Review Article



# Pediatric minimal invasive surgery—bile duct diseases

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**Abstract:** The laparoscopic surgery brings a new horizon to conventional treatments. In this article, we reviewed the minimal invasive surgeries in the treatment of bile duct diseases in children. The results demonstrated that laparoscopic treatments for bile duct diseases in children are safe, and can achieve comparable or superior surgical outcomes as those of open surgery.

**Keywords:** Minimal invasive surgery; laparoscopy; bile duct disease; children

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With the development of laparoscopy, surgical treatment for bile duct diseases enters into a new era. The magnified view and agile observation perspective provided by telescope update the knowledge of diseases, associated anomalies, and anatomical variations, improve the techniques, and facilitate individualized surgical strategies. The advantages includes: (I) it minimizes the surgical trauma, reduces complications, hence accelerates postoperative recovery; (II) umbilical-to-hepatic hilum observation provided by telescope is beneficial for maneuver at hepatic hilum; (III) magnified view facilitates precise dissection and anastomosis; (IV) the wide vision field allows observation of whole abdominal cavity, the laparoscope and/or cystoscope can inspect intrahepatic duct, common channel, pancreatic duct, hence resolve multiple anomalies with the same setting.

The laparoscopic techniques increasingly applied in children with the following bile duct diseases.

## Choledochal cysts (CDC)

### *Evolution of laparoscopic cyst excision and Roux-Y hepaticojejunostomy*

The laparoscopic CDC excision and hepaticojejunostomy was started since 1995 (1). A few small series of 1–3 CDC children were reported afterwards (2-8). Our center firstly published a larger series (n=35) in 2004 (9). Laparoscopic cyst excision and hepaticojejunostomy becomes an alternative surgical treatment for CDC children. Since 2011, we firstly started single-incision laparoscopic cyst excision and hepaticojejunostomy (SILH) (10,11), and extended this technique to complicated cases (12-15). To date, 1401 CDC children successfully underwent SILHs in our center.

SILH is described as followings. A 2–2.5 cm umbilical longitudinal incision is made and stretched horizontally. A 5-mm telescope is placed at the midline of incision. Two 3-mm conventional laparoscopic instruments are placed at the ends of umbilical incision bilaterally. A series of trans-abdominal retraction sutures are placed through serosa of gallbladder fundus, proximal common hepatic duct (CHD), proximal to distal CDC to facilitate dissection and anastomosis (*Figure 1*).

The laparoscopic cyst excision and hepaticojejunostomy is performed according to different subtypes (16,17). In patient with stenotic distal common bile duct, dissection is carried out along lateral-anterior-distal-posterior direction. The distal CDC is transected at the level of stenotic segment. Distal stump ligation is optional to avoid pancreatic duct injury induced by excessive dissection. Ten percent of this subgroup associated hepatic duct stricture,

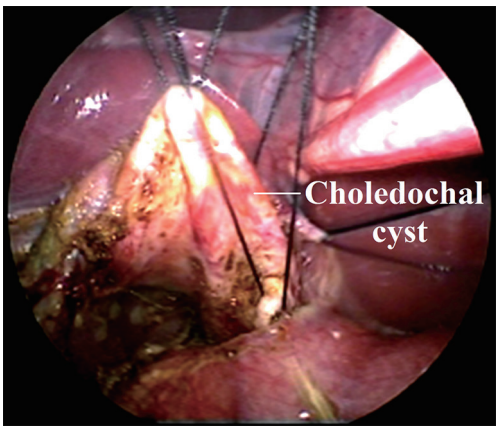


**Figure 1** Single-incision laparoscopic surgery for choledochal cyst.

requiring surgical correction. Intrahepatic ducts are inspected and irrigated by normal saline to remove stone debris. Thus, prevents post-operative stone formation and cholangitis. In patient with non-stenotic distal common bile duct, the cyst was transected at middle segment. Dissection was conducted circumferentially and continued distally to proximal biliary-pancreatic junction to prevent pancreatic duct injury. Distal stump should be ligated to avoid pancreatic leak. In this subgroup, 62.3% patients combined with protein plugs in common channel. Hence, clearance of protein plugs is necessary to prevent pancreatitis.

The advantages of SILH are (11):

(I) Trans-umbilical approach through the access that the gallbladder and cyst removal and Roux loop establishment;

(II) Using conventional laparoscopic instruments saves the cost of expensive curved instruments and special training. Hence, this technique can be promoted in each laparoscopic center;

(III) Similar operative time and post-operative complication rate compared with conventional laparoscopic hepaticojejunostomy;

(IV) Immature muscle and fascial layer and relatively small surgical field in children allows more freedom of instrument movements and facilitates SILH with straight instruments.

Hence, SILH can be a viable option as a surgical treatment of CDC.

Robotic-assisted cyst excision and hepaticojejunostomy has been utilized in clinical practice since 2006. The 3D visualization and the additional degrees of freedom are major advantages. One camera port, three robotic instrument ports and one assistant port are required. The mean operative time is significantly longer than that of conventional laparoscopic procedures (18-23). Postoperative bile leak was reported (18). However, larger size, repeated launches of robotic system in different steps of one operation, lack of haptics, increased hospital cost restricts its application in CDC treatment. Recently, a hybrid of conventional laparoscopy and robotic technique, i.e., using robotic system in the CDC dissection and hepaticojejunostomy and conventional laparoscopy in the rest surgical steps, is adopted to avoid repeated launches of robotic platform. It maximizes the advantages of two approaches, minimizes the difficulty of robotic manipulation, and shortens operative time.

With the technical improvement, refined transumbilical robotic system and artificial intelligence will provide an optimal alternative to achieve the advanced goal of minimal invasive surgery, i.e., scarless, minimal surgical trauma, and steep learning curve.

### *General principles in laparoscopic hepaticojejunostomy*

#### Individualized short Roux loop

Conventionally, adult's standard 35–40 cm Roux loop is applied in CDC children. With increased age, the small bowel is significantly lengthened. A redundant Roux loop is prone to induce intestinal obstruction requiring redo surgery (24), bile stasis (25), bacterial overgrowth (26), cholangitis (26), stone formation, and mal-absorption of fat and fat-soluble vitamins (27). The Roux loop was tailored in our series according to the distance between umbilicus and hepatic hilum. The comparison study indicated that the individualized short Roux loop provides a comparable antireflux effect (28), and prevents Roux loop rotation and necrosis.

#### Laparoscopic clearance of protein plugs in the common channel

Post-operative pancreatic stone formation and pancreatitis are attributed to unresolved protein plugs/calculi in the common channels (29,30). In the era of open surgery, inspection and irrigation of the common channel are difficult, and increase the risk pancreatic duct injury (31).

We utilized laparoscopic-assisted irrigation through a catheter (32). Repeated intra-operative cholangiogram via the catheter can verify the clearance (32). The liver function and serum amylase in all the patients normalized in long-term follow-up (32). No pancreatic leak occurred

 

(32,33). Our outcomes demonstrate that the catheter irrigation provides a comparable effect compared to flexible endoscope irrigation. The fine-caliber catheter is suitable for neonates and infants. It is an alternative option for protein plug removal in the centers where the flexible endoscopes are not available.

**Management of single/multiple hepatic duct strictures**

Our large-scale series verifies associated hepatic duct strictures in 11% CDC children (33,34), which often results in cholestasis, intrahepatic stone formation, and cholangitis. As an alternative of choledochoscope, laparoscope can be inserted into intrahepatic bile duct for careful inspection. The optimal proximal CDC resection level could be easily decided without hepatic duct injury or cyst remaining (35). The ductoplasty is carried out as followings.

Single hepatic duct stricture: the anterior wall of common/left/right hepatic duct is split. Ductoplasty and wide hepaticojejunostomy are carried out.

Multiple hepatic duct strictures: the stenotic segments should be split to the proximal dilated bile duct. After remove the intrahepatic duct calculi, a wide hepaticojejunostomy is conducted at this level to prevent post-operative biliary obstruction (33,34). No intrahepatic duct stone formation or cholangitis are developed in our mid- to long-term follow-up.

**Management of aberrant hepatic duct (AHD)**

Improper management of AHD often leads to bile leak or biloma after CDC operation, requiring further surgery (36-38). Recognizing different subtypes of AHDs and treating them accordingly is effective to prevent relevant complications.

We categorize AHD into 4 subtypes, and treat them individually:

(I) Type 1: AHD locates close to the conjunction of cystic duct and CHD. The AHD and CHD are combined as one stoma or sutured along their lateral walls to form an anastomotic stoma;

(II) Type 2: AHD locates in the mid-portion of cystic duct. Anastomosis of AHD and CHD to jejunum are carried out separately;

(III) Type 3: duplication of cystic duct. The duplicated cystic duct is ligated before being divided to prevent bile leak;

(IV) Type 4: associated with aberrant right hepatic artery (RHA). In case of RHA anteriorly compressing CHD and AHD (type 4a). Aberrant RHA is repositioned behind. The AHD and CHD are combined as one anastomotic stoma. In case of RHA anteriorly compressing CHD alone, the connection between AHD and CHD is transected (type 4b). After repositioning the RHA behind CHD, the lateral walls of AHD and CHD are sutured to form an anastomotic stoma.

In our series, none of patients developed anastomotic stricture, cholangitis, or bile leak. Postoperative liver function tests and serum amylase level normalized within 1 year. Individualized laparoscopic ductoplasty and hepaticojejunostomy provides an efficacious surgical option for CDC children with AHDs.

**Management of aberrant RHA**

We firstly found that the aberrant RHA anteriorly compressing proximal CHD is a major factor attributed to the postoperative biliary re-obstruction (27%). To prevent postoperative biliary obstruction, the aberrant RHA is carefully mobilized from the proximal CHD, and repositioned behind the proximal CHD (39-41).

To prevent RHA injury, the assistant pulls up a retraction suture through proximal CHD to increase the space between RHA and CHD in dissection, and expose posterior wall and repositioned RHA in anastomosis.

### *Laparoscopic application in complicated CDCs*

**Neonatal CDCs**

With the advents of ultrasonographic screenings, perinatally diagnosed CDCs significantly increase (42). In our center, the percentage of prenatally diagnosed CDC increased from 16% (2001 to 2010) to 37.7% (in 2017). Conventionally, postponed definitive surgery is postponed for 6 months because of difficulties of anaesthesia and surgery. In era of open surgery, anastomotic stricture and leakage have been reported (43,44). For neonates with giant CDCs, staged surgeries with external biliary drainages are utilized.

However, ultrasonographic studies found a large amount of sludge in CDC at birth in majority of antenatally diagnosed asymptomatic CDC patients (45,46). The accumulation of sludge often results in severe distal obstruction, even perforation (47). We randomized prenatally diagnosed CDC into early and late operation groups, i.e., underwent definitive surgery within and after the first month of life. The biliary obstruction and liver damage in the late operation group were significantly severer, and was similar to that in cystic biliary atresia. Liver

 

fibrosis rate was significantly higher in the late operation group (45). Perforation and coagulopathy, including subdural hemorrhage requiring craniectomy frequently occurred and severer in infants younger than 1 year old (48-52). While, staged surgery often induced dehydration, electrolyte imbalance, infection, adhesion induced incidental injury. Hence, early definitive surgery in neonatal period is advocated.

Laparoscope with magnified vision and finer instruments increases the accuracy of anastomosis. It minimizes the anastomotic leakage and stricture, which are the main concerns in open surgery (43). Hence, more and more CDC neonates or antenatally diagnosed CDC patients undergo definitive surgery in the neonatal periods. In our series, the youngest CDC patients (cyst diameter: 10 cm) successfully underwent one-stage cyst excision and hepaticojejunostomy at Day 3 after birth, and recovered uneventfully.

**Giant CDCs**

Giant CDC occupies large space in abdominal cavity, particularly in neonates and younger infants. It significantly increases the difficulty and risk of incidental injury. Traditionally, it is thought to be a contra-indication of one-stage laparoscopy definitive surgery.

To create adequate working space, we puncture the cyst with a 20 G angiocatheter and evacuate the contents (13). A series of retraction sutures were placed from proximal to distal CBD to facilitate distal and posterior wall dissections (13). Severe CHD dilatation in these patients obscures the border of CHD and CDC. Transecting the proximal CDC to (I) identify the CHD orifice under direct vision to prevent injury of hepatic ducts; (II) clearly detect portal vein and hepatic artery from transection plane, and dissect posterior wall bi-directionally to gradually minimize the difficult dissection area (13).

**Perforated CDCs**

Conventionally, perforated CDC is thought to be a contraindication for laparoscopic treatment because of adhesions, deranged anatomy, and demanding techniques. Two-stage surgery is usually adopted. The one-stage definitive surgery via open approach has been utilized in case reports with selected patients (53,54). An attempt of laparoscopic one-stage cyst excision and hepaticojejunostomy was made for perforated CDC recently (55).

In our practice, placing a series of retraction suture and adjusting the tension and direction of suture retraction are helpful to facilitate dissection, particularly when severe inflammation obscures the margin between perforated site and surrounding tissues (15). The electro-hook and forceps are alternately used in cyst dissection. The forceps have a superior hemostasis effect for actively bleeding from intramural vessels of cyst wall (15). In case that severe inflammatory cyst wall was too fragile to place the retraction suture, blunt dissection is adopted. The intrapancreatic dissection area is closed by a double layer running suture to minimize oozing (15). The anterior cyst wall is incised to differentiate the border between perforated site and surrounding tissues in direct vision. Localized mucosectomy in the perforated area is recommended to prevent the injuries of surrounding tissues (15). In some patients, the perforated site sealed with surrounding tissues and forms a bile pseudocyst. The pseudocyst should be distinguished from CDC. Pseudocyst fenestration is recommended to prevent iatrogenic injury of normal anatomical structures (15).

**Redo surgery**

We categorize the cause of post-operative biliary re-obstruction into 2 groups: congenital and technical. Congenital factors include aberrant RHA anteriorly compressing proximal CHD, and unsolved single or multiple hepatic duct strictures. Technical factor is anastomotic stricture (14).

In our series, 27% re-obstruction originates from aberrant RHA. After dissection of adhesions, a series of sutures are placed through gallbladder fossa for liver retraction. The second suture is placed through anterior wall of proximal CHD to facilitate dissection and redo anastomosis. The vascular pulse should be noticed during dissection, particularly when adhesions cover the RHA and obscure vascular pulse. Aberrant RHA is carefully mobilized and repositioned behind CHD. Intrahepatic duct stones are removed. Ductoplasties and redo hepaticojejunostomy are carried out (14).

Thirty percent re-obstruction results from unrecognized single or multiple hepatic duct strictures. The stenotic segments should be split to the proximal dilated bile duct. A wide hepaticojejunostomy is then performed at this level to prevent biliary re-obstruction (14).

Forty-three percent re-obstructions are attributed to anastomotic stricture. Ductoplasty and redo hepaticojejunostomy are conducted (14).

***Long-term follow-up results***

Our large series demonstrated that after the learning curve,

 

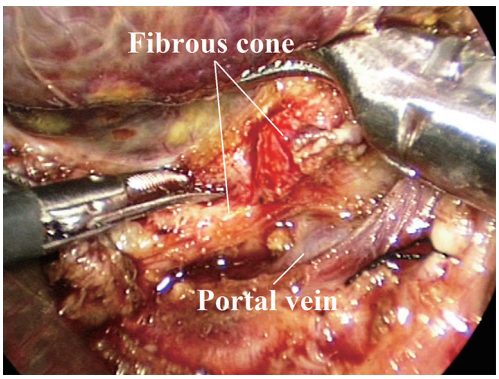


**Figure 2** Laparoscopic Kasai portoenterostomy for type III biliary atresia.

the operative time in laparoscopic group significantly decreased, and comparable to that in the open procedure (3.04 hours of laparoscopic group *vs.* 2.95 hours of open group, P=0.56) (33). By now, 1,855 CDC children successfully underwent laparoscopic definitive surgeries in our center. The laparoscopic definitive surgery could be accomplished within 1.5 hours for uncomplicated CDCs, and 3 hours for complicated CDCs. So far, 115 CDC patients have been followed up over 10 years. Compared with data in open group in previous reports and our series, patients in laparoscopic group have faster recovery, superior cosmetic outcomes, and lower morbidities (33). The results are accorded with those in Vietnam report (56). The application of laparoscope significantly decreases the morbidity in the era of open surgery. It attributed to meticulous dissection and anastomosis under the magnified view provided by telescope, technical improvement, and an accumulation of experience.

## Biliary atresia

The biliary atresia is treated according to different subtypes (12): (I) correctable biliary atresia with proximal CHD diameter ≥1 cm: hepaticojejunostomy; (II) uncorrectable biliary atresia, including type I and II biliary atresia with proximal CHD diameter <1 cm, and type III biliary atresia: Kasai portoenterostomy.

Laparoscopic cyst excision and hepaticojejunostomy for correctable biliary atresia are similar to those in CDC (12). Laparoscopic Kasai portoenterostomy are described in previous studies (*Figure 2*) (57-63). The fibrous cone is recommended to be dissected by a 3-mm laparoscopic scissors because the electro-cautery or ultrasonic scalpel may destroy the small bile duct, subsequently affect post-operative biliary drainage.

The outcomes of laparoscopic treatment for type III biliary atresia are debatable to date. Our short- to intermediate-term results of laparoscopic group (n=48), including jaundice clearance and native liver survival rates, were comparable to those of open group (n=47) (57). The operative time of laparoscopic group was longer than that of the open group (57). These results are similar to larger series reports from Japan (n=22) (58) and mainland China (n=80 and n=49 respectively) (59,60). While, small series from Germany (n=12) (61), Hong Kong (n=9, n=11 respectively) (62,63) showed that the native liver survival rates of laparoscopic group were lower than that of open group.

$CO_2$ pneumoperitoneum is assumed to contribute to the poor results of laparoscopic Kasai procedure for biliary atresia patients with liver dysfunction and fibrosis because it temporarily altered the metabolism and function of hepatic macrophages (61). However, $CO_2$ pneumoperitoneum does not affect the results of CDC children with liver dysfunctions and/or hepatic fibrosis who undergo laparoscopic surgeries. Poor results in previous small series of laparoscopic Kasai portoenterostomy may result from (I) difficult portal plate dissection, hemostasis, and anastomosis. Large case accumulation is required to improve techniques. Recent study reviewed outcomes of 80 patients with type III biliary atresia who underwent laparoscopic Kasai procedures. They divided the learning curve into 4 phases, each included 20 patients. After of the first 40 patients, the operative time and blood loss were remarkably reduced, and jaundice clearance and 2 years native liver survival rate remarkably increased (59); and (II) biliary atresia classification. There is no comparative study to evaluate the efficiency of laparoscopic versus open surgery in different subtypes of biliary atresia. A large sample RCT study with long-term follow-up is warranted.

## Solitary liver cyst/cystic dilatation of main hepatic duct

Solitary hepatic cysts with biliary communications (HC) and cystic dilatations of the main intrahepatic ducts (CIHD) are often associated biliary obstruction, causing cholestasis, stone formation, cholangitis, liver damage, and carcinoma in adulthood (64-66). Conventionally, Roux-Y cysto-jejunostomy is adopted. However, this procedure disrupts major biliary system (64,65). The bacteria migration increases the risk of cholangitis (67,68). Post-operative

 

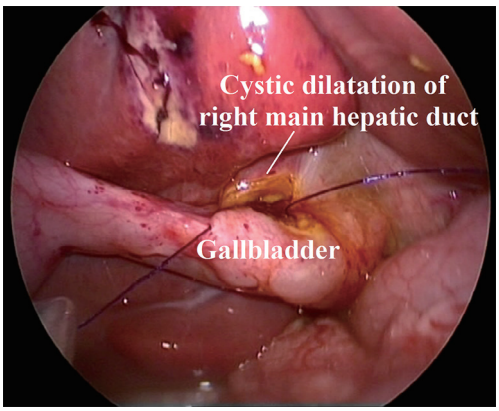


**Figure 3** Laparoscopic cysto-cholecystostomy for cystic dilatations of right main hepatic duct.

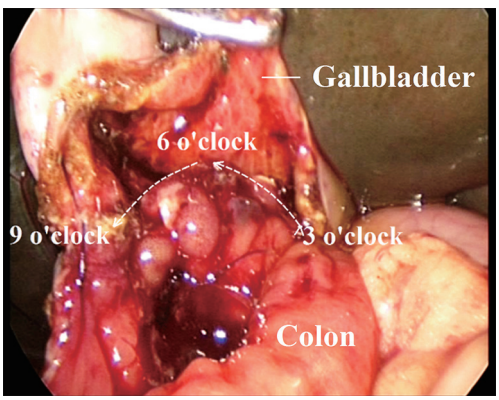


**Figure 4** Laparoscopic cholecystocolostomy for progressive familial intrahepatic cholestasis.

morbidities include anastomotic leak (67,68), anastomotic stenosis (69), hemorrhage (67), wound infection (67,68), intra-abdominal abscess (67,68), and Roux loop necrosis (70). We first employed laparoscopic cysto-cholecystostomy for children who have HC with biliary communication or CIHD (n=20 respectively) (71).

The cysts are dissected out and de-roofed. The orifice of intrahepatic ducts in HC can be detected by telescope. A longitudinal incision is made on the gallbladder based on the caliber of the de-roofed cysts. A side-to-side cysto-cholecystostomy is carried out (*Figure 3*) (71).

No bile leak, anastomotic stenosis, stone formation or cholangitis was detected in the intermediate-term follow-up (71). Liver function reversed to normal level after surgery (71). Laparoscopic cysto-cholecystostomy provides a safe, simpler, less disruptive and more physiological biliary drainage for HC with biliary communication and CIHD in children.

## Progressive familial intrahepatic cholestasis (PFIC)

PFIC usually induces hepatic cirrhosis early childhood. Surgical treatments, including partial external biliary diversion (72,73), ileoileal bypass procedure (74), partial internal biliary diversion (75), and liver transplantation (76) are often associated chronic diarrhea, cholangitis, or lifelong immuno-suppression. Furthermore, recurrence is a major concern even after liver transplantation (77). We herewith developed a cholecystocolostomy with anti-reflux Y-loop. It diverts bile from the gallbladder to the descending colon (78).

The transverse colon is transected proximal to splenic flexure. To establish the Y-loop, an end-to-side anastomosis is carried out between the distal transverse colon and mid-descending colon extracorporeally. The Y-loop is individually tailored based on the distance between the umbilicus and the gallbladder. With assistance of laparoscope, a longitudinal incision is made on the gallbladder, and end-to-side cholecystocolostomy is performed. The diameter of the anastomosis ranged from 2.5 to 3.0 cm (*Figure 4*) (78).

The mean operative time was 2.02 hours. All patients were jaundice free after 7 to 20 days and pruritus subsided in 3 to 14 days. Liver function parameters significantly improved postoperatively. Success rate (normalization of serum bile acids at postoperative 12 months) (79) was 85%. No mortality, diarrhea, cholangitis, or intrahepatic reflux was observed.

A cholecystocolostomy with anti-reflux Y-loop simplifies the surgery, remains the integrity of the small intestine, decreases post-operative complication rate, and offers comparable even superior results with previous surgical interventions (80).

## Symptomatic gallstone disease, hemolytic diseases, and biliary dyskinesia

Laparoscopic cholecystectomy is adopted in children with symptomatic gallstone disease, hemolytic diseases, and biliary dyskinesia (81,82). A review of 20,246 patients who underwent laparoscopic cholecystectomies showed that after learning curve, the postoperative recovery and cosmetic outcomes were superior to those in open cholecystectomies (83).

## Conclusions

In summary, laparoscopic treatments for bile duct diseases in children are safe. The outcomes are comparable or superior to those of open surgical procedures. Both patients and pediatric surgeons have benefited from these revolutionary techniques.

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*Ethical Statement:* The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.



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