RESEARCH



Single-incision laparoscopic hepaticojejunostomy with selective ductoplasty for type IV-A Choledochal cysts in children: a retrospective study



Wei Liu^{1,2}, Tong Yin^{1,3}, Xinyuan Chen^{1,2}, Mei Diao^{1,4*} and Long Li^{1,4,5*}

Abstract

Introduction Type IV-A choledochal cyst (CDC) has been considered to have a poor prognosis due to the high incidence of postoperative anastomotic strictures and intrahepatic stones. This study aimed to evaluate the surgical outcomes of children with type IV-A CDC and to provide insights for clinical diagnosis and treatment.

Methods The study retrospectively analyzed patients from June 2015 to December 2018 at our center, 76 children were diagnosed with type IV-A CDC. All patients underwent single-incision laparoscopic choledochal cyst excision and Roux-en-Y hepaticojejunostomy. The decision to perform ductoplasty was made by comprehensively considering the intrahepatic duct dilatation (IHDD) and stricture. All patients were followed up 1, 3, and 6 months postoperatively, and then every 1 year thereafter. Patients were categorized into two groups based on IHDD changes postoperatively: the long-term group (LTG), with IHDD persisting for over a year, and the short-term group (STG), where IHDD normalized within a year. Single/multiple factor logistic regression was used to analyse the factors influencing postoperative IHDD.

Results The median follow-up period was 80 months, with a range from 64 to 101 months. The decrease in postoperative liver function parameters, compared to preoperative levels, was statistically significant. Two patients (2.63%) developed bile leaks. One patient (1.32%) developed anastomotic stricture. All patients' IHDD returned to normal size. Fifty-six (73.7%) patients showed normalization of IHDD within one-year postoperatively. The median recovery time for IHDD in patients was 1.65 months, ranging from 3 days to 74 months postoperatively. There were significant differences in intrahepatic biliary sludge and stones and maximum diameter of IHDD between STG and LTG. Logistic regression was used to analyse the factors and found that intrahepatic biliary sludge and a wider maximum diameter of IHDD were risk factors for postoperative long-term IHDD in patients.

*Correspondence: Mei Diao psps3001@hotmail.com Long Li lilong23@126.com

Full list of author information is available at the end of the article



© The Author(s) 2024. **Open Access** This article is licensed under a Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License, which permits any non-commercial use, sharing, distribution and reproduction in any medium or format, as long as you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons licence, and indicate if you modified the licensed material. You do not have permission under this licence to share adapted material derived from this article are provide in the article's Creative Commons licence, unless indicated otherwise in a credit line to the material. If material is not included in the article's Creative Commons licence, unless indicated by statutory regulation or exceeds the permitted use, you will need to obtain permission directly from the copyright holder. To view a copy of this licence, visit http://creativecommons.org/licenses/by-nc-nd/4.0/.

Conclusion Single-incision laparoscopic hepaticojejunostomy with selective ductoplasty are safe and effective for children with Type IV-A CDC. Long-term follow-up is recommended for children presenting with intrahepatic biliary sludge and larger IHDD.

Keywords Choledochal cyst, Type IV-A, Intrahepatic duct dilatation, Single-incision ductoplasty

Introduction

Type IV-A choledochal cysts (CDC) are characterized by extrahepatic choledochal cysts and intrahepatic duct dilatation (IHDD) [1–4]. Traditionally, type IV-A CDC has been associated with a poor prognosis due to the high incidence of postoperative anastomotic strictures and intrahepatic stones [5, 6]. A study involving 14 patients with type IV-A CDC revealed that six patients experienced persistent dilatation postoperatively, and one developed multiple intrahepatic stones, eventually lead-ing to cholangiocarcinoma [7].

Single-incision laparoscopic surgery has been increasingly utilized in various pediatric surgical procedures because of its minimal invasiveness, favorable cosmetic results, and quicker postoperative recovery [8–10]. Selective ductoplasty can address strictures at the hepatic hilum and improve surgical outcomes [11].

The optimal surgical intervention for type IV-A CDC remains controversial, and there is a lack of related research focusing on children with type IV-A CDC. This study evaluates the surgical outcomes of children with type IV-A CDC at our center and aims to provide insights for clinical diagnosis and treatment.

Methods

Study population

From June 2015 to December 2018, a total of 717 children were diagnosed with CDC at our center, of which 78 were classified as type IV-A according to the Todani classification [Figure 1] [4, 12]. Demographics, preoperative ultrasonography, computed tomography (CT), perioperative laboratory results (within one week before and after surgery) and intraoperative cholangiography data were collected. Preoperative and intraoperative imaging examination revealed the shape of cysts, the diameter of IHDD, and existence of intrahepatic bile sludge or stones. Diagnosis was based on preoperative CT, abdominal ultrasonography, and intraoperative cholangiography. Two patients were excluded due to loss to follow-up, resulting in a final cohort of 76 patients.

Surgical methods

All patients underwent a single-incision laparoscopic choledochal cyst excision and Roux-en-Y hepaticojejunostomy [8, 13]. The patient was positioned supine with the head elevated and feet lowered. The surgeon stood between the patient's legs, and the laparoscopic assistant was positioned on the patient's left side. A single incision was made at the umbilicus for the insertion of the camera and operational trocars [Figure 2]. The gallbladder was suspended by piercing the serosal layer at the base with a silk suture. Intraoperative cholangiography was then performed. A second suspension suture was placed through the proximal common hepatic duct, and a third suture was used to suspend the middle of the cyst for easier dissection and removal. The distal end of the cyst was severed proximally to the pancreaticobiliary junction. If there was no stricture at the distal end, it was ligated using a Hem-o-Lok clip; otherwise, no ligature was applied. The proximal end of the cyst was transected along its upper edge. Intrahepatic bile ducts were inspected with a laparoscopic camera. The decision to perform ductoplasty was made based on a comprehensive assessment of IHDD and the strictures, guided by intraoperative findings and cholangiographic evaluation. Ductoplasty was performed as previously reported [11], with splitting the anterior wall of stricture vertically to the place of dilatation and suturing transversely [Figure 3. When stenosis involved the exit of the left or right hepatic ducts, the lateral wall of the stenotic segment is split longitudinally into the dilated portion. The incision is then spread open transversely and anastomosed with the jejunum [Figure 4]. For strictures at the exits of the left and right hepatic ducts, the stenotic segment is carefully split along the lateral wall and extended into the dilated portion of the duct, followed by a bi-ductal hepaticojejunostomy. Bile sludge and stones were evacuated by irrigation.

Follow-up and grouping

Postoperative follow-ups were conducted at 1, 3, and 6 months, and annually thereafter. These evaluations included laboratory tests and abdominal ultrasonography. Based on the changes of postoperative IHDD, the patients were divided into 2 groups: (1) long-term group (LTG): patients whose IHDD remained dilated for more than one year postoperatively; and (2) short-term group (STG): patients whose IHDD returned to normal size within one year postoperatively. Normal size was defined as the diameter of the intrahepatic duct corresponding to that of normal children of the same age, or the absence of noticeable intrahepatic duct dilatation, as determined by ultrasonography [14–16].





Fig. 1 Intraoperative cholangiography showing the IHDD and stricture at the exits of the left and right hepatic ducts

Ethics approval

This study is a retrospective study, which adhered to the ethical standards set by the World Medical Association's Declaration of Helsinki and was approved by the Ethical Committee of the Capital Institute of Pediatrics (SHERLL2022047), and written informed consent was received from the legal guardians of all participants.

Statistical analysis

Statistical analysis was performed using SPSS 27.0. Normally distributed data were presented in the form of Mean±SD, while non-normally distributed data were represented using Median (P25, P75). Categorical variables were presented as numbers and percentages. Data comparisons between the two groups was performed using t-test, rank sum test, chi-square test, and Fisher's exact test: Student's t-test was used for normal distribution data, Mann-Whitney U test was used for non-normally distribution data, chi-square test and Fisher's exact test were used for categorical variables data. Univariate and multivariate logistic regression analyses were used to determine the factors influencing the time of postoperative IHDD. A *p*-value <0.05 was considered statistically significant.

Results

Demographics and characteristics

All 76 children included in this study were diagnosed with type IV-A CDC, with a male to female ratio of 1:4.43 【Table 1】. The median age at surgery was 20.25 (8.47, 43.93) months, ranging from 3 days to 177 months. Twenty-five patients (32.9%) were diagnosed prenatally. Sixty-four patients (84.2%) presented with clinical symptoms, The primary symptoms included abdominal pain (63.2%), vomiting (55.3%), fever (27.6%), and jaundice (59.2%). Six patients (7.9%) presented with triad of



Fig. 2 Instrument arrangement for single-incision laparoscopic hepaticojejunostomy and placement of suspension sutures (arrow)

jaundice, pain and abdominal mass. Abnormal liver function was observed in 64 patients. Abnormal liver function was defined as elevated serum transaminase levels or elevated bilirubin levels. Bile sludge or stones were confirmed in 15 patients (19.7%) in the intrahepatic ducts and in 56 patients (73.7%) in the extrahepatic cysts. Perforation of extrahepatic cyst wall occurred in 18 patients (23.7%). Patients in STG and LTG showed no statistical differences in age, gender, prenatal diagnosis, preoperative symptoms, extrahepatic bile sludge or stones, and perforations (p > 0.05), and there were significant differences in intrahepatic biliary sludge and stones (p = 0.02).

Imaging examinations

A total of 69 patients (90.8%) had cystic choledochal cysts, while 7 (9.2%) exhibited fusiform cysts [Table 2].



Fig. 3 The surgical method for stricture at the common hepatic duct. a: The common hepatic duct (*black arrow*). b: Splitting the anterior wall of the common hepatic duct (*black arrow*). c: The split common hepatic duct, showing the exits of the left and right hepatic ducts. the split anterior walls of the hepatic duct (*black arrows*). d: A wide anastomosis

The mean diameter (the horizontal extent) of the CDC was 47.76 ± 23.39 mm, with an average length (the vertical extent) of 72.14 ± 31.83 mm. IHDD involved a single lobe in 32 patients (42.1%)—with the left lobe in 26 patients (34.2%) and the right lobe in 6 patients (7.9%)— and spanned both lobes in 44 patients (57.9%). The median maximum diameter of IHDD was 20.00 (15.00, 30.00) mm. There were no statistically significant differences between the short-term group (STG) and long-term group (LTG) in terms of the shape, diameter, length

of the CDC, and the location of IHDD (p>0.05). However, significant differences were noted in the maximum diameter of IHDD (p=0.003).

Operative information

The mean surgical duration was 225.95 ± 78.87 min. All patients underwent single-incision laparoscopic hepaticojejunostomy, with no patients undergoing robotic surgery. However, three patients (3.9%) were converted to open surgery intraoperatively due to pre-existing



Fig. 4 The surgical method for stricture at the exit of the left hepatic duct. a: Stricture at the exit of the left hepatic duct (*white arrow*). b: Splitting the anterior wall of the left hepatic duct. c: The split left hepatic duct, with the split lateral walls (*black arrows*). d: Schematic diagram of the surgical method, showing the stricture at the exit of the left hepatic duct (*white arrow*) and the split lateral walls of the left hepatic duct (*black arrows*).

complicated conditions: one had severe adhesions and significant bleeding, another presented a heavily adhered cyst wall to the duodenum with unclear boundaries and a 0.2 cm intestinal wall perforation, and a third was due to the large size and extensive perforation of the cyst, with unclear adhesions to the duodenum and pancreas. Additionally, 19 patients (25.0%) required ductoplasties due to excessive strictures at the hepatic hilum: 6 cases in the LTG and 13 cases in the STG (p > 0.05).

Perioperative laboratory results

Laboratory results were collected within one week preand post-surgery. Postoperative liver function parameters, jaundice and serum amylase levels, all decreased and compared to preoperative levels were statistically significant. No significant statistical differences were observed between STG and LTG [Table 3].

 Table 1
 Demographics and characteristics of patients

Characteristics	STG	LTG (n = 20)	р	
	(<i>n</i> = 56)			
Age (month), M(IQR)	20.45 (4.7,	18.28 (10.83,	0.591	
	41.20)	56.50)		
Gender			0.058	
Male	7 (12.5%)	7 (35.0%)		
Female	49 (87.5%)	13(65.0%)		
Prenatal diagnosis	20 (35.7%)	5 (25.0%)	0.381	
Preoperative symptoms (n)				
Abdominal pain	32 (57.1%)	16 (33.3%)	0.069	
Vomiting	29 (51.8%)	13 (65.0%)	0.308	
Fever	16 (28.6%)	5 (25.0%)	0.759	
Jaundice	34 (60.7%)	11 (55.0%)	0.655	
Preoperative liver dysfunction (n)	48 (85.7%)	16 (80.0%)	0.807	
Intrahepatic bile sludge or stones	7 (12.5%)	8 (40.0%)	0.020	
(n)				
Extrahepatic bile sludge or stones	42 (75.0%)	14 (70.0%)	0.663	
Perforation	11 (19.6%)	8 (40.0%)	0.071	
STG short-term group /TG long-term g	roup			

STG short-term group, LTG long-term group

Table 2 Shape of Type-IV-A CDC

Characteristics	STG (n = 56)	LTG (n = 20)	р	
Shape of CDC			0.553	
Cystic	52 (92.9%)	17 (85.0%)		
Fusiform	4 (7.1%)	3 (15.0%)		
Diameter of CDC	50.50 ± 23.67	40.10±21.30	0.088	
Length of CDC	75.10 ± 32.00	63.85 ± 30.60	0.176	
Location of IHDD			0.147	
Single lobe	24 (42.9%)	5 (25.0%)		
Left lobe	19 (33.9%)	5 (25.0%)		
Right lobe	5 (8.9%)	0 (0.0%)		
Both lobes	23 (41.1%)	14 (70.0%)		
Maximum diameter of	15.00 (13.85,	22.50 (20.00,	0.003	
IHDD (mm)	25.00)	33.75)		

Postoperative complications

Following surgery, patients resumed a full diet after an average of 3.43±1.16 days, had drainage for an average of 4.08 ± 1.94 days, and the mean postoperative hospital stay was 6.38±1.90 days. Two patients (2.63%) experienced bile leaks, and neither had perforations. One patient did not undergo ductoplasty at the primary surgery and developed a postoperative bile leak, requiring a repair surgery on the fifth day after surgery. The other patient underwent ductoplasty during the primary surgery due to a stricture at the hepatic hilum. Postoperative bile leakage occurred, and a repeated hepaticojejunostomy was performed on the fifth day. One child (1.32%) developed anastomotic stricture, underwent necessitating another hepaticojejunostomy five months postoperatively. There were no reports of cholangitis, intrahepatic duct stones, pancreatitis, or other long-term complications.

Postoperative IHDD and factors influencing the time of postoperative IHDD

The median follow-up period was 80 months operatively, ranging from 64 to 101 months. All patients' IHDD returned to normal size **(**Figure 5**)**. Within one month postoperatively, IHDD had returned to normal size in 26 patients (34.2%). By three months, 45 patients (59.2%) had normalized IHDD. At six months postoperatively, 46 patients (60.5%) had returned to normal IHDD. Within one year, 56 patients (73.7%) exhibited normalized IHDD. The median recovery time for IHDD in patients was 1.65 months, ranging from 3 days to 74 months.

Univariate logistic regression analysis revealed that preoperative general information and imaging differences between the STG and LTG were significant **[** Table 4 **]**. It was determined that intrahepatic biliary sludge and the maximum diameter of IHDD were correlated with prolonged postoperative IHDD. Further multivariate logistic regression analysis identified these two factors as significant risk factors for long-term IHDD.

Discussion

Single-incision laparoscopic surgery has been widely adopted across various pediatric surgical procedures due to its minimal invasiveness and excellent outcomes [8, 9, 13, 17]. Our findings indicated it a viable option for the surgical treatment of type IV-A CDC, providing patients with significant recovery advantages while reducing surgical trauma.

In 2012, single-incision laparoscopic surgery was first reported in the treatment of children with CDCs [8]. Subsequently, studies had reported the use of singleincision laparoscopy for giant CDCs [18] and perforated CDCs [19]. Compared to traditional laparoscopic surgery, single-incision laparoscopy has shown comparable short-term and mid-term outcomes [13, 20, 21]. Moreover, it achieves a virtually scarless cosmetic outcome, meeting patient demands while advancing the pursuit of minimally invasive goals in laparoscopic technology. The management of primary strictures at the hepatic hilum is controversial. In 1997, Ando reported on stricture resection surgery [22], and in 1998, Todani described hepatic hilar anastomosis [23]. Experiences with ductoplasty have also been reported [11, 24], demonstrating that it is a safe and effective technique for correcting strictures at the hepatic hilum.

Single-incision laparoscopic hepaticojejunostomy presents unique challenges when applied to CDC in children. First, placement of the telescope and instruments through a single umbilical port often results in "instrument crowding." To minimize clashing, we used a longer laparoscope and shorter instruments to create a steeper triangulation angle, while the multi-channel single-incision approach further enhanced instrument

 Table 3
 Perioperative Liver function, jaundice and serum amylase levels in patients

Characteristics	Total	p (pre-operation vs. post-operation)	STG (n = 56)	LTG (n = 20)	р
ALT					
Pre-operation	24.50(15.25, 110.70)	0.004	26.45 (15.85, 110.70)	21.00 (12.60, 106.625)	0.483
Post-operation	26.45(15.55, 40.325)		27.30 (14.95, 42.40)	24.85 (15.90, 39.40)	0.878
AST					
Pre-operation	44.95(32.75, 73.975)	<0.001	45.45 (35.475, 79.40)	37.50 (30.15, 59.575)	0.313
Post-operation	30.95 (25.15, 39.50)		30.60 (24.70, 37.65)	32.25 (27.175, 46.00)	0.308
ALP					
Pre-operation	231.50(174.250, 306.75)	<0.001	231.00 (182.50, 351.25)	232.00 (163.50, 292.50)	0.633
Post-operation	129.50(106.75, 176.75)		135.00 (104.50, 193.50)	127.50 (113.00, 170.50)	0.719
GGT					
Pre-operation	234.50(30.45, 448.75)	<0.001	234.50 (30.45, 473.50)	254.45 (26.675, 437.75)	0.962
Post-operation	90.35(33.00, 177.975)		83.70 (33.50, 186.25)	112.00 (25.725, 167.025)	0.750
TBIL					
Pre-operation	14.35(7.85, 34.35)	<0.001	13.25 (7.85, 38.45)	15.45 (6.725, 25.275)	0.684
Post-operation	9.35(7.40, 15.20)		9.75 (7.40, 19.475)	8.70 (6.025, 11.60)	0.291
DBIL					
Pre-operation	4.90(1.80, 14.325)	0.037	3.35 (1.725, 14.70)	5.70 (1.95, 10.675)	0.850
Post-operation	2.90(2.00, 7.30)		2.90 (2.00, 7.55)	2.75 (1.95, 3.55)	0.532
AMY					
Pre-operation	48.00(16.25, 73.00)	0.002	51.50 (18.00, 76.00)	31.50 (15.25, 65.75)	0.450
Post-operation	27.50(13.00, 68.75)		27.00 (14.25, 74.50)	34.50 (11.00, 56.75)	0.645

ALT Alanine Aminotransferase, AST Aspartate Aminotransferase, ALP Alkaline Phosphatase, GGT Gamma-Glutamyl Transferase, TBIL Total Bilirubin, DBIL Direct Bilirubin, AMY Amylase





Fig. 5 Ultrasound images from the same patient. a: day 3 after surgery. b: month 14 after surgery

Table 4 Univariate an	d multivariate logistic	regression analyse	es of factors influencing the time	of postoperative IHDD
-----------------------	-------------------------	--------------------	------------------------------------	-----------------------

Variables	Univariate logistic regression				Multivariate logistic regression					
	β	S.E.	Wald	<i>p</i> value	95%CI	β	S.E.	Wald	P value	95%CI
Maximum diameter of IHDD	-0.73	0.28	6.846	0.009	0.929 (0.880–0.982)	-0.071	0.030	5.589	0.018	0.931 (0.878–0.988)
Intrahepatic bile sludge or stones	-1.54	0.610	6.386	0.012	0.214 (0.065-0.708)	-1.485	0.656	5.127	0.024	0.226 (0.063-0.819)

maneuverability and flexibility. Furthermore, suspension sutures were applied to expose deep anatomical structures. A series of suspension sutures were placed relay-style through the abdominal wall, with the assistant adjusting the tension and direction of the sutures. This technique aids in identifying the cyst boundaries, thereby reducing the risk of iatrogenic injury. Single-incision laparoscopic hepaticojejunostomy not only allows for effective visualization and manipulation of biliary structures but also minimizes scarring, which is particularly beneficial given the thinner abdominal walls of children that enhance instrument maneuverability. During ductoplasty, we believe it is safer to split the anterior stenotic wall to widen the hepatic duct because the hepatic artery and portal vein are located closely near the posterior wall of the hepatic duct. The anterior wall is relatively free from these critical vascular structures, reducing the risk of vascular injury. Oozing from the ductal wall during the procedure can be well controlled using cautery.

Type IV-A CDC involves extrahepatic cysts combined with IHDD, complicating differentiation from secondary IHDD caused by type I-C. Todani suggested that type I CDC presented as fusiform dilatation extending into the intrahepatic duct, while type IV-A CDC often involves primary strictures [4]. This distinction is critical as it influences surgical methods and postoperative management. Previous research had demonstrated that strictures commonly occur at the exits of the left and right hepatic ducts within the intrahepatic bile ducts [25]. In our study of 19 patients who underwent ductoplasties, 12 had stricture at the common hepatic duct, and 7 involved the exits of the left and/or right hepatic ducts. When addressing strictures at the common hepatic duct, we performed a vertical incision on the anterior wall of the stricture, followed by transversely suturing. This technique ensures that the hepaticojejunostomy anastomosis spans at least 0.5 cm (neonates \geq 0.5 cm, older children \geq 1 cm) to prevent postoperative anastomotic strictures [26]. Similarly, strictures at the exits of the left and right hepatic ducts require vertical incisions, and subsequent reshaping of the hepatic ducts to prevent cholestasis.

Type IV-A CDC was considered to have a higher probability of postoperative complications such as anastomotic stricture and cholangiocarcinoma [27–29]. Anand reported 45 Type IV-A CDC patients aged 15 to 40 years who underwent only hepaticojejunostomy, 20% developed cholangitis and/or intrahepatic stones, and 13.33% developed anastomotic stricture postoperatively [30]. In Xia's study, 59 adult patients with type IV-A CDC who underwent hepaticojejunostomy and partial hepatectomy, 8.47% experienced bile leakage, 11.86% had delayed wound healing, 5.08% suffered from abdominal infections, and 12.2% faced recurrent cholangitis postoperatively [31]. Our findings indicate that Type IV-A CDC children have a favorable prognosis following singleincision laparoscopic hepaticojejunostomy with selective ductoplasty. This improved outcome may be attributed to ductoplasty reduced the possibility of postoperative cholestasis caused by strictures at the hepatic hilum. For children with type IV-A CDC undergoing initial surgery, single-incision laparoscopic hepaticojejunostomy with selective ductoplasty is feasible. However, for patients suffering from recurrent cholangitis or liver infections after the primary surgery, and with persistent IHDD involving second-order or higher bile ducts that cannot be addressed at the hepatic hilum, partial hepatectomy may be considered.

Persistent postoperative IHDD may obstructed bile flow which might cause biliary infections, leading to the formation of bilirubin calcium stones, cholangitis, intrahepatic duct stones, and even cholangiocarcinoma [7, 32, 33]. However, there is no established consensus regarding the postoperative recovery from IHDD [6, 25, 34]. In our study, all children's postoperative IHDD recovered to normal size, with 73.7% of patients achieving recovery within one year. Studies have shown that bile duct dilatation is related to the pressure [6, 35], which increases after biliary obstruction. The rapid resolution of IHDD may be associated with relief of obstructions at the hepatic hilum and/or distal common bile duct. As mentioned in Hill's study [6], effective surgical intervention and subsequent pressure reduction resulted in a dramatic decrease in intrahepatic duct diameter, suggesting that duct dilation in children can be altered.

Analysis of postoperative risk factors identified intrahepatic biliary sludge and a larger IHDD diameter as predictors of persistent IHDD. The presence of intrahepatic biliary sludge and stones are associated with cholestasis [36]. During surgery, laparoscopic techniques were employed to carefully examine the intrahepatic ducts and remove any sludge and stones thoroughly. However, the presence of undetectable micro-stones deep within the ducts remains uncertain, potentially delay the recovery of IHDD. In addition, severe IHDD requires a longer recovery period. Although long-term IHDD might be asymptomatic, it could also lead to recurrent cholangitis. Consequently, patients had intrahepatic biliary sludge and a wider IHDD diameter require extended postoperative monitoring for cholangitis and intrahepatic stones.

However, our center is a pediatric treatment center and there are age limitations on our patients, further research is needed to explore the relationship between patient age and the recovery of IHDD. Besides, the single-center nature of this study has the limitation of small sample size, additional multi-center studies are essential to validate our findings. Although our study suggests that the bile ducts can return to normal size, this result warrants more comprehensive, long-term follow-up to accurately monitor bile duct recovery and evaluate the potential for recurrent dilation or other complications.

Conclusion

In conclusion, our findings support the safety and effectiveness of single-incision laparoscopic hepaticojejunostomy with selective ductoplasty. Additionally, we suggest long-term follow-up for children with intrahepatic biliary sludge and an increased IHDD diameter to effectively monitor for potential complications.

Author contributions

WL and TY: data acquisition; WL: drafting of manuscript; XYC: analysis and data interpretation; MD and LL: critical revision of manuscript.

Funding

The project is supported by the Research Unit of Minimally Invasive Pediatric Surgery on Diagnosis and Treatment, Chinese Academy of Medical Sciences 2021RU015, Beijing Hospital Authority's Ascent Plan, Code: DFL20221101, and Capital's Funds for Health Improvement and Research 2020-2-2103.

Data availability

The data generated and/or analyzed in this study are not open to the public to safeguard individual privacy but are available from the corresponding author on reasonable request.

Declarations

Competing interests

The authors declare no competing interests.

Ethics approval

This study is a retrospective study, which adhered to the ethical standards set by the World Medical Association's Declaration of Helsinki and was approved by the Ethical Committee of the Capital Institute of Pediatrics (SHERLL2022047), and written informed consent was received from the legal guardians of all participants.

Data accessibility

The data generated and/or analyzed in this study are not open to the public to safeguard individual privacy but are available from the corresponding author on reasonable request.

Declaration of competing interest

No authors report any conflict of interest.

Author details

¹Department of General Surgery, Capital Institute of Pediatrics, Beijing, China

²Graduate School of Peking Union Medical College, Chinese Academy of Medical Sciences, Beijing, China

³Capital Institute of Pediatrics-Peking University Teaching Hospital, Beijing, China

⁴Research Unit of Minimally Invasive Pediatric Surgery on Diagnosis and Treatment, Chinese Academy of Medical Sciences 2021RU015, Beijing, China

⁵Department of Pediatric Surgery, Tsinghua University Affiliated Beijing Tsinghua Changgung Hospital, Beijing, China

Received: 16 July 2024 / Accepted: 28 October 2024 Published online: 15 November 2024

References

- Singham J, Schaeffer D, Yoshida E, Scudamore C. Choledochal cysts: analysis of disease pattern and optimal treatment in adult and paediatric patients. HPB (Oxford). 2007;9(5):383–7.
- Edil BH, Cameron JL, Reddy S, Lum Y, Lipsett PA, Nathan H, Pawlik TM, Choti MA, Wolfgang CL, Schulick RD. Choledochal cyst disease in children and adults: a 30-year single-institution experience. J Am Coll Surg. 2008;206(5):1000–5. discussion 1005–1008.
- Miyano T, Yamataka A, Kato Y, Segawa O, Lane G, Takamizawa S, Kohno S, Fujiwara T. Hepaticoenterostomy after excision of choledochal cyst in children: a 30-year experience with 180 cases. J Pediatr Surg. 1996;31(10):1417–21.
- Todani T, Watanabe Y, Toki A, Morotomi Y. Classification of congenital biliary cystic disease: special reference to type ic and IVA cysts with primary ductal stricture. J Hepatobiliary Pancreat Surg. 2003;10(5):340–4.
- Kim JH, Choi TY, Han JH, Yoo BM, Kim JH, Hong J, Kim MW, Kim WH. Risk factors of postoperative anastomotic stricture after excision of choledochal cysts with hepaticojejunostomy. J Gastrointest Surg. 2008;12(5):822–8.

- Hill R, Parsons C, Farrant P, Sellars M, Davenport M. Intrahepatic duct dilatation in type 4 choledochal malformation: pressure-related, postoperative resolution. J Pediatr Surg. 2011;46(2):299–303.
- Ono S, Fumino S, Shimadera S, Iwai N. Long-term outcomes after hepaticojejunostomy for choledochal cyst: a 10- to 27-year follow-up. J Pediatr Surg. 2010;45(2):376–8.
- Diao M, Li L, Dong N, Li Q, Cheng W. Single-incision laparoscopic roux-en-Y hepaticojejunostomy using conventional instruments for children with choledochal cysts. Surg Endosc. 2012;26(6):1784–90.
- Kobayashi M, Mizuno M, Sasaki A, Arisue A, Akiyama S, Wakabayashi G. Singleport laparoscopic Heller myotomy and Dor fundoplication: initial experience with a new approach for the treatment of pediatric achalasia. J Pediatr Surg. 2011;46(11):2200–3.
- Jones RE, Zagory JA, Clark RA, Pandya SR. A narrative review of the modern surgical management of pediatric choledochal cysts. Transl Gastroenterol Hepatol. 2021;6:37.
- Li L, Liu SL, Hou WY, Cui L, Liu XL, Jun Z, Liu-Ming H, Gang L, Kamal NA. Laparoscopic correction of biliary duct stenosis in choledochal cyst. J Pediatr Surg. 2008;43(4):644–6.
- Todani T, Watanabe Y, Narusue M, Tabuchi K, Okajima K. Congenital bile duct cysts: classification, operative procedures, and review of thirty-seven cases including cancer arising from choledochal cyst. Am J Surg. 1977;134(2):263–9.
- Diao M, Li L, Li Q, Ye M, Cheng W. Single-incision versus conventional laparoscopic cyst excision and Roux-Y hepaticojejunostomy for children with choledochal cysts: a case-control study. World J Surg. 2013;37(7):1707–13.
- 14. Matcuk GR Jr., Grant EG, Ralls PW. Ultrasound measurements of the bile ducts and gallbladder: normal ranges and effects of age, sex, cholecystectomy, and pathologic states. Ultrasound Q. 2014;30(1):41–8.
- McGahan JP, Phillips HE, Cox KL. Sonography of the normal pediatric gallbladder and biliary tract. Radiology. 1982;144(4):873–5.
- Hamada Y, Ando H, Kamisawa T, Itoi T, Urushihara N, Koshinaga T, Saito T, Fujii H, Morotomi Y. Diagnostic criteria for congenital biliary dilatation 2015. J Hepatobiliary Pancreat Sci. 2016;23(6):342–6.
- Muensterer OJ, Adibe OO, Harmon CM, Chong A, Hansen EN, Bartle D, Georgeson KE. Single-incision laparoscopic pyloromyotomy: initial experience. Surg Endosc. 2010;24(7):1589–93.
- Diao M, Li L, Li Q, Ye M, Cheng W. Challenges and strategies for single-incision laparoscopic roux-en-Y hepaticojejunostomy in managing giant choledochal cysts. Int J Surg. 2014;12(5):412–7.
- 19. Diao M, Li L, Cheng W. Single-incision laparoscopic hepaticojejunostomy for children with perforated choledochal cysts. Surg Endosc. 2018;32(7):3402–9.
- 20. Tang Y, Li F, He G. Comparison of single-incision and conventional laparoscopic cyst excision and Roux-en-Y hepaticojejunostomy for children with Choledochal cysts. Indian J Surg. 2016;78(4):259–64.
- Xu D, Tang K, He S. A modified technique of single-incision laparoscopic hepaticojejunostomy for children with choledochal cysts. BMC Surg. 2019;19(1):36.
- Ando H, Kaneko K, Ito F, Seo T, Ito T. Operative treatment of congenital stenoses of the intrahepatic bile ducts in patients with choledochal cysts. Am J Surg. 1997;173(6):491–4.
- 23. Todani T, Watanabe Y, Toki A, Ogura K, Wang ZQ. Co-existing biliary anomalies and anatomical variants in choledochal cyst. Br J Surg. 1998;85(6):760–3.
- Li S, Wang W, Yu Z, Xu W. Laparoscopically assisted extrahepatic bile duct excision with ductoplasty and a widened hepaticojejunostomy for complicated hepatobiliary dilatation. Pediatr Surg Int. 2014;30(6):593–8.
- Koshinaga T, Inoue M, Ohashi K, Sugito K, Ikeda T, Hagiwara N, Tomita R. Persistent biliary dilatation and stenosis in postoperative congenital choledochal cyst. J Hepatobiliary Pancreat Sci. 2011;18(1):47–52.
- 26. Diao M, Li L, Cheng W. Recurrence of biliary tract obstructions after primary laparoscopic hepaticojejunostomy in children with choledochal cysts. Surg Endosc. 2016;30(9):3910–5.
- Takimoto A, Fumino S, Iguchi M, Takemoto M, Takayama S, Kim K, Higashi M, Aoi S. Current treatment strategies for postoperative intrahepatic bile duct stones in congenital biliary dilatation: a single center retrospective study. BMC Pediatr. 2022;22(1):695.
- de Kleine RH, Schreuder AM, Ten Hove A, Hulscher JBF, Borel Rinkes IHM, Dejong CHC, de Jonge J, de Reuver P, Erdmann J, Kazemier G, et al. Choledochal malformations in adults in the Netherlands: results from a nationwide retrospective cohort study. Liver Int. 2020;40(10):2469–75.
- Ray S, Dhali A, Khamrui S, Mandal TS, Das S, Dhali GK. Surgical outcomes after re-operation for excision of choledochal cyst with delayed biliary complications: a retrospective study on 40 patients. Am J Surg. 2023;226(1):93–8.

- Anand U, John AG, Priyadarshi RN, Kumar R, Singh BN, Parasar K, Kumar B. Long-term complications after extrahepatic cyst excision for type IV-A choledochal cysts. Ann Hepatobiliary Pancreat Surg. 2023;27(4):433–6.
- Xia HT, Dong JH, Yang T, Zeng JP, Liang B. Extrahepatic cyst excision and partial hepatectomy for Todani type IV-A cysts. Dig Liver Dis. 2014;46(11):1025–30.
- Maki T. Pathogenesis of calcium bilirubinate gallstone: role of E. Coli, betaglucuronidase and coagulation by inorganic ions, polyelectrolytes and agitation. Ann Surg. 1966;164(1):90–100.
- Li FY, Cheng NS, Mao H, Jiang LS, Cheng JQ, Li QS, Munireddy S. Significance of controlling chronic proliferative cholangitis in the treatment of hepatolithiasis. World J Surg. 2009;33(10):2155–60.
- Congo K, Lopes MF, Oliveira PH, Matos H, Basso S, Reis A. Outcomes of choledochal cysts with or without intrahepatic involvement in children after

extrahepatic cyst excision and Roux-en-Y hepaticojejunostomy. Ann Hepatol. 2012;11(4):536–43.

- Lv Y, Liu N, Wu H, Li Z. Etiological classification and treatment strategies for secondary bile duct dilatation. Exp Biol Med (Maywood). 2021;246(3):281–5.
- 36. Ran X, Yin B, Ma B. Four major factors contributing to Intrahepatic stones. Gastroenterol Res Pract. 2017;2017:7213043.

Publisher's note

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.