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DYNAMIC MANUSCRIPT

Changes in outcomes and operative trends with pediatric robot-assisted resection of choledochal cyst

Kyong Ihn[1] · In Geol Ho[1] · Young Ju Hong[2] · Ho Jong Jeon[1] · Dongeun Lee[1] · Seok Joo Han[1]

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Abstract

Background This study aimed to report our experience with a robot-assisted resection of choledochal cysts (CCs) in pediatric patients, especially focusing on changes in outcomes and operative trends.

Methods We retrospectively reviewed medical records of all 158 patients under 18 years of age who underwent robot-assisted resection of CC in a single tertiary center between July 2008 and January 2021. Patients were divided into the first period (P1, July 2008–March 2016; $N=79$) and second period (P2, April 2016–January 2021; $N=79$) with equal number of participants. The patients of P2 were compared with those of P1 to assess clinical outcomes with operative details. Operative characteristics and postoperative prognosis were compared for each group.

Results The mean operative time was 383.6 min for the P2 group and 462.6 min for the P1 group ($p<0.001$). The mean estimated blood loss was 28 mL in the P2 group and 63 mL in the P1 group ($p=0.025$). The rate of emergency department visit after the operation was lower in the P2 group (3.8% vs. 13.9%, respectively, $p=0.047$). The two groups showed no significant differences in the rate of late postoperative complications and reoperations.

Conclusion With the increase in the center's experience, robot-assisted resection of CC can be safely adopted and feasible, especially for pediatric patients.

Levels of evidence Treatment Study, Level III.

Keywords Choledochal cyst · Robot-assisted surgery · Pediatric · Minimally invasive surgical procedures · Pancreaticobiliary maljunction

A choledochal cyst (CC) is a pathological condition characterized by various degrees of congenital dilatation of the extrahepatic and/or intrahepatic biliary tract. CC is a rare disease mainly affecting Asian populations up to 1 in 1000 people in Japan [1, 2]. Although CCs are recognized as benign and correctable congenital anomalies, cholangitis, pancreatitis, and malignant transformation may be anticipated if not properly treated. The standard treatment is complete resection of the cyst with Roux-en-Y hepaticojejunostomy.

With the advent of pediatric laparoscopic surgical techniques, several authors have reported the feasibility and safety of laparoscopic resection of CC [3, 4]. However, the laparoscopic approach is limited by the inflexible laparoscopic instruments and technically demanding procedures [5]. Meanwhile, the robotic system offers potential solutions through its more sophisticated features. It provides articulating instruments and a magnified, three-dimensional view of the operative fields, motion scaling and tremor filtering, thereby increasing surgical dexterity. The role of robotic surgery has been limited especially in the pediatric surgical field, because of the complete lack of haptic feedback and relatively high cost compared with open or laparoscopic surgery. However, many authors believe that robots are particularly well-suited for resection of CC because of the increased dexterity and degrees of freedom over rigid laparoscopic instruments in dissecting the intra-pancreatic CC and fine

✉ Seok Joo Han
sjhan@yuhs.ac

[1] Department of Pediatric Surgery, Department of Surgery, Severance Children's Hospital, Yonsei University College of Medicine, 50 Yonsei-ro, Seodaemun-gu, Seoul 03722, Republic of Korea

[2] Department of Surgery, Yong-in Severance Hospital, Yonsei University College of Medicine, 363 Dongbaekjukjeon-daero, Giheung-gu, Yongin-si, Gyeonggi-do, 16995, Republic of Korea

anastomosis of hepaticojejunostomy [6–10]. We have also previously reported our successful surgical experiences of this technique [11, 12].

Here, we intended to verify the feasibility of pediatric robot-assisted resection of a choledochal cyst (RCC) using the da Vinci Si and Xi systems (Intuitive Surgical Inc., Sunnyvale, CA, USA), with special emphasis on the improvements and refinements of surgical outcomes through accumulated experiences in a single tertiary center.

Materials and methods

Study design

The Institutional Review Board of the Severance Hospital of Yonsei University (No. 4-2020-0157) approved this study and waived the requirement for written informed consent from the patients to access their medical records. We retrospectively reviewed and analyzed 160 children who underwent RCC in Severance Children's Hospital of Yonsei University College of Medicine between July 2008 and January 2021. Patients with a body weight of more than 6 kg during the study period were included regardless of the type of Todani classification or presence of perforation of the CC. We excluded two patients because they underwent simultaneous resection of other organs during robotic surgery (partial hepatectomy in one case and splenectomy in the other case). The remaining 158 patients were divided into the first period (P1, July 2008–March 2016; $N=79$) and second period (P2, April 2016–January 2021; $N=79$) with equal number of participants. All surgical procedures were performed by the same experienced pediatric surgeon (S. J. Han.).

Preoperative investigations included routine blood examination, ultrasonography, magnetic resonance imaging (MRI), and hepatobiliary scintigraphy if necessary. Data regarding sex, age at the time of RCC, body weight at the time of the operation, types of Todani classification, and follow-up period as the time from the operation to the end of the study were obtained retrospectively from the medical records. The perioperative and postoperative variables evaluated were operative time, estimated blood loss, open conversion rate, reconstruction method of Roux-en-Y jejunojejunostomy, length of hospital stay, morbidity, and readmission rate in 30 days after the RCC.

Surgical procedure for robot-assisted resection of choledochal cyst

All RCCs were performed using the da Vinci Si and Xi systems. Each robotic surgery was performed as previously reported [11]. The patients were placed supine in a 15° reverse Trendelenburg position with the elevation of the whole-body trunk using sponge pads. Five ports were used, including two 12-mm ports and three 8-mm robotic arm ports. The 12-mm trocar was inserted into the umbilicus with an open technique, and a pneumoperitoneum was established up to 12 mmHg. A 30° robotic camera was inserted through the umbilical port, and a laparoscopic exploration was performed throughout the abdominal cavity.

In most cases, the jejunojejunostomy was made by an extracorporeal hand-sewn anastomosis, although in selected cases, a side-to-side jejunojejunostomy was created with an endoscopic staple firing intracorporeally. The jejunal Roux limb was fashioned extracorporeally after identifying and marking the proximal jejunum 20–30 cm from the ligament of Treitz under optic view. After capturing the proximal jejunum with a laparoscopic grasper, the umbilical port incision was extended vertically by approximately 2–3 cm, and marked jejunum was exteriorized from the abdominal cavity. The jejunojejunostomy was made using 5/0 or 4/0 absorbable multifilament interrupted sutures. The Roux limb was returned to the abdomen, and the umbilical wound was partially closed to fit a 12-mm trocar. In cases of intracorporeal jejunojejunostomy anastomosis, the jejunum was divided with an endoscopic linear stapling device about 20–30 cm from the duodenal-jejunal junction. The mesentery of the jejunum was divided with either an energy device or a stapling device to obtain enough length of the Roux limb. Two enterotomies were made on the antimesenteric borders. A side-to-side isoperistaltic stapled jejunojejunostomy was created between the two jejunal segments by aligning with a traction suture. The remaining enterotomy site at the proximal end of the stapler was then closed using intracorporeal interrupted absorbable sutures.

Pneumoperitoneum was re-established, the robotic system was docked over the head of the patient, and the assistant surgeon stood between the patient's left side and the anesthesiologist. The assistant's 12-mm trocar was positioned in the left lower quadrant for suction, passing on needles, handling adjacent tissue, and extracting the resected tissues using laparoscopic instruments. The surgeon grasped and retracted the gallbladder and falciform ligament in the cephalad direction by two or three percutaneous hitch-stitches. An electric cauterization was used to free and then to resect the CC as proximal as possible to the distal common channel. The groove between the pancreas and CC could be identified when a robotic arm retracted a lower portion of the cyst and the duodenum was pulled down by the assistant surgeon. Intraoperative distal cholangiopancreatography (IOC) could be adopted to identify the proper resection line and to avoid injuring the pancreatic duct in some patients with complex distal common bile duct (CBD) anatomy. Saline irrigation of the distal duct was conducted to clear out the stones and protein plugs, if found. Once the transection line of the distal

CC was determined, the distal stump was ligated using endoloops or Hem-o-lok polymeric clips (Teleflex, Morrisville, NC) and then resected. Thereafter full-thickness transection of the proximal cyst was performed closest to the level of the common hepatic duct bifurcation by a sharp robotic Metzenbaum scissor. The Roux limb was passed up to the hepatic hilum through the retrocolic route. End-to-side hepaticojejunostomy was accomplished with 5/0 or 6/0 absorbable multifilament interrupted sutures. Additional tacking sutures between the serosa of the jejunum and capsule of hilum were added to both ends of the hepaticojejunostomy to prevent kinking of Roux limb. The residual mesenteric defect was closed using 5/0 absorbable multifilament interrupted sutures. One or two drains were left near the hepaticojejunostomy and/or pelvic cavity and removed under the surgeon's discretion in the ward. Routine postoperative ultrasonography was performed 7 days after the surgery, to detect residual intrahepatic bile duct dilatation and intra-abdominal fluid collection.

Statistical analyses

Statistical software (SPSS, version 25.0, Chicago, IL) was used for statistical analyses. A p value < 0.05 was considered as a statistically significant difference. Qualitative variables were expressed as numbers (n) and percentages, and quantitative variables as means ± standard deviations. The normality of distribution was verified with the Kolmogorov–Smirnov test and graphically checked with a histogram. Categorical variables were compared with the Fischer exact or Chi-squared test as appropriate; quantitative variables were compared between groups with the Student's t test.

Results

Patient characteristics

During the study period, 158 consecutive pediatric patients underwent RCC at our institution and met our study criteria. The baseline demographics and outcomes of these patients are summarized in Table 1. The two groups of patients (P1 and P2 groups) were comparable in age, sex, the incidence of cesarean section, gestational age, weight at operation, and birth weight (all $p > 0.05$). The body weight at the operation was lower in the P2 group than in the P1 group (15.9 kg vs. 19.2 kg, $p = 0.073$). However, this trend of lower body weight was not statistically significant. The proportion of Todani classification for type IV was higher in the P2 group than in the P1 group (51.9% vs. 31.6%, $p = 0.015$).

Perioperative and postoperative characteristics of the P1 and P2 groups

The perioperative and postoperative characteristics are summarized in Table 2. All patients who underwent RCC had a complete excision on the final pathologies. There was a statistically significant reduction in operative time between the two groups. Patients in the P2 group had a significantly shorter total operative time and console time than those in the P1 group (462.6 vs. 383.6 min, and 252.2 vs. 190.1 min, respectively, $p < 0.001$). The amount of bleeding during the RCC was 63 mL in the P1 group and 28 mL in the P2 group ($p = 0.025$). The incidence of postoperative intensive care unit admission was 32.9% after RCC in the P1 group and 11.4% after RCC in the P2 group ($p = 0.001$).

Table 1 Clinicopathological findings of patients categorized by the period

Variable	Period 1 ($n = 79$)	Period 2 ($n = 79$)	p value
Age (years)	4.56 ± 4.23	3.71 ± 3.26	0.158
Sex (male)	20 (25.3)	21 (26.6)	1.000
Cesarean section	30 (38.0)	31 (39.2)	1.000
Gestational age (weeks)	39.0 ± 1.7	38.8 ± 2.1	0.442
Birth weight (kg)	3.2 ± 0.5	3.1 ± 0.5	0.380
Prenatal diagnosis	7 (8.9)	10 (12.7)	0.609
Mean weight at operation (kg)	19.2 ± 13.4	15.9 ± 8.8	0.073
Median weight at operation (kg)	14.8 [6.3–72.0]	13.5 [6.3–47.7]	0.308
Weight less than 10 kg at operation	11 (13.9)	16 (20.3)	0.291
Todani type			0.015*
1	54 (68.4)	38 (48.1)	
4	25 (31.6)	41 (51.9)	
Follow-up period (months)	92.9 ± 20.0	32.7 ± 15.3	< 0.001*

Values are presented as numbers (%), means ± standard deviations, or medians [ranges] accordingly

*Represents statistically significant p values, ($p < 0.05$)

Table 2 Perioperative and postoperative characteristics

Variable	Period 1 ($n=79$)	Period 2 ($n=79$)	p value or comments
Total operation time (min)	462.6 ± 91.5	383.6 ± 92.4	< 0.001*
Total console time (min)	252.2 ± 78.1	190.1 ± 62.5	< 0.001*
Bleeding (mL)	63 ± 129	28 ± 54	0.025*
Postoperative stay (days)	8.4 ± 3.1	8.2 ± 3.5	0.736
Postoperative ICU stay	26 (32.9)	9 (11.4)	0.001*
Extracorporeal Roux-en-Y reconstruction	75 (94.9)	69 (87.3)	0.160
Open conversion	1 (1.3)	1 (1.3)	1.000
Postoperative complication	14 (17.7)	8 (10.1)	0.250
Biliary complication	4	1	2 requiring PTBD
Twisted Roux limb	0	2	1 requiring redo hepaticojejunostomy
Intestinal obstruction	3	2	All requiring reoperation
Intestinal perforation	1	0	Laparoscopic primary repair
Intra-abdominal fluid collection	1	1	1 requiring percutaneous drainage
Hydrothorax	1	0	Central venous catheter dislocation
Wound complication	4	1	2 requiring reoperation
Pancreatitis	0	1	Settled conservatively
Reoperation within 30 days	6 (7.6)	4 (5.1)	0.746
In 30-days ED visit	11 (13.9)	3 (3.8)	0.047*
Abdominal pain	6	3	1 requiring PTBD 2 requiring reoperation
Fever	3	0	1 requiring percutaneous drainage
Wound discharge	2	0	Settled conservatively

Values are presented as numbers (%) or means ± standard deviations

ICU Intensive care unit, *ED* Emergency department, *PTBD* Percutaneous transhepatic biliary drainage

*Represents statistically significant p values, ($p < 0.05$)

Five patients had postoperative biliary complications, with four patients in the P1 group. Of these, 2 patients required a percutaneous transhepatic biliary drainage (PTBD), and 3 recovered from minor bile leakage conservatively. Postoperative wound complications occurred in five cases. All five patients with intestinal obstructions required reoperation. There was only one case of intestinal perforation, which was repaired by a laparoscopic approach. Other complications included iatrogenic hydrothorax after central venous catheter dislocation during anesthesia ($n=1$), intra-abdominal fluid collection ($n=2$), pancreatitis ($n=1$), and a twisted Roux limb ($n=2$). Revisional surgery was performed in one patient with a twisted Roux limb; the remaining patient gained full oral intake without additional surgery.

The rate of emergency department visits after the RCC was higher in the P1 group (13.9% vs. 3.8%, $p=0.047$). Of the 14 patients who visited the emergency department within 30 days after surgery, 9 complained of abdominal pain, accounting for 64.3% of the total. Of these, 2 patients underwent reoperation for intestinal obstruction, and 1 underwent PTBD due to bile duct stricture. There was one conversion case in each group (technical problems with an inadequate view in early P1 and a twisted Roux limb in P2). However, the overall rates of postoperative complications, reoperation, and open conversion were similar between both groups.

Perioperative and postoperative characteristics under 10 kg

See Table 3 for outcomes of children after RCC under 10 kg of body weight. The P1 group had longer total operative times (424.6 min vs. 334.7 min, $p=0.002$) and longer console times (200.1 min vs. 147.6 min, $p=0.003$). Postoperative complications occurred in only one case in the P1 group. The patient was diagnosed with stenosis of the hepaticojejunostomy 2 months after RCC, and PTBD was performed, and the inserted catheter was removed in 5 months. The patient's course was uneventful during the 9-year follow-up. There was no open conversion and no reoperation in either group. There were no statistically significant differences in other perioperative and postoperative variables between the two groups.

Table 3 Patients' perioperative and postoperative characteristics under 10 kg

Variable	Period 1 ($n=11$)	Period 2 ($n=16$)	p value
Total operation time (min)	424.6 ± 37.0	334.7 ± 78.5	0.002*
Total console time (min)	200.1 ± 36.5	147.6 ± 44.5	0.003*
Bleeding (mL)	8 ± 8	11 ± 9	0.406
Postoperative stay (day)	7.2 ± 0.6	7.4 ± 0.9	0.535
Postoperative ICU stay	5 (45.5)	4 (25.0)	0.411
Extracorporeal Roux-en-Y reconstruction	11 (100)	13 (81.2)	0.248
Open conversion	0 (0)	0 (0)	1
Postoperative complication	1 (9.1)	0 (0)	0.407
Biliary complication	1 (9.1)	0 (0)	0.407
Reoperation within 30 days	0 (0)	0 (0)	1
In 30-days ED visit	0 (0)	0 (0)	1

Values are presented as numbers (%) or means ± standard deviations

ICU Intensive care unit, *ED* Emergency department

*Represents statistically significant p values, ($p<0.05$)

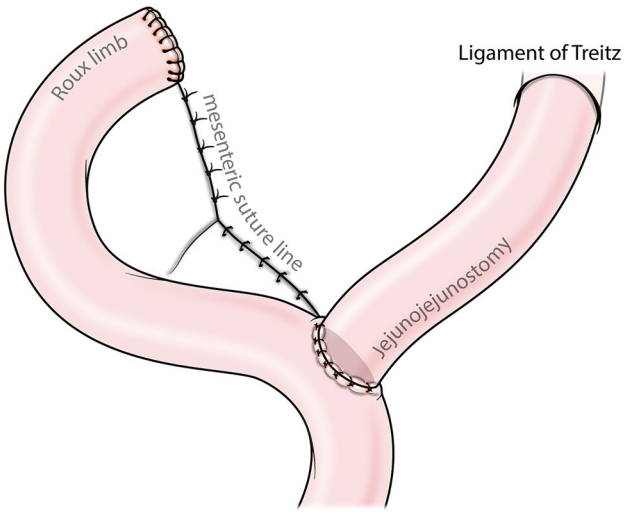

Fig. 1 Ideal Roux-en-Y jejunojejunostomy anastomosis and identification of the mesenteric suture line for preventing complications related to Roux-en-Y jejunojejunostomy. (The demonstration of mesenteric suture line is available in the Supplementary video at 4:40 s of the video.)

Discussion

To the best of our knowledge, this study represents the largest single institutional cohort assessment of long-term outcomes after pediatric RCC to date. We implemented a novel and standardized performance for 158 RCCs under 13 years. Optimization of RCC was established in the early period, allowing for the expansion of selection criteria such as young pediatric patients under 10 kg of body weight and complex bile duct anomalies without adversely affecting patient outcomes. This experience could serve as a model for the safe implementation and adoption of robot-assisted surgery for the resection of pediatric CCs.

A paradigm shift occurs toward minimally invasive surgery as the standard therapeutic option in various pediatric abdominal surgeries. The advantages of robotic systems for the resection of CCs, such as surgeon-controlled three-dimensional view, endo-wristed instruments, could be helpful for the meticulous dissection of the liver hilum and safe resection of the distal CBD without injuring the pancreatic duct. Recent studies have documented the technique and feasibility of RCC, and our results, which revealed low morbidity and superior outcomes, are in line with these studies [8, 9].

Postoperative ICU stay and readmission incidence via emergency department 30 days after the operation were significantly lower in the P2 group than in the P1 group. The reason for the high rate of postoperative ICU care in the P1 group was not because of its high patient comorbidities but because of the need for intensive monitoring for pediatric robotic surgery in the early period. This could be verified because there was no difference in the total hospital stay and complication rates for periods 1 and 2. However, our study showed that the operation time of P1 was 462.6 min and that of P2 was 383.6 min, which was longer than that in prior published reports (192.7 min for Pham et al. and 180.6 min for Xie et al.) [8, 9]. This could be explained by several outlier patients, such as three patients who had severe inflammation with a sealed-off cyst perforation, one patient who had redo hepaticojejunostomy after initial hepaticojejunostomy with a twisted Roux limb, and one patient who had severe intra-abdominal adhesions following prior repair of ileal atresia when he was a neonate. Utilizing IOC and the interrupted hepaticojejunostomy suture method might be another attributable factor for relatively longer operative time. Given these heterogeneous factors, it will be difficult to simply conclude that our operation time is longer than that in other series.

After we encountered twisted Roux limb and redo hepaticojejunostomy cases, we drew lessons to perform a routine check for the mesenteric suture line before retrocolic tunneling of the Roux limb (Fig. 1). When jejunojejunostomy is reconstructed using the extracorporeal method, the accurate identification of the ligament of Treitz may sometimes be confusing. Therefore, we recommend checking that the shape of the mesenteric suture line is formed as a vertical streamline toward the end of the Roux limb by the robotic intraperitoneal view. (The demonstration of the mesenteric

suture line is available in the Supplementary video at 4:40 s of the video.)

Subset analysis of the 26 patients who weighed under 10 kg confirmed that RCC could be safely adopted and showed superior outcomes in our cohort. There are no studies specifically evaluating RCC outcomes with pediatric patients under 10 kg after the report from Dawrant et al. [13]. The total operative and console times were shorter in the subset P2 group than in the subset P1, similar to the overall P1 and P2 cohort results. The minimum body weight of our study participants was 6.3 kg. Seven patients weighed less than 7 kg, of whom five were included in the P2 group. Patients with an extremely small body weight tend to be operated more on in the P2 period. Robot-assisted surgery performed in these patients is difficult due to the narrow space in the abdominal cavity. However, RCC for such patients is often decided before the inflammation becomes severe; therefore, there is an advantage of less bleeding during surgery. The intestine diameter is small, which is more advantageous for patients weighing less than 10 kg when performing extracorporeal jejunojejunostomy. Xie et al. also did not recommend intracorporeal stapling jejunojejunostomy in patients with an extremely small body weight because space is insufficient to use the stapler in the abdominal cavity [14]. Moreover, while there are some difficulties such as applying 8-mm robotic working ports and a small intra-abdominal working space, increased attention for the placement of trocar with a proper angle may mitigate collisions among robotic arms and avoid inadvertent visceral injury. Expanded criteria of pediatric robot-assisted surgery for smaller babies could be achieved by further miniaturization of the robotic system. Currently, the da Vinci SP surgical system has been developed as a novel robotic platform for successfully performing robotic single-site surgery in various adult operations. As this kind of new technology evolves, we will need to continue investigating the benefits of pediatric robot-assisted surgery.

One of the severe long-term complications after resection of the CC is subsequent malignant transformation following incomplete resection of the intra-pancreatic portion of the CC. The remnant distal CC might be related to the pathogenesis of biliary carcinogenesis, with an incidence estimated to be 0.7–6.0% [15]. Xia et al. reported that the malignancy rate was 15% (6/41) among the remnant intra-pancreatic CCs, and the mean time between original incomplete resection of CC and the diagnosis of malignancy was 140 months [16]. Endoscopic cholangioscopy might be helpful in identifying the location of the pancreatic duct to ensure maximal resection of the distal CBD without pancreatic injury. In our center, the status of the CC was routinely assessed using preoperative magnetic resonance cholangiopancreatography (MRCP) (Fig. 2). In doubtful cases such as the presence of anomalous pancreaticobiliary ductal union (APBDU), which was hard to discern, or pancreatic duct anomalies, IOC was performed after irrigation and evacuation of multiple protein plugs or stones (Fig. 2). This procedure is advantageous because invasive procedures such as preoperative endoscopic retrograde cholangiopancreatography can be omitted, and complex anatomical structures that are not well-discriminated in MRCP can be clearly observed. IOC can be easily performed while maintaining the robotic trocars in place during RCC procedures. Fortunately, remnant intrapancreatic bile duct or progression to malignancies was not observed during the follow-up period. The use of IOC is our important management strategy to eliminate the possibility of remnant bile duct tissue or protein plugs.

The goal of the minimally invasive resection of the CC is to provide patients with the benefits of a minimally invasive approach while maintaining the same anatomical resection and reconstruction with conventional open resection of the CC. Throughout the study period, our treatment strategy consistently followed the principles mentioned above. As our teams gained surgical experience using robots, the indication for RCC has not been intentionally expanded. Improved results have widened the indications for RCC over time, including pediatric patients with various comorbidities and patients with various body weights. The senior surgeon (S. J. Han.) of this study already possessed high-volume surgical experiences of other kinds of robotic surgery and also had been actively engaged in open and laparoscopic pediatric hepatobiliary surgeries.

As with any study conducted using a retrospective single-center review, there were some limitations to analyzing and interpreting its results. Although our study includes the largest cohort numbers of pediatric RCC, it was limited to a single surgeon's experience; nonetheless, this could be a positive aspect considering the surgical procedure was consistent throughout the entire study period. Although most patients diagnosed with CC in our center were enrolled in this study, patients with prominent obstructive cholestasis weighing less than 6 kg inevitably underwent open resection of the CC. In addition, patients who decided to undergo open or laparoscopic resection due to various reasons may have introduced selection bias. However, there was no exclusion criterion except for the body weight of 6 kg in determining whether or not to proceed with robot-assisted surgery. Even if our results may not be generalizable, the inclusion of a broadly representative pediatric patient cohort not only increased statistical power but also strengthened the validity of our findings. Therefore, our results might be of interest to more surgeons who are willing to improve the quality of minimally invasive pediatric surgery.

In summary, this study represents the largest analysis of pediatric RCC to date. Our data suggest continued improvements beyond an initial steep learning curve, with no compromise in postoperative and long-term outcomes after

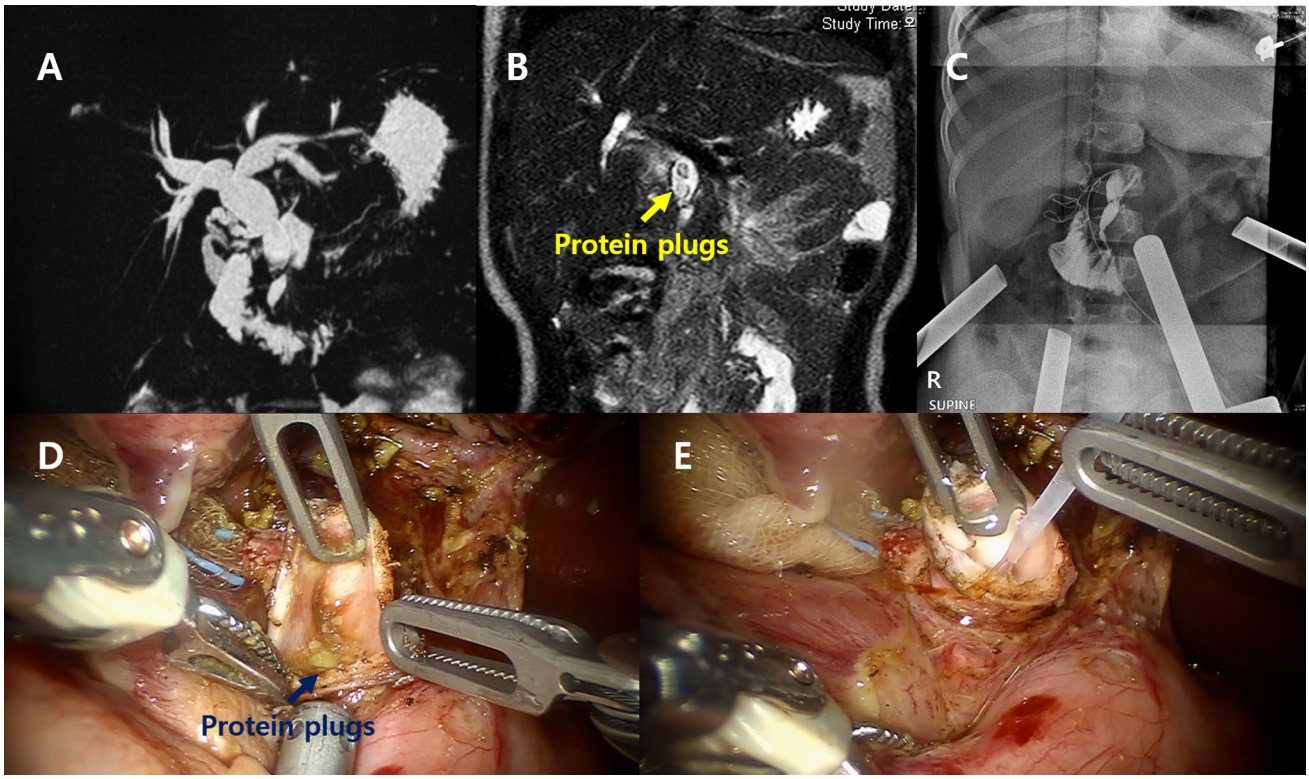

Fig. 2 Representative MRI, IOC, and optic findings of the distal CBD during robotic choledochal cyst excision for the same patient. **A**, **B** MRI shows cystic dilatation of the extrahepatic bile duct and proximal intrahepatic bile duct with anomalous pancreaticobiliary ductal union (APBDU), suggesting choledochal cyst type 4a. Multiple protein plugs or stones are observed just above APBDU. **C** IOC shows the disappearance of the CBD protein plugs or stones after saline irrigation. **D**, **E** Magnified optic findings of protein plug in the distal CBD before and after saline irrigation

gradual expansion of selection criteria. This study provides evidence for the use of robot-assisted surgery to improve outcomes in patients with pediatric CCs.

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Declarations

Disclosures Kyong Ihn, In Geol Ho, Young Ju Hong, Ho Jong Jeon, Dongeun Lee and Seok Joo Han have no have no relevant financial conflicts of interest to disclose.

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