopic choledochal cyst excision in children since 2007. In this paper, we report our experience with the laparoscopic approach, and the early and mid-term outcomes in comparison with the conventional open approach. **Materials and Methods:** Thirty-five consecutive cases by a single surgeon between May 2006 and April 2012 were retrospectively reviewed. Patient characteristics and surgical outcomes were analysed. **Results:** There were 13 laparoscopic and 22 open cases. Baseline patient characteristics were similar. Operative time was longer in the laparoscopic group. Three cases in the laparoscopic group were converted to open in our early experience. There were no differences in time to feeds or length of hospitalisation. One laparoscopic case developed minor bile leak that resolved on conservative management. There were no complications in the laparoscopic group on median follow-up of 35 months. In the open group, there was 1 case of pancreatitis, cholangitis, and hypertrophic scarring respectively. There were 3 cases of suspected adhesive colic that resolved without surgery. **Conclusion:** Laparoscopic choledochal cyst excision enjoys excellent early and mid-term outcomes compared to open excision, even in centres with smaller patient volume. It should be the approach of choice where technical expertise is available.

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**Key words:** Hepaticojejunostomy, Minimally invasive, Postoperative outcomes

### Introduction

Paediatric choledochal cyst is an uncommon benign cystic dilatation of the biliary tree, and is more prevalent in Asia than in the West. Accepted treatment consists of complete excision of the cyst with bilio-enteric reconstruction. Left untreated, choledochal cysts can lead to significant morbidity and mortality, including spontaneous rupture, pancreatitis, cholangitis, biliary cirrhosis, and an increased risk of developing cholangiocarcinoma.

With the advent of minimally invasive surgery in children, the laparoscopic approach has increasingly gained acceptance since its first description by Farello in 1995.<sup>1</sup> Although laparoscopic excision of choledochal cyst is technically demanding, several authors have since reported on its feasibility and short-term safety. However, the mid-term and long-term outcomes have yet to be established.

The KK Women's and Children's Hospital is one of two tertiary paediatric hospitals in Singapore. Since 2007, we have been performing the laparoscopic procedure on children. This is the first local series of laparoscopic choledochal cyst excision reported in Singapore.

### Materials and Methods

Thirty-five consecutive choledochal cyst excisions performed by a single surgeon between May 2006 and April 2012 were retrospectively reviewed. The decision between laparoscopic and open surgery was based on surgeon and parental preference. Excluded from review were 2 cases performed by another surgeon, and 2 cases performed in an emergency setting for perforated choledochal cysts.

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The data retrospectively collected from inpatient and outpatient electronic, and written medical records included patient age, gender, clinical presentation, cyst size, operative time, time to commencement of feeds, time to discharge and all postoperative complications. Cyst size was represented by maximum measured dimension on preoperative imaging, either on ultrasonography, computed tomographic scans or magnetic resonance cholangiopancreatography (MRCP).

Roux-en-Y hepatico-jejunostomy (HJ) was the preferred bilio-enteric reconstruction for all cases. Among the laparoscopic cases, an epigastric port was used for liver retraction for the first 3 cases. Subsequent operations were done via a 4-port technique, with liver retraction effected by hitch stitches on the falciform ligament and gallbladder. A combination of 3 mm and 5 mm instruments were used. Laparoscopic dissection was largely carried out with the monopolar hook. Proximally, the duct was transected just below the level of the hepatic bifurcation. Distally, the intrapancreatic portion of the cyst was followed until it tapered to the calibre of the normal common bile duct, as shown in Figure 1. This was followed by extra-corporeal transumbilical jejunostomy and intra-corporeal HJ. Open excision was performed through a right subcostal incision, with subsequent cyst excision and enteral reconstruction performed in a manner similar to the laparoscopic approach.

Postoperative management of all patients followed standard guidelines, with commencement of feeds on return of bowel function, and discharge from hospital after establishing full feeding. Follow-up was at 2 weeks post-discharge with clinical, biochemical and ultrasound



Fig. 1. Site of distal common bile duct transection beyond choledochal cyst.

assessments, repeated at 3 months and thereafter, annually if well.

Patient characteristics and surgical outcomes were analysed using SPSS 19.0 (Chicago). Mann-Whitney U tests were used for statistical analysis. A level of  $P < 0.05$  was considered to be statistically significant. The review had obtained approval by the hospital ethics board.

## Results

Of the 35 cases, there were 13 laparoscopic cases and 22 open cases. The demographic data of the patients in both groups are summarised in Table 1. There were no significant differences between patients' age at the time of surgery or preoperative cyst size. One patient in each group was antenatally diagnosed and asymptomatic before surgery. All other patients were symptomatic and presented variably with recurrent abdominal pain, cholangitis, pancreatitis or obstructive jaundice. Table 2 shows details of the clinical presentations in each group.

Table 1. Patient Characteristics

	Laparoscopic (n = 13)	Open (n = 22)	P value
Median age at operation (months)	36.5	36.5	0.824
Gender ratio (Female : Male)	8 : 5	19 : 3	
Type of choledochal cyst (based on Todani Classification <sup>14</sup> )	Type 1 (100%)	Type 1 (90.9%) Type 4 (9.09%)	
Median cyst Dimension (cm) (on preoperative imaging)	4.4 (Range, 1.7 – 15.1)	2.45 (Range, 0.7 – 10.7)	0.091

Table 2. Clinical Preoperative Presentation of Patients

Clinical Presentation	Laparoscopic (n = 13)	Open (n = 22)
Asymptomatic	1	1
Cholangitis	8	5
Obstructive jaundice	1	3
Pancreatitis	0	8
Recurrent abdominal pain	3	5

Median operative time was longer in the laparoscopic group compared to the open group (536 vs 300 minutes,  $P < 0.01$ ). There were no intraoperative complications in either group. Postoperatively, there were no significant differences in time to commencement of feeds ( $P = 0.473$ , Table 3), or total length of hospitalisation ( $P = 0.184$ , Table 3). The median follow-up period was 35 months in the laparoscopic group, and 41 months in the open group. No patient was lost to follow-up.

In the early postoperative period, a patient in the laparoscopic group developed minor bile leak. This resolved conservatively with intravenous antibiotics, and the child remained asymptomatic with normal liver biochemistry and ultrasound imaging on follow-up of 11 months. There were no early complications in the open group. On subsequent outpatient follow-up, there were no complications in the laparoscopic group. In the open group, there was 1 case of postoperative pancreatitis, 1 case of postoperative cholangitis that resolved with intravenous antibiotics, and 1 case of asymptomatic intra-hepatic biliary dilatation that resolved spontaneously after 62 months of follow-up. There was also 1 case of hypertrophic scar formation at the subcostal incision, and 3 cases of non-obstructive abdominal colic. These were possibly adhesive in nature, but did not require re-operation.

Three cases in the laparoscopic group which were performed early in our experience necessitated conversion to open surgery. Two were for bleeding due to dense adhesions from previous inflammation; and one was for difficult anatomy requiring open ductoplasty. However, a subsequent case with cyst extension into the proximal ducts requiring extensive duct reconstruction was completed laparoscopically with good early results.

## Discussion

The role of laparoscopic surgery in infants and children is now widely accepted. Minimal access and lesser tissue handling accelerates recovery with reduced postoperative pain, ileus and adhesions formation. Wound complications

are reduced and cosmesis is improved.<sup>2</sup> Magnification allows improved visualisation of deep-seated anatomical structures, resulting in a decreased risk of bleeding and inadvertent injury.<sup>3</sup> These advantages are amplified when the open surgery necessitates a sizeable incision, and when the incision is in the upper abdomen, impacting postoperative respiratory rehabilitation. These conditions apply particularly to the surgical excision of choledochal cyst, where a large muscle-dividing right subcostal incision is required to access the deep hepatic hilum for accurate anastomosis at the hepatic bifurcation.

Despite these benefits, laparoscopic excision of choledochal cyst is not widely practised. For many surgeons, it remains a technically demanding operation. The condition is rare, and bilio-enteric anastomosis demands the advanced skillset of intra-corporeal laparoscopic suturing. The notorious variability of the hepatobiliary vascular anatomy, added to the difficult surgical planes from recurrent episodes of inflammation in most patients, contribute significantly to the surgical challenge. This is particularly so in centres with lower patient volumes.

Notwithstanding these challenges, authors in small case series have demonstrated early to mid-term safety of laparoscopic choledochal cyst excision in children.<sup>4,9</sup> Larger series from a few major centres in China and Vietnam (Table 4) show similar or improved long-term outcomes when compared to historical data from the open method.<sup>3,10-12</sup> A key shortcoming in drawing conclusions from historical data is that factors other than the surgical approach, such as changes in perioperative practices or improved equipment, may introduce bias. Our report compares outcomes between the laparoscopic and open group performed within the same study period, with patients from both groups subject to similar perioperative practice guidelines, thus eliminating this bias. Limitations of this study are its small study population, and lack of randomisation with resultant potential selection bias. Nonetheless, both patient groups were found to be evenly matched in terms of cyst size and age—the 2 factors most likely to influence procedural difficulty and outcomes.

Table 3. Postoperative Outcomes for Laparoscopic and Open Choledochal Cyst Excision

Sample size	Hospital stay (days)	Postoperative feeding (median, days)	Overall complications (%)	Bile leak	Pancreatitis	Cholangitis	Anastomotic stenosis	Adhesive ileus or obstruction	Roux loop obstruction
Lap: 13	6 (4–25)	3 (3–4)	7.7	1 (7.7%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)
Open: 22	5 (5–7)	3 (2–5)	31.8	0 (0.0%)	1 (4.5%)	1 (4.5%)	0 (0.0%)	3 (13.6%)	0 (0.0%)

Table 4. Summary of Literature Comparing Outcomes for Laparoscopic and Open Choledochal Cyst Excision

Author/ Year	Sample size	Hospital stay (mean ± SD) (days)	Postoperative feeding (mean ±SD) (days)	Overall complications (%)	Bile leak (%)	Pancreatitis (%)	Cholangitis (%)	Anastomotic stenosis (%)	Adhesive ileus or obstruction (%)	Roux loop obstruction (%)
She 2009 <sup>10</sup>	Lap: 10	NA	NA	20.0	NA	1.5	1.5	10.0	0.0	NA
	Open: 65			30.0		0.0	0.0	1.5	1.5	
Liuming 2011 <sup>11</sup>	Lap: 39	5.5 ± 0.9	3.5 ± 0.7	20.5	2.6	0.0			0.0	2.6
	Open: 38	7.0 ± 1.4	4.9 ± 0.9	15.8	2.6	2.6	NA	NA	2.6	0.0
Diao 2011 <sup>3</sup>	Lap: 218	7.4 ± 2.4	2.86 ± 1.23	2.7	0.9	0.0	0.0	0.0	0.0	0.9
	Open: 200	9.9 ± 3.5	3.78 ± 1.52	41.0	5.5	2.5	4.5	6.0	4.0	0.0
Liem 2011 <sup>12</sup>	Lap: 309	7.0 ± 0.2	2.5 ± 0.1	3.9	2.3	0.6				
	Open: 307	9.1 ± 0.2	3.7 ± 0.1	5.5	2.0	0.0	NA	NA	NA	NA

We used time to commencement of feeds and length of hospitalisation as measures of recovery time, and showed that the laparoscopic approach was comparable to the open. Our small study population is the likely reason why we were unable to demonstrate any clear benefits, though other larger series have shown shorter recovery times in the laparoscopic group.<sup>3,11,12</sup>

One patient in the laparoscopic group developed early bile leak that was minor and resolved on conservative management. Bile leak is a potentially serious complication that may necessitate re-operation or result in late strictures and its sequelae. However, reported rates of bile leak from laparoscopic series worldwide, ranging from 0.9% to 8.1%,<sup>3,5,7,12</sup> compares favourably with that of 2.0% to 7.3% in the open series.<sup>3,11-13</sup>

All patients in the laparoscopic group were complication-free on longer term follow-up. In contrast, 2 patients in the open group developed significant postoperative events: 1 with cholangitis and 1 with pancreatitis. Both cholangitis and pancreatitis are known complications following cyst excision. Postoperative cholangitis can be caused by bilio-enteric anastomotic strictures or the formation of intrahepatic stones, especially in Type 4a cysts with multiple intra-hepatic and extra-hepatic biliary tree dilatations.<sup>14,15</sup> Pancreatitis can be due to protein plug or calculi formation obstructing the pancreatic duct. This reflects the importance of ensuring complete cyst excision, particularly the intra-pancreatic portion of the cyst.<sup>16</sup> In this series, however, there were no biliary abnormalities on imaging surveillance to account for these occurrences. Both patients were conservatively treated and respectively remained asymptomatic for 5 to 6 years thereafter. Three patients in the open group also experienced adhesive colic, and 1 patient developed a complication directly related to the means of surgical access—a hypertrophic subcostal scar. Diao et al<sup>3</sup> similarly reported significantly decreased

early and mid-term morbidity from anastomotic strictures, pancreatitis, cholangitis and adhesive obstruction in laparoscopic excisions. The authors attributed this to the better visualisation and improved precision of the bilio-enteric anastomosis with laparoscopy.

In our early experience, 1 case was converted to open surgery due to hepatic duct stricture and the need for ductoplasty. With increased experience, laparoscopic hilar ductoplasty was performed in a subsequent case with proximal cyst extension. Other authors have recently demonstrated that similar laparoscopic hilar ductoplasty can be performed safely for strictures near the hepatic duct confluence.<sup>17</sup> Complex ductal anatomy need not necessitates open conversion with careful preoperative planning and meticulous technique.

Two patients in our laparoscopic series were infants, at 5 and 6 months of age. One was asymptomatic, while the other had obstructive jaundice. In both cases, operative times were shorter than the median operative times. It has been the authors' experience that in infants, surgical planes were easier. A shorter history associated with fewer prior inflammatory episodes is the likely reason. Others have similarly reported that in experienced hands, laparoscopic excision and reconstruction can be achieved even in the very young.<sup>3</sup>

One key disadvantage shown by our study was that laparoscopic excision entailed significantly longer operative times. Despite the potentially increased risks of prolonged anaesthesia and carbon dioxide pneumoperitoneum, we observed no adverse outcomes. There is a steep learning curve for choledochal cyst excisions, with some authors reporting an improvement of operative times only after the initial 30 to 35 procedures, and more recent cases with operative times not inferior to that of open surgery.<sup>3</sup> Centres with smaller patient volumes may thus take longer to achieve equivalent operating times to the open approach. But it is

our contention that where appropriate paediatric anaesthetic support is available, meticulous operative technique should not be traded off for shorter operative times if surgical outcomes are not to be compromised.

Longer operative times in laparoscopic choledochal cyst excision have triggered a revival of interest in hepatico-duodenostomy (HD) as an alternative to HJ. Without the Roux-en-Y jejunostomy, HD can be completed in a shorter time, whether open or laparoscopically.<sup>18,19</sup> Santore et al reported shorter operative times, faster recovery and fewer complications with HD, and argued that HD was the more physiological option.<sup>18</sup> However, the increased rates and implications of asymptomatic bile gastritis and the potential long-term carcinogenic risk of HD have yet to be ascertained.<sup>19-21</sup> For an infant or child undergoing surgery early in life, this is an important consideration. We contend that until data concerning long-term safety of HD is available, HJ, in spite of longer operative times, remains the proven option.

Robotic surgery, with articulating arms allowing for greater dexterity to aid intracorporeal suturing, is a new frontier in minimally-invasive surgery. Several authors in scattered case series have begun exploring robotic choledochal cyst surgery in a bid to reduce operating time.<sup>22</sup> However, its benefits will perhaps be truly realised when robotic equipment become sufficiently miniaturised for use in infants and small children, made more widely available, and more cost-friendly.

## Conclusion

In conclusion, our study shows that laparoscopic surgery is feasible, safe and enjoys excellent short- and mid-term outcomes in comparison with the open technique. This is achievable even in centres with low patient volumes. Young age and complex ductal anatomy need not be deterrents with careful preoperative planning and meticulous technique.

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