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NEW TECHNOLOGY

Laparoscopic management for aberrant hepatic duct in children with choledochal cysts

Mei Diao[1] · **Long Li**[1] · **Wei Cheng**[2]

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Abstract

Background The aim of the current study is to evaluate efficacy of laparoscopic treatment for aberrant hepatic duct (AHD) in children with cholecochal cysts (CDC).

Methods CDC children with AHDs who successfully underwent laparoscopic ductoplasties and hepaticojejunostomies between October 2001 and October 2017 were reviewed. The AHD variations were categorized into four subtypes and the surgical management varied according the subtypes.

Results Sixty CDC patients with AHDs were reviewed. The mean age at surgery was 3.91 years. Two patients with Type 2 anomaly developed bile leaks after primary surgeries, and underwent laparoscopic anastomosis of AHD to jejunum in redo surgeries. In the remaining 58 patients, the average operative time was 3.75 h. The mean postoperative hospital stay was 6.02 days. The mean duration for full diet resumption was 2.25 days. The mean drainage time was 4.05 days. The median follow-up period was 30 months. Two patients with giant cysts had fluid collections, and were cured by drainages. One patient encountered duodenal injury at perforation site, and underwent laparoscopic repair. None of the patients had anastomotic stenosis, bile leak, cholangitis, intrahepatic reflux, pancreatic leak, pancreatitis, Roux-loop obstruction, or adhesive intestinal obstruction. Postoperative liver function tests and serum amylase level normalized within 1 year.

Conclusions Recognition and treatment based on different subtypes of AHDs effectively prevent relevant complications. Individualized laparoscopic ductoplasty and hepaticojejunostomy is an efficacious management for AHDs in CDC children.

Keywords Aberrant hepatic duct · Ductoplasty · Single-incision laparoscopy · Choledochal cysts · Children

The aberrant hepatic duct (AHD) is an extra- or sub-segmental bile duct that often drains a part of the liver and can join the biliary system at any level, such as at the cystic duct, the gall bladder, the right hepatic duct, the common hepatic duct (CHD), or the common bile duct [1–6]. Improper management of AHD often leads to bile leak or biloma after cholecochal cyst (CDC) operation, requiring further surgery [1–3].

We treated CDC children with AHDs laparoscopically according to different AHD subtypes. To the best of our knowledge, the current series is the first large study to assess the efficacy of individualized laparoscopic ductoplasty for CDC children with AHDs.

Materials and methods

Between October 2001 and October 2017, all CDC children with AHDs underwent laparoscopic operations by the same surgical team were reviewed. 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.

Preoperative ultrasonographic studies, CT scans, MRCPs, and intraoperative cholangiograms were carried out to delineate biliary system. Peri-operative liver function tests were evaluated.

✉ Long Li
lilong22@hotmail.com

✉ Wei Cheng
wei.cheng@monash.edu

[1] Department of Pediatric Surgery, Capital Institute of Pediatrics, Beijing 100020, People's Republic of China

[2] Department of Paediatrics and Department of Surgery, Southern Medical School, Faculty of Medicine, Nursing and Health Sciences, Monash University, Clayton, VIC 3168, Australia

Prior to 2012, conventional laparoscopic surgeries for CDC were carried out. The approach of conventional laparoscopic hepaticojejunostomy has been previously described [7]. After 2012, single-incision laparoscopic technique was adopted [8]. All instruments were placed through the umbilicus. Carbon dioxide pneumoperitoneum was established at a pressure of 10 mmHg for CDC children younger than 1 year of age, and 12 mmHg for those older than 1 year of age.

A trans-abdominal wall suture was placed through serosa of gallbladder fundus for cephalad liver retraction to expose the hepatic hilum. The second trans-abdominal wall retraction suture was placed through the proximal CHD to facilitate dissection and hepatico-jejunal anastomosis. In patient with AHD located in the middle portion of cyst duct, an additional retraction suture was placed through middle portion of gallbladder fossa to retract liver and facilitate anastomosis.

Patients were managed individually based on the subtypes:

Type 1: AHD locates close to the conjunction of cystic duct and CHD (Fig. 1A): The AHD and CHD were combined as one stoma or sutured along their lateral walls to form an anastomotic stoma (Fig. 2A);
Type 2: AHD locates in the mid-portion of cystic duct (Fig. 1B): Anastomosis of AHD and CHD to jejunum were carried out separately (Fig. 2B);
Type 3: Duplication of cystic duct (Fig. 1C): The duplicated cystic duct was ligated before being divided to prevent bile leak (Fig. 2C);
Type 4: Associated with aberrant right hepatic artery (RHA): In case of RHA anteriorly compressing CHD and AHD (Type 4a, Fig. 1D): Aberrant RHA was repositioned behind. The AHD and CHD were combined as one anastomotic stoma. In case of RHA anteriorly compressing CHD alone (Fig. 1E), the connection between AHD and CHD was transected (Type 4b, Fig. 2D). After repositioning the RHA behind CHD, the lateral walls of AHD and CHD were sutured to form an anastomotic stoma (Fig. 2E). A drainage tube was placed.

Patients were followed up in our clinic 1, 2, 3, 6 months postoperatively and every 6 months thereafter. The laboratory results and findings of ultrasonographic and upper gastrointestinal studies were assessed.

Statistic analysis

Data were analyzed with SPSS 13.0 package. Paired *t* tests were applied to compare peri-operative laboratory values. $p < 0.05$ was considered to be statistically significant.

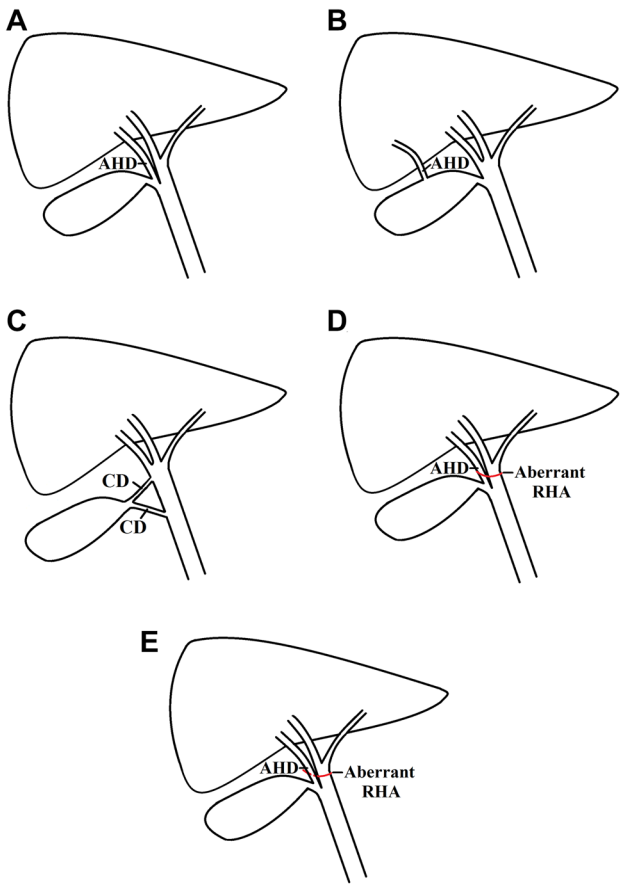

Fig. 1 The categories of aberrant hepatic ducts in our series. **A** Type 1: Aberrant hepatic duct (AHD) locates close to junction of cystic duct and common hepatic duct. **B** Type 2: Aberrant hepatic duct (AHD) locates in mid-portion of cystic duct. **C** Type 3: Duplication of cystic duct (CD). **D** Type 4a: Combined with aberrant right hepatic artery (RHA) anteriorly compressing both common hepatic duct (CHD) and aberrant hepatic duct (AHD). **E** Type 4b: Combined with aberrant right hepatic artery (RHA) anteriorly compressing common hepatic duct (CHD) alone

Results

Sixty CDC children with AHDs were successfully treated laparoscopically (F/M: 46/14, Type 1: $n = 47$, Type 2: $n = 6$, Type 3: $n = 3$, Type 4a: $n = 1$, Type 4b: $n = 3$). The mean age at surgery was 3.91 ± 3.00 years (25 days–15.52 years). Two (3.3%) patients from Type 2 subtype group developed bile leaks after primary surgeries because the dense adhesions near the perforated site. They underwent laparoscopic anastomosis of AHD to jejunum in the redo surgeries. The drain tubes were placed for 7 and 9 days respectively. The patients were discharged at day 11 after primary surgeries, respectively. In the remaining 58 patients, the average operative time was 3.75 ± 1.20 h (range 1.5–6 h). The mean postoperative hospital stay was

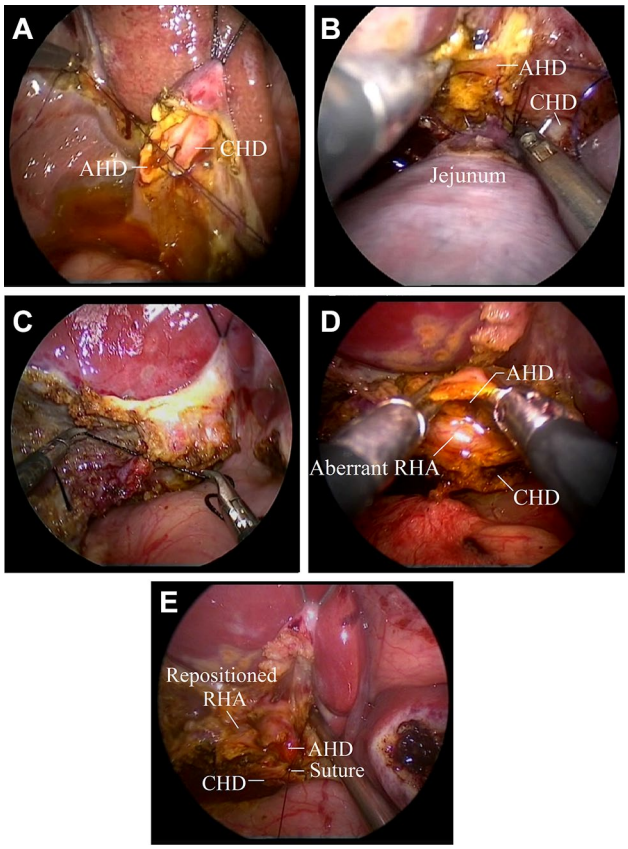

Fig. 2 Individualized laparoscopic ductoplasties and hepaticojejunostomies for management of aberrant hepatic ducts in children with choledochal cysts. **A** The lateral walls of aberrant hepatic duct (AHD) and common hepatic duct (CHD) were sutured to form an anastomotic stoma. **B** The anastomosis of aberrant hepatic duct (AHD) and common hepatic duct (CHD) to jejunum were carried out separately. **C** The duplicated cyst duct was ligated before division to prevent bile leak. **D**, **E** In case of aberrant right hepatic artery (RHA) compressing CHD alone, the connection between AHD and CHD was transected. After repositioning the RHA behind CHD, the lateral walls of AHD and CHD were sutured to form an anastomotic stoma for hepaticojejunostomy

6.02 ± 1.62 days. The average duration for full diet resumption was 2.25 ± 0.86 days. The mean duration of drainage was 4.05 ± 1.71 days. The median follow-up period was 30 months (12–216 months). Two (3.3%) patient with giant cysts had fluid collections because of extensive dissection of the intrapancreatic segment of cysts. They were successfully cured by 9 and 11 days drainage, respectively. One (1.6%) patient encountered duodenal injury because of perforation induced severe adhesions, and underwent laparoscopic repair. None of patients had anastomotic stenosis, bile leak, cholangitis, intrahepatic reflux, pancreatic leak, pancreatitis, Roux-loop obstruction, or adhesive intestinal obstruction. Postoperative liver function tests and serum amylase level returned to normal within 1 year (Table 1, $p < 0.001$).

Discussion

Previously published studies reported that 5–17% patients with bile duct injury had AHDs [9–12]. Severe inflammation or perforation, particularly at the hepatocystic junction increased the risk of AHD injury. Improper management of AHD frequently leads to bile leak or biloma, requiring redo surgeries.

In the era of open surgery, there were a few of case reports on AHDs in CDC children (1–2 patients in each case report respectively) [13–16]. In the era of laparoscopy, with the magnified view and easier inspection of intrahepatic duct and cystic duct provided by the telescope, more AHDs can be detected in the primary surgery. To date, there is a lack of large cohort on AHDs in CDCs, category and laparoscopic management strategy. Our study is the first large cohort on AHDs in CDC children, and provides individual laparoscopic management strategy.

Pre- and intraoperative investigations

In addition to understanding the morphology of the cyst, associated pancreatico-biliary malunion, and intestinal malformations, the ultrasonographic study and CT scans are helpful in establishing the relationship between the cyst and blood vessels, such as portal vein, hepatic artery, and their variations. It effectively prevents accidental injury of

Table 1 Pre- versus postoperative liver function parameters and serum amylase levels in choledochal cyst children with aberrant hepatic ducts undergoing laparoscopic ductoplasties and hepaticojejunostomies

	ALT (U/L) Ref: < 40	AST (U/L) Ref: < 40	ALP (U/L) Ref: < 400	GGT (U/L) Ref: 7–50	TBIL (μmol/L) Ref: 3.4–20	SAMY (U/L) Ref: 25–125
Pre-operation	145.59 ± 89.49	150.77 ± 93.10	442.58 ± 297.88	397.86 ± 300.96	84.62 ± 30.13	238.76 ± 200.25
Post-operation	20.35 ± 7.82	24.04 ± 7.48	156.57 ± 55.11	20.79 ± 10.81	9.61 ± 3.95	47.61 ± 28.09
p	< 0.001	< 0.001	< 0.001	< 0.001	< 0.001	< 0.001

NB: *ALT* alanine transaminase, *AST* aspartate aminotransferase, *ALP* alkaline phosphatase, *GGT* γ-glutamyl transpeptadase, *TBIL* total bilirubin, *SAMY* serum amylase

vessels. In case of aberrant right hepatic artery anteriorly compressing the proximal common hepatic duct, repositioning aberrant hepatic artery behind the proximal common hepatic duct is necessary to avoid postoperative biliary obstruction [17].

MRCP and intraoperative cholangiogram are more useful than ultrasonographic study and CT scan to assess the AHD. Conscious of overlapping of the gallbladder, cystic duct, and intrahepatic duct which may mask the AHDs, we routinely perform intraoperative laparoscopic assessment of possible AHD and artery.

Surgical strategy

Individualized management based on different AHD subtypes is advised to achieve the best outcome: (1) AHD opening near the CHD: combining AHD and CHD as one anastomosis; (2) AHD opening at middle cystic duct: separate AHD- and CHD-jejunostomy; (3) duplicated cystic duct: ligation before excision; (4) associated aberrant RHA: combining AHD and CHD as one anastomosis after replacing aberrant RHA behind.

Surgical techniques

Precaution of improper management of AHD

Improper management of AHD is preventable by recognition of anatomical variation and careful inspection in surgery. Based on our experiences, the following two conditions require surgeon's attention:

(1) Perforated CDC with dense adhesions: There is an extensive inflammatory adhesion between cystic duct and visceral surface of the liver when perforated site is located close to the conjunction of cystic duct, CHD, and CDC. In Type 2 patient, the connection between AHD and cystic duct is often mistaken for adhesions. Because the bile flow is intermittent, the AHD orifice is prone to be overlooked when adhesions covers it. Splitting the cyst duct from mid-portion to Calot's triangle before dissection is helpful to detect AHD orifice. In our series, two of Type 2 patients with perforated CDC developed bile leaks due to this oversight. We hence split the cystic duct to detect AHD orifice under direct vision. None of patients with AHDs encountered bile leak thereafter.
(2) Giant or large CDC associated gross dilatation of CHD: The border between CHD and CDC is blurred. The Type 1, i.e., the AHD locates close to conjunction of cystic duct and CHD, is prone to injury when severely dilated CHD is misjudged and dissected as upper portion of CDC. Transecting CDC at mid-portion is helpful to identify the border between CHD and CDC, and detect the AHD orifice indirect vision.

Precise dissection and anastomosis

Retraction sutures are placed at mid-portion of gallbladder fossa close to the AHD, and anterior wall of proximal CHD to obtain adequate surgical exposure, and facilitate dissection and anastomosis.

The diameter of AHD is usually small (2–3 mm). In patients with Type 2 AHD, the anastomosis between AHD and jejunum is technically demanding. To gain adequate working space, particularly for suture at 3 o'clock direction (near to the CHD), the AHD-jejunostomy is carried out prior to the CHD-jejunostomy, and starts from 3 o'clock position. The 6-0 PDS running suture is applied. This prevents anastomotic stricture or leakage.

Prevention of vascular injury

In patients with Type 4 AHD, the retraction suture is pulled upward to increase the space between RHA, CHD, and AHD during dissection, RHA repositioning, anastomosis between AHD and CHD for stoma formation, and anastomosis between newly established stoma and jejunum.

In conclusion, recognizing different subtypes of AHDs and treating them accordingly is effective to prevent complications caused by AHD injury. Individualized laparoscopic ductoplasty and hepaticojejunostomy provides an efficacious surgical option for CDC children with AHDs.

Compliance with ethical standards

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

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