Challenges and strategies for single-incision laparoscopic Roux-en-Y hepaticojejunostomy in managing giant choledochal cysts

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ABSTRACT

Background/purpose: Giant choledochal cyst (CDC) is thought to be a challenge for one-stage single-incision laparoscopic hepaticojejunostomy (SILH). We herewith designed the strategies for SILH in surgical management of giant CDC children, and reported its outcomes.

Methods: Twenty-eight patients with giant CDCs successfully underwent SILH between April 2011 and October 2013. With guidance of an extra-long 5-mm 30°/C14 laparoscope, anterior cyst wall was punctured extra-corporeally using a 20-gauge angiocatheter. Cyst content was evacuated to create working space. A series of trans-abdominal retraction sutures were placed through serosa of gallbladder fundus, common hepatic duct and proximal to distal portion of anterior cyst wall to facilitate dissection. Cyst excision and hepaticojejunostomy was performed.

Results: Mean age at operation was 6.24 months. Mean operative time was 3.18 h, significantly shorter than 6.3 h in our historical group undergone conventional laparoscopic hepaticojejunostomy. No blood transfusion was required. Post-operative recovery was comparable to that of our historical CLH controls. Median follow-up period was 24 months. No mortality or morbidities of anastomotic stenosis, bile leak, cholangitis or pancreatic leak was observed. Liver function normalized post-operatively.

Conclusions: Following the strategy, SILH for giant CDCs is safe and one can achieve outcomes comparable to those of CLH in experienced hands.

1. Introduction

Recently, single-incision laparoscopic surgery has become popular in pediatric surgeons to achieve minimal surgical trauma [1-4]. We have adopted single-incision laparoscopic hepaticojejunostomy (SILH) strategy in CDC children [5]. However, the limitations of single-incision laparoscopic approach render the one-stage definitive surgery for giant choledochal cyst (CDC) more challenging because of 1) extremely limited working space; 2) limited exposure of displaced portal vein, hepatic artery, and surrounding tissues; and 3) difficult dissection for intra-pancreatic portion of CDC and posterior cyst wall. One or two stages open surgery with large painful muscle-cutting wound are usually adopted [6], which significantly increases the mortality and morbidities. We have modified our SILH techniques to treat children with giant CDCs. The current study is the first series to evaluate its safety and efficacy.

2. Methods

Giant CDC is defined as CDC with diameter greater than 10 cm [7,8]. Patients with giant CDCs (Fig. 1a and b, CDC diameter >10 cm) who underwent SILH between April 2011 and October 2013 were reviewed. All SILH procedures were performed by the same surgical team. Ethics approval from the Ethics Committee of Capital Institute of Pediatrics was obtained. Written informed consents were obtained from the parents of CDC patients prior to the study.

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Displacement of vessels, severe inflammatory adhesion, wide and deep dissection of intra-pancreatic portion of common bile duct increases the difficulty and risk to the vessels and surrounding tissues injuries. Preoperative computerized tomography and magnetic resonance cholangiopancreatogram images were carried out to identify the displaced portal vein, hepatic artery and adjacent structures (Fig. 1b), the ratio of intra-pancreatic portion to whole common bile duct, and cyst wall thickness.

Giant CDC is often accompanied by severe biliary obstruction, coagulopathy and consequently the increased the risk of perioperative hemorrhage. Preoperative correction of coagulopathy and postoperative anti-hemorrhage remedy are necessary. Vitamin K1 was routinely given. Fibrinogen and/or human prothrombin complex were administered perioperatively if FIB value was lower than 2 g/L and/or partial thromboplastin time was more than 10 s above upper normal limit.

The patients were placed in anti-Trendelenberg position. The operating surgeon stood on the foot of the table and the assistant surgeon stood on the patient’s left side holding the camera. A vertical umbilical skin incision was made. Huge CDC occupies large abdominal space and interferes operative field. Hence, the patients are susceptible to accidental tissue injury. We performed biliary decompression before working trocar insertion. With the guidance of an extra-long 5-mm 30° laparoscope (Fig. 2, 26046BA, Karl Storz GmbH & Co. KG, Tuttlingen, Germany), the anterior cyst wall was punctured extra-corporeally by 20-gauge angiocatheter which was connected to a suction tube (Fig. 3a). After intraoperative cholangiograms was completed, the cyst was evacuated by suction to create a working space (Fig. 3b and c). Two 3-mm conventional laparoscopic instruments were inserted through the working ports which were placed each side of the camera port at the ends of horizontal umbilical incision (Fig. 2). Carbon dioxide pneumoperitoneum was established at a pressure of 10 mmHg.

To facilitate dissection of giant CDC with distal cyst deeply embedded in pancreas, a series of retraction sutures were routinely placed through abdominal wall and 1) serosa of gallbladder fundus (this suture was used to retract right liver lobe after subserosal dissection of gallbladder); 2) proximal common hepatic duct; 3) mid- to distal portion of anterior cyst wall (Fig. 3d). Additional transabdominal retraction sutures were placed through falciform ligament, pons hepatis or left margin of incised hepatoduodenal ligament in patient with enlarged left liver lobe to expose medial lateral of cyst wall because portal vein and hepatic artery are situated medial-posteriorly to the cyst (Fig. 3d). The assistant adjusted the tension and direction of suture retraction extracorporeally to facilitate dissection and anastomosis. The cyst dissection was carried out close to the cyst wall from anterior-lateral to posterior-medial direction orientation direction using a 3-mm hook cautery.
Localized mucosectomy was performed in patients with occult perforations to prevent injury of adjacent structures caused by dense adhesions. The cysts were dissected out and incised at the distal ends. The distal stumps were left unligated because distal common bile ducts were stenotic in all patients with cystic type of CDC [9]. The proximal cyst was transected at the level of common hepatic duct (CHD) to identify CHD orifice by inspection, and identify portal vein and hepatic artery (Fig. 3e). Ductoplasties were performed in patients combined with hepatic duct strictures. The proximal common hepatic duct with a diameter greater than 1.0 cm was kept for anastomosis. An individualized jejunal Roux loop length was tailored according to the distance between the umbilicus and the hepatic hilum [10]. Extra-corporeal Roux loop formation was carried out via the enlarged umbilical wound. The hepato-jejunal anastomosis was carried out as previously described [5]. According to the diameter of anastomotic stoma, one double-armed 5/0 or 6/0 PDS (PDS®, Z148, Ethicon Inc., US) was divided into two 10–15 cm segments with a needle (Taper RB-2, 1/2 Circle, 13 mm, Ethalloy, Ethicon Inc., US) at each end. Hepaticojejunostomy was started at 6 o’clock position, i.e. mid-point of posterior wall (the knot of 2 trimmed short arms was placed at 6 o’clock position), and it was continued to 9 and 3 o’clock positions separately by running suture. The sutures were knotted at 9 and 3 o’clock positions simultaneously after completing the posterior wall anastomosis. The anterior wall anastomosis was performed using continuous suture along the 3–12–9 o’clock direction by the same PDS suture. The sutures were knotted at 9 o’clock position [5]. The diameter of the anastomosis was greater than 1.0 cm. The midline 5-mm incision for camera port insertion and two 3-mm incisions for working ports insertion were closed by 2/0 absorbable sutures separately. The mid-longitudinal umbilical skin incision was closed by 5/0 VICRYL RAPIDE suture. A drainage tube was placed.

According to the intervention protocol in our department, patients were routinely given antibiotics, Glycyrrhizin and Ursodeoxycholic acid postoperatively till the laboratory results returned to normal level.

![Fig. 3. Single-incision laparoscopic hepaticojejunostomy for giant choledochal cyst. 3a. The anterior cyst wall is punctured extra-corporeally with a 20-gauge angiocatheter. 3b. The cyst is evacuated by suction to create the working space. 3c. The working space is created after evacuation. 3d. A series of trans-abdominal retraction sutures to facilitate dissection. 3e. Posterior wall dissection.]
Patients were followed up in our clinic at postoperative 1, 2, 3, 6 months and every 6 months thereafter. The laboratory results, and findings of ultrasonographic and upper gastrointestinal studies were reviewed.

2.1. Statistic analysis

Data were analyzed with SPSS 13.0 package. Paired t-tests were applied to compare perioperative laboratory values in SILH group. P < 0.05 was considered to be statistically significant.

3. Results

We performed laparoscopic cyst excision and hepaticojejunostomy for choledochal cyst children without peritonitis or cyst perforation. In our series, all patients with giant choledochal cysts during the study period did not have peritonitis or complete cyst perforation. Hence, all of them were recruited to this study. Thirty patients with giant CDCs underwent SILH initially. Two patients (a 8.5 years old girl and a 9 years old boy) were converted to cyst excision and hepaticojejunostomy; 3) mid-portion of anterior cyst wall for exposure of hepatic hilum to facilitate CDC dissection and posterior cyst wall, which aggravated adhesion with portal vein and hepatic artery; 4) falciform ligament or pons hepaticus; 5) left margin of distal hepatic artery and require localized mucosectomies. Pathological results showed grade III and IV liver fibrosis in 7 of 28 (25%) patients, and hepatic cell damage in 12 of 28 (42.9%) patients.

The mean age at operation was 6.24 months (8 days–7.75 years), similar to 6.3 months (28 days–9 months) in our historical control group who underwent conventional laparoscopic hepaticojejunostomy (CLH, n = 7) [11]. The cyst diameter in both SILH group and historical CLH group were greater than 10 cm. The diameter of anastomotic stoma was 1–1.5 cm. With technical improvement, the mean operative time was significantly shorter than that of our historical CLH controls [11] (3.18 ± 0.73 h vs. 6.3 h). Intraoperative blood loss was minimal. No blood transfusion was required. Postoperative hospital stay and resumption of feed in SILH group were 10.56 months, range: 8 days–10.56 months). Twelve of them (42.9%) were diagnosed by prenatal ultrasonographic studies. Except for one asymptomatic prenatally diagnosed CDC patient, 25 patients (89.3%) presented jaundice and additional 2 patients (7.1%) presented vomiting. Coagulopathy was found in 10 of 28 (35.7%) patients, with the mean fibrinogen level of 1.34 ± 0.07 g/L. Human fibrinogen was given to correct their coagopathies. All CDC were cystic type. The cyst diameters and lengths were 10.2–11.5 cm and 10.5–13 cm respectively. Fifteen of 28 (53.6%) patients had additional intrahepatic duct dilatations. Four of 28 patients (14.3%) had additional common hepatic duct strictures, which were corrected by ductoplasties. Protein plugs/sludge were detected in all patients and were completely removed by saline irrigations. Twenty patients (71.4%) had mild adhesions, while 8 (28.6%) patients had severe adhesions. The pancreatic tissues grew into the cyst walls in additional common hepatic duct strictures, which were corrected in 28 patients (F/M: 20/8). With exception of one patient being 7.75 years old, the remaining 27 patients (96.4%) were young infants (mean age: 3.03 ± 2.16 months, range: 8 days–10.56 months). Twelve of them (42.9%) were diagnosed by prenatal ultrasonographic studies. Except for one asymptomatic prenatally diagnosed CDC patient, 25 patients (89.3%) presented jaundice and additional 2 patients (7.1%) presented vomiting. Coagulopathy was found in 10 of 28 (35.7%) patients, with the mean fibrinogen level of 1.34 ± 0.07 g/L. Human fibrinogen was given to correct their coagopathies. All CDC were cystic type. The cyst diameters and lengths were 10.2–11.5 cm and 10.5–13 cm respectively. Fifty of 28 (53.6%) patients had additional intrahepatic duct dilatations. Four of 28 patients (14.3%) had additional common hepatic duct strictures, which were corrected by ductoplasties. Protein plugs/sludge were detected in all patients and were completely removed by saline irrigations. Twenty patients (71.4%) had mild adhesions, while 8 (28.6%) patients had severe adhesions. The pancreatic tissues grew into the cyst walls in 4 of 28 (14.3%) patients, which obscured the border between common bile duct and pancreas and significantly increased the difficulty of dissection. Occult (self-sealed) perforation (3/28, 10.7%), including one old perforation covered by the adjacent tissues (3.6%) and 2 subserosal perforations (7%), were detected at posterior cyst wall, which aggravated adhesion with portal vein and hepatic artery and required localized mucosectomies. Pathological results showed grade III and IV liver fibrosis in 9 of 28 (32.1%) patients, grade I and II liver fibrosis in 7 of 28 (25%) patients, and hepatic cell damage in 12 of 28 (42.9%) patients.

The mean age at operation was 6.24 months (8 days–7.75 years), similar to 6.3 months (28 days–9 months) in our historical control group who underwent conventional laparoscopic hepaticojejunostomy (CLH, n = 7) [11]. The cyst diameter in both SILH group and historical CLH group were greater than 10 cm. The diameter of anastomotic stoma was 1–1.5 cm. With technical improvement, the mean operative time was significantly shorter than that of our historical CLH controls [11] (3.18 ± 0.73 h vs. 6.3 h). Intraoperative blood loss was minimal. No blood transfusion was required. Postoperative hospital stay and resumption of feed in SILH group were 6.03 ± 0.64 days and 2.50 ± 0.88 days respectively, which were comparable to those in our historical CLH controls [11]. Drainage tubes were kept for 4–8 days in 8 patients for ascites resulted from severe liver damage and excessive exudations from wide dissection area. The median follow-up period was 24 months (1–50 months). No mortality or morbidities of anastomotic stenosis, bile leak, intrahepatic reflux, cholangitis, pancreatic leak, pancreatitis, intestinal obstruction, wound infection, injury of intra-abdominal organs or incisional hernia was observed. Coagulation function in those with coagulopathies normalized after surgery (average postoperative fibrinogen level: 3.15 ± 0.57 g/L). Liver function parameters reversed to normal level within 1 year after operation (Table 1, p < 0.001). No appreciable scar was detected at 1 month follow-up (Fig. 4).

4. Discussion

Giant CDC occupies large intra-abdominal space and displaces adjacent structures. Limitation of single-incision laparoscopic surgery [2], including trocar crowding, limited intra-abdominal exposure, clashing of the instruments, and limited number of working ports further increases difficulties of one-stage definitive surgery for giant CDC. We herewith formulated the following strategies for single-incision laparoscopic cyst excision and hepaticojejunostomy in treatment of giant CDCs, and reported its outcomes.

4.1. Working space creation

Giant CDC often crosses the mid-line and almost reaches to pelvic cavity. To avoid accidental tissue injury during working port insertion, we placed camera port through vertical umbilical incision under the direct vision. With guidance of telescope, the cyst was extra-corporally punctured with a 20 G angiocatheter. It was connected to a suction tube. The cyst contents were evacuated till adequate working space was established.

4.2. Exposure

A series of trans-abdominal suture retractions were placed through 1) serosa of gallbladder fundus for cephalad retraction of right liver lobe; 2) anterior wall of proximal common hepatic duct for exposure of hepatic hilum to facilitate CDC dissection and hepaticojejunostomy; 3) mid-portion of anterior cyst wall to distal CDC to posterior cyst wall for exposure of intra-pancreatic segment, portal vein and hepatic artery to facilitate CDC dissection and prevent vessel injury; 4) falciform ligament or pons hepaticus and anchored at left costal margin of anterior line axillary for cephalad retraction of enlarged left liver lobe; 5) left margin of incised hepatoduodenal ligament and anchored at left costal margin of anterior line axillary for exposure medial lateral of CDC to avoid vessel injury. Flexible utilization of suture retractions, especially sequential suture retractions along CDC dissection direction effectively substitutes the role of the first assistant in conventional 4-port laparoscopic surgery. Furthermore, the assistant adjusted

### Table 1

<table>
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<tr>
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<th>Pre-operation</th>
<th>Post-operative 12 month P</th>
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<tbody>
<tr>
<td>ALT (U/L) Ref</td>
<td>&lt;40</td>
<td>195.45 ± 94.34</td>
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<tr>
<td>AST (U/L) Ref</td>
<td>&lt;40</td>
<td>190.23 ± 87.26</td>
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<tr>
<td>ALP (U/L) Ref</td>
<td>&lt;400</td>
<td>461.19 ± 269.62</td>
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<tr>
<td>GGT (U/L) Ref</td>
<td>7–50</td>
<td>524.45 ± 366.61</td>
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<tr>
<td>TIBL (μmol/L) Ref</td>
<td>3.4–20</td>
<td>108.57 ± 73.43</td>
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<tr>
<td>DBIL (μmol/L) Ref</td>
<td>1.7–13.2</td>
<td>63.93 ± 40.97</td>
</tr>
<tr>
<td>SAMY (U/L) Ref</td>
<td>25–125</td>
<td>93.78 ± 87.52</td>
</tr>
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NB: ALT: alanine transaminase; AST: aspartate aminotransferase; ALP: alkaline phosphatase; GGT: γ-glutamyl transpeptidase; TIBL: total bilirubin; DBIL: direct bilirubin; SAMY: serum amylase.
4.3. Dissection

In our series, dissection difficulties in children with giant CDCs resulted from 1) inflammatory adhesions (8/28, 28.6%); 2) occult perforation (3/28, 10.7%) at posterior cyst wall aggravated the adhesion with portal vein and hepatic artery; 3) large interface between intra-pancreatic segment of CDC and pancreas requiring wide and deep dissection; 4) in-growth of pancreatic tissue into the cyst wall obscured the border between common bile duct and pancreas (4/28, 14.3%). We established the following rules to ensure successful procedure minimizing accidental injury: 1) very careful dissection close to the cyst wall. Even if the cyst wall is accidentally ruptured during dissection, it can be used as guidance to differentiate CDC and surrounding tissues, hence to avoid incidental injury; 2) transecting the proximal CDC to i) identify the orifice of common hepatic duct (CHD) under direct vision. Severe CHD dilatation obscures the border of CHD and CDC. Intraoperative cholangiogram usually requires much larger volume of contrast agent to achieve comparable results to outer observation or intraoperative cholangiogram. Accidental injury of CHD, left or right hepatic duct has been reported in laparoscopic-assisted giant CDC dissection [6]. Traversing the proximal CDC allows observation of CHD outlet. Dissection is then performed along the rim of CHD orifice to prevent CHD injury; and ii) clearly detect portal vein and hepatic artery from transection plane, and dissect posterior cyst wall bi-directionally, i.e. proximal → distal and distal → proximal directions, to narrow the difficult dissection area gradually; 3) adoption of localized Lilly’s procedure, i.e. mucosectomy, in the area with occult perforation because dense adhesion significantly increases risk of injury of portal vein and hepatic artery. Mucosectomy has been proven not to increase the morbidity of cancer in CDC patients [7]; 4) leaving stump of distal CDC unligated cause all giant CDC were stenotic subtypes. None of our patients encountered postoperative pancreatic fluid leak. Previous literature reported 5 of 112 (4.5%) CDC children suffered injuries of pancreatic duct from excessive distal dissections [12]. Four of them had large or giant CDCs (diameter: 7 cm, 8 cm, 12 cm and 11 cm; length: 10 cm, 16 cm, 15 cm and 12 cm respectively) [12]. Our management strategy of distal stump effectively prevents injury of pancreatic duct [9].

4.4. Bleeding control

Peri-operative hemorrhage in giant CDCs was mainly attributed to 1) vessel injury induced by dense adhesion or vessels displacement, 2) oozing caused by severe inflammation or broad dissection of intra-pancreatic portion, and 3) coagulopathy resulted from liver damage. Preoperative computerized tomography and magnetic resonance cholangiopancreatogram scans are valuable to map the displaced vessels. A series of suture retractions, dissection along the anterior-lateral wall → distal end → posterior-medial wall direction, i.e. from non-vascular region to vascular region, transection from proximal CDC to detect portal vein and hepatic artery directly, localized mucosectomy in posterior cyst wall with occult perforation are helpful to decrease the risk of vessel injury. Peri-operative anti-hemorrhage management is necessary. Coagulopathy should be corrected preoperatively by administration of Vitamin K1, fibrinogen, human prothrombin complex, and plasma.

4.5. Drainage tube placement

Drainage was necessary for children with giant CDCs because of excessive fluid exudation from broad dissection area, inflammatory oozing, and ascites caused by liver dysfunction. Human SeroAlbumin was given based on serum albumin monitoring results.

Following the strategy, one-stage single-incision laparoscopic cyst excision and hepaticojejunostomy was safely performed in children with giant CDCs in experienced hands. With technical improvement, the average operative time of SILH was significantly shortened compared to that in our historical CLH data [11]. No blood transfusion was required. Postoperative recovery was comparable to those in our historical controls. So far, no intraoperative and postoperative complications in SILH were encountered. The outcome was comparable to that in historic CLH groups. The postoperative pain was minimal. There was no requirement of additional analgesics. Cosmetic outcomes were superior to open and conventional 4-port laparoscopic hepaticojejunostomy, and achieved “virtually scarless wound”.

The advantages include: 1) SILH is carried out through the umbilical incision, which used for gallbladder and choledochal cyst removal and extracorporeal Roux loop formation in conventional laparoscopic cyst excision and hepaticojejunostomy; 2) SILH can be completed by conventional straight laparoscopic instruments. It does not require the expensive and specially-designed instruments and specific training. Thus allows this technique to be applied in each laparoscopic center; 3) the operative time and postoperative morbidities of SILH are comparable to those of conventional laparoscopic hepaticojejunostomy [5,13]; 4) SILH is more suitable for children because their immature muscle and fascial layer of abdominal wall allow more freedom of instrument movements, and the relatively small operating field in hepatic hilum facilitates SILH with straight instruments.

Our result shows that most neonatal biliary cystic lesions are detected prenatally and majority of them presented persistent jaundice after birth. For those with atypical symptoms, such as vomiting, crying, insomnia, abdominal distention, parents should consult with specialists and perform ultrasonographic study as early as possible. Giant CDC indicates severe biliary obstruction. Pathological results verified that they had similar extent of hepatic damages to some age-matched biliary atresia patients. Although
the liver damage is reversed eventually, it usually requires half to one year for complete recovery. Hence, early one-stage definitive surgery is strongly advocated, i.e. the progression of liver damage may be similar to that of unoperated biliary atresia patients. It avoids external biliary drainage induced electrolyte and fluid deficits and patient's suffering. Proper surgical strategy ensures safe surgery and effectively prevents the development of progressive hepatic lesions. Surely, the surgical approach depends on patient’s condition. Conversion to open approach should not be considered as failure.

Admittedly, the current study suffers from the short-comings of small size and relatively short follow-up period. Long-term follow up in a larger scale cohort is warranted to compare the efficacy between SILH and CLH for giant CDCs.

5. Conclusions

With case accumulation and technical improvement, SILH is safe and feasible for majority of giant CDC children. Young infants with shorter clinical histories and less adhesion (71.4% in our series) are more suitable for this procedure. Its results are comparable to CLH for giant CDCs. SILH potentially provides a viable option as a surgical treatment of giant CDCs.

Disclosure of funding

Dr. Mei DIAO, Prof. Long LI, Dr. Qi LI, Dr. Mao YE and Prof. Wei CHENG declare no conflicts of interest or financial ties to disclose.

Conflicts of interest

No conflicts of interest to disclose.

Ethical approval

The current research obtained the Ethical Approval from Ethical Committee of Capital Institute of Pediatrics (Reference number: 2011-A06).

Author contribution

Prof Long LI contributed to study design and writing.

Prof Wei CHENG contributed to study design and manuscript writing.

Dr. Mei DIAO contributed to study design, data collections, data analysis, and writing.

Dr. Qi LI contributed to data collections.

Dr. Mao YE contributed to data collections.

References