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Laparoscopic common bile duct exploration to treat choledocholithiasis in situs inversus patients: A technical review

Bo-Ya Chiu, Shu-Hung Chuang, Shih-Chang Chuang, Kung-Kai Kuo

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Abstract

Situs inversus (SI) is a rare congenital condition characterized by a mirror-image transposition of the major visceral organs. Since the 1990s, more than one hundred SI patients have been reported to have successfully undergone laparoscopic cholecystectomy. In these cases, the major problem is to overcome is the left-right condition for right-handed surgeons. Laparoscopic common bile duct exploration (LCBDE), an alternative to treat patients with bile duct stones, has shown equivalent efficacy and is less likely to cause pancreatitis than endoscopic retrograde cholangiopancreatography. Recent updated meta-analyses revealed that a shorter postoperative hospital stay, fewer procedural interventions, cost-effectiveness, a higher stone clearance rate, and fewer perioperative complications are additional advantages of LCBDE. However, the technique is technically demanding, even for skilled laparoscopic surgeons. Conducting LCBDE in patients with difficult situations, such as SI, is more complex than usual. We herein review published SI patients with choledocholithiasis treated by LCBDE, including our own experience, and this paper focuses on the technical aspects.

Key Words: Choledocholithiasis; Choledochotomy; Laparoscopic common bile duct exploration; Single incision; Situs inversus; Transcystic

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Core Tip: Laparoscopic common bile duct exploration (LCBDE) is an alternative option to treat choledocholithiasis. Compared to endoscopic retrograde cholangiopancreatography, it has been demonstrated that LCBDE has resulted in shorter hospital stays, fewer procedures, and greater cost-effectiveness in recent studies. Nevertheless, LCBDE is a technically demanding procedure. It is even more challenging in difficult circumstances such as situs inversus (SI). Herein, we present an analysis of published SI patients with choledocholithiasis treated by LCBDE and our own case, and this paper focuses on the technical aspects.

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INTRODUCTION

For more than two decades, laparoscopic common bile duct exploration (LCBDE) and endoscopic retrograde cholangiopancreatography (ERCP) with stone removal have been widely accepted techniques for managing common bile duct (CBD) stones[1-3]. As LCBDE is a technically demanding procedure, ERCP has gradually become mainstream in clinical practice. In 2018, an updated network meta-analysis including 13 trials and 1757 patients revealed that laparoscopic cholecystectomy (LC) plus LCBDE had better outcomes than LC plus ERCP in terms of not only length of hospital stay but also stone clearance rate and perioperative complications[4]. Other studies exhibited inconsistent results[3,5,6]. The trend in favor of ERCP resulted in decreased familiarity with LCBDE by surgeons. The complex steps and delicate tools used in LCBDE also make it challenging to perform.

Situs inversus (SI) is a rare condition causing left-right positioning of thoracic and abdominal organs [7]. Clinically, SI by itself is asymptomatic; however, when it is associated with CBD stones, the diagnosis can be challenging due to the reversed anatomical location[8]. This is also true for LC and LCBDE. As LC in SI has been studied in two meta-analyses[9,10], LCBDE in SI is limited to a small number of case reports. This review is aimed at assessing published SI patients treated by LCBDE and shares our own experience in the technical aspects.

LITERATURE REVIEW AND OUR OWN EXPERIENCE

Data collection

A search for all articles regarding CBD exploration in patients with SI was conducted in The Cochrane Library, PubMed, Embase, and Web of Science without language restriction until October 5, 2022. The keywords used were "Situs inversus" plus "laparoscopic bile duct exploration", "choledocholithotripsy", "choledocholithotomy", "choledocholithiasis", and "bile duct stone". All relevant studies reporting one or more cases of CBD stone exploration in SI that were found as full texts, structured abstracts or conference reports were included. Studies that described procedures conducted in open surgery or other than CBD exploration were excluded. Duplications of the same patient reported in different studies were also excluded. A total of 12 records were identified by the literature search. In accordance with the study exclusion criteria, two articles were excluded for reporting the same patient [11,12], and the other two articles were excluded for open surgery[13,14]. Finally, eight case reports were included in this review[8,15-21]. The detailed information is presented in Tables 1-3.

Our own experience

A 79-year-old female with SI had underlying hypertension, type II diabetes mellitus, dyslipidemia, and an operation history of ectopic pregnancy 50 years prior. This time, she suffered from postprandial epigastric pain accompanied by a jaundice episode 5 d before her medical seeking. At the emergency room, physical examination was unremarkable. Laboratory values showed white blood cell 7,200/mm (normal range: 4000-10000), C-reactive protein 33.31 mg/dL (0-8), total bilirubin 1.17 mg/dL (1.0-2.0), direct bilirubin 0.5 mg/dL (0.2-1), alanine transaminase 39 IU/L (10-40), aspartate aminotransferase 89 IU/L (10-42), and alkaline phosphatase 147 IU/L (28-94). Abdominal computed tomography (CT) showed cholelithiasis and choledocholithiasis in the distal CBD (Figure 1A and B). Other anomalies included distal CBD draining into the duodenal 3rd portion, duplication of the right ureter, and right hydroureteronephrosis (Figure 1A and C). Under the impression of acute calculous cholecystitis and choledocholithiasis with obstructive jaundice and liver function impairment, she was admitted for single-incision LC and LCBDE. The detailed surgical procedure is discussed below, and the patient's

Table 1 Preoperative characteristics of situs inversus patients treated by laparoscopic bile duct exploration in the literature review

| Ref. | Year | Age | Sex | Medical history | Presentation | Diagnostic tools for CBD stone | Preoperative intervention |
|---------------------------------|------|-----|-----|---|--|--------------------------------|---------------------------|
| Kang <i>et al</i> [21] | 2004 | 64 | F | Atrial fibrillation | Jaundice, abnormal liver function | Echo, CT, MRCP | Nil |
| Tai <i>et al</i> [20] | 2004 | NA | F | Arrhythmia with pacemaker implantation | NA | NA | ERCP |
| Weber-Sánchez <i>et al</i> [15] | 2011 | 60 | M | NA | Jaundice, cholecystitis | NA | NA |
| Han <i>et al</i> [16] | 2012 | 71 | F | NA | NA | CT | ERCP |
| Liu <i>et al</i> [8] | 2017 | 51 | F | Nil | Cholecystitis, abnormal liver function | CT, MRCP | Nil |
| Senthilnathan <i>et al</i> [17] | 2017 | 76 | F | Gallbladder stone s/p open cholecystectomy | NA | NA | ERCP |
| Takalkar <i>et al</i> [19] | 2018 | 50 | F | NA | Jaundice, abnormal liver function, cholecystitis | Echo, MRCP | ERCP + ERBD |
| Simkhada <i>et al</i> [18] | 2021 | 63 | F | HTN, hypothyroidism, CKD | NA | MRCP | ERCP |
| Our case | 2022 | 79 | F | HTN, hyperlipidemia, DM, CKD, ectopic pregnancy | Jaundice, abnormal liver function, cholecystitis | CT | Nil |

F: Female; M: Male; s/p: Status post; HTN: Hypertension; DM: Diabetes mellitus; CKD: Chronic kidney disease; CT: Computed tomography; MRCP: Magnetic resonance cholangiopancreatography; ERCP: Endoscopic retrograde cholangiopancreatography; ERBD: Endoscopic retrograde biliary drainage; NA: Not available; CBD: Common bile duct.

postoperative recovery was uneventful. She was discharged on the 3rd postoperative day. No complications occurred after a 24.5-mo follow-up.

The single-incision LCBDE (SILCBDE) technique has been described in detail previously[22,23]. The patient was given general anesthesia and placed in the reverse Trendelenburg position. The surgeon and the assistant stood on the right side of the patient owing to the mirror positioning of the intraabdominal organs. The surgery began with the insertion of a commercial multichannel port through a 2 cm left paraumbilical incision. A 5-mm atraumatic grasper was inserted through the left 5-mm port to retract the gallbladder to the 2 o'clock direction. A 5-mm 50-cm-long 30-degree laparoscope was passed through the lower 12-mm port to provide visualization. The working ports were served by the upper 12-mm and right 5-mm ports. After dissecting Calot's triangle (Figure 2A) and securing the proximal cystic duct, a small incision was made into the cystic duct, and a 5 French feeding tube was passed into the nondilated cystic duct (Figure 2B and C). We conducted a diagnostic intraoperative cholangiogram (IOC), and distal CBD stones were confirmed (Figure 3A). Then, we used a modified technique named "basket-in-catheter"(BIC)[24]: The Dormia basket was inserted into a six French feeding tube and then went forward to the predestined distance. After entering the distal CBD, the basket was opened to trap the stone (Figures 2B and 3B). After stone clearance, completion IOC was performed to confirm that there were no retained stones (Figure 3C). Finally, the cystic duct was closed, and the gallbladder was detached from the liver bed (Figure 2D) with a closed suction drain left in the subhepatic space. The surgery took 152 min with an estimated blood loss of 5 ml. The details of the operation are shown in the Video.

ACUTE BILIARY TRACT DISEASE IN SITUS INVERSUS PATIENTS

Overview of *Situs inversus*

SI is a rare defect of situs orientation. As SI fails to generate normal left-right asymmetry, it results in a spectrum of laterality disturbances[25]. The most common of these is immotile cilia syndrome, known as Kartagener syndrome[26-28]. Ivemark's syndrome, cardiac malformation and biliary atresia are also found in patients with SI[29-31]. In our case, duplication of the right ureter and abnormal insertion of the distal CBD into the 3rd portion of the duodenum were found on CT (Figures 1 and 3C). Although their association with SI remains unclear[32,33], we should always pay attention to SI patients to identify possible anomalies in preoperative surveys.

Table 2 Intraoperative characteristics of situs inversus patients treated by laparoscopic bile duct exploration in the literature review

| Ref. | Year | IOC | Incision number | Style | Position | Surgeon site | Working hand | Choledochoscopy | CBDE route | Approach technique |
|---------------------------------|------|-----|-----------------|--------|-----------------------|--------------|--------------|-----------------|--------------------------------------|--------------------------|
| Kang <i>et al</i> [21] | 2004 | N | 4 | MirA | NA | Right side | Left | Yes | Choledochotomy with T-tube closure | Choledochoscopy |
| Tai <i>et al</i> [20] | 2004 | N | 4 ^a | NA | NA | NA | NA | NA | NA | NA |
| Weber-Sánchez <i>et al</i> [15] | 2011 | N | 4 ^a | NA | NA | NA | NA | NA | NA | NA |
| Han <i>et al</i> [16] | 2012 | N | 4 | MirCon | NA | NA | NA | Yes | Choledochotomy | Choledochoscopy |
| Liu <i>et al</i> [8] | 2017 | N | 4 ^a | NA | NA | NA | NA | NA | NA | NA |
| Senthilnathan <i>et al</i> [17] | 2017 | Y | 4 | MirCon | Supine, split legs | Between legs | NA | No | Choledochotomy with CDD ^b | Balloon and saline sweep |
| Takalkar <i>et al</i> [19] | 2018 | N | 4 | MirA | NA | NA | NA | No | Choledochotomy with CDD ^c | NA |
| Simkhada <i>et al</i> [18] | 2021 | N | 4 | MirA | Lithotomy | Right side | Left | Yes | Choledochotomy with primary closure | Endo-forceps |
| Our case | 2022 | Y | 1 | SP | Reverse Trendelenburg | Right side | Right | No | Transcystic approach | Basket in catheter |

^aDefined as conventional in study.^bUnderwent laparoscopic cholecystectomy six months prior. Found common bile duct stone and small residual gallbladder containing stone this time.^cDue to significant dilation of the common bile duct (> 2 cm).

IOC: Intraoperative cholangiogram; MirA: Mirrored American; MirCon: Mirrored conventional; SP: Single-port. CDD: Choledochoduodenostomy; CBDE: Common bile duct exploration; NA: Not available; N: No; Y: Yes.

Table 3 Operative results of situs inversus patients treated by laparoscopic bile duct exploration in the literature review

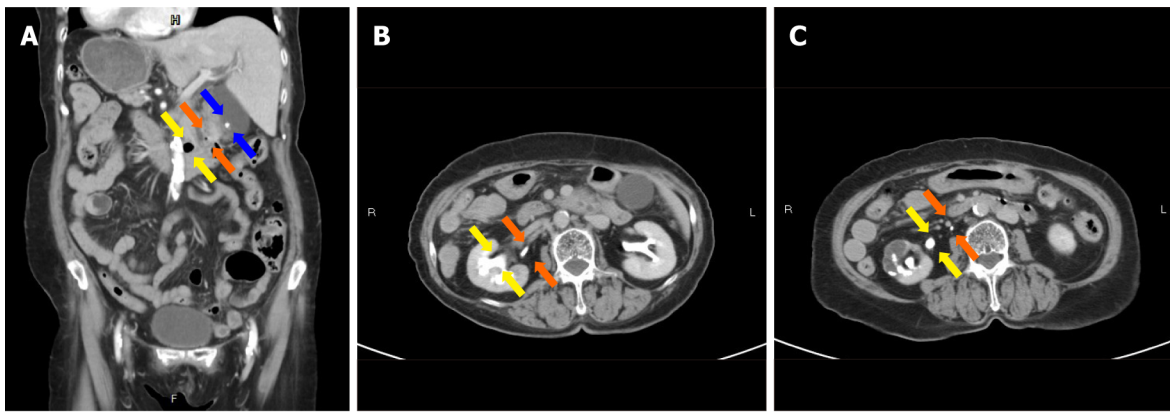
| Ref. | Year | OP time (min) | Conversion | PLOS (d) | Complication | Mortality | Follow-up (mo) |
|---------------------------------|------|---------------|------------|----------|--------------|-----------|----------------|
| Kang <i>et al</i> [21] | 2004 | 240 | No | NA | NA | Nil | Nil |
| Tai <i>et al</i> [20] | 2004 | NA | No | NA | Nil | Nil | Nil |
| Weber-Sánchez <i>et al</i> [15] | 2011 | NA | No | NA | NA | Nil | Nil |
| Han <i>et al</i> [16] | 2012 | 129 | No | NA | Nil | Nil | Nil |
| Liu <i>et al</i> [8] | 2017 | NA | No | 5 | Nil | Nil | Nil |
| Senthilnathan <i>et al</i> [17] | 2017 | NA | No | 7 | Nil | Nil | Nil |
| Takalkar <i>et al</i> [19] | 2018 | NA | No | NA | NA | Nil | Nil |
| Simkhada <i>et al</i> [18] | 2021 | NA | No | NA | NA | Nil | Nil |
| Our case | 2022 | 152 | No | 3 | Nil | Nil | 24.5 |

OP: Operative; PLOS: Postoperative length of hospital stay; NA: Not available.

Diagnosis

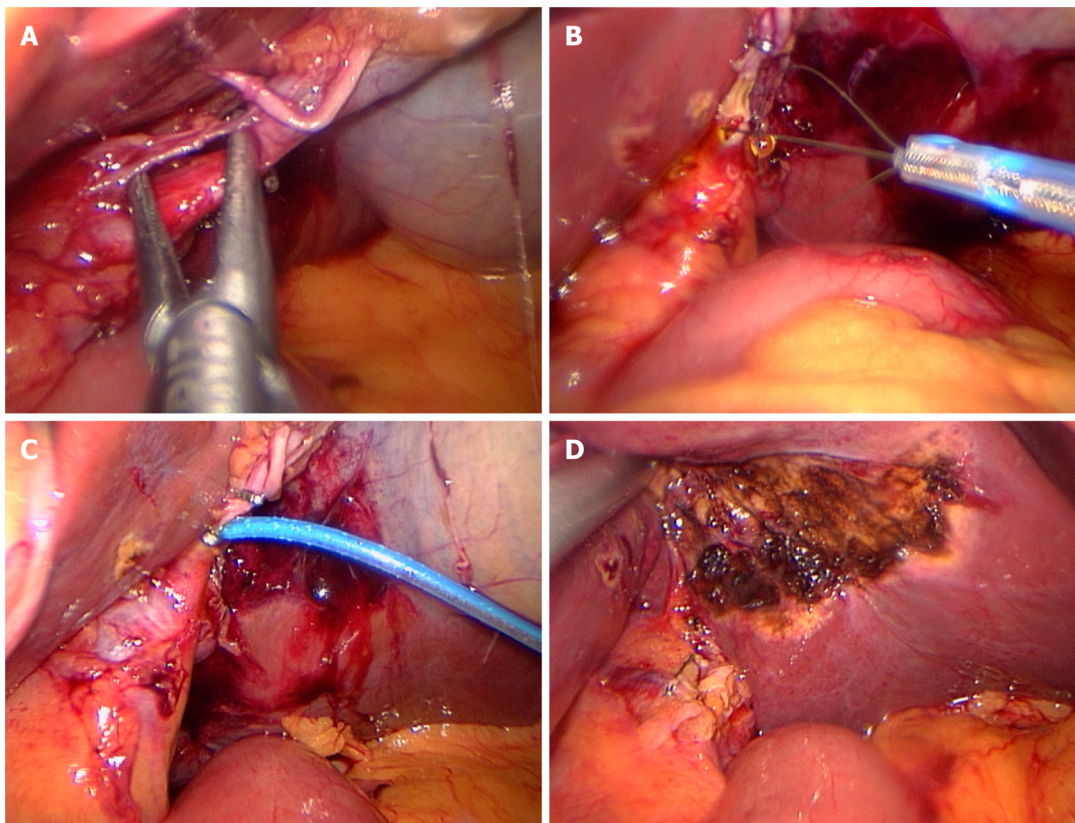
A total of 9 case reports including our case, was comprised of eight women and one man with ages ranging from 50 to 79 years of age, as detailed in Table 1. Systemic disease was reported in four patients [18,20,21], and one of them was equipped with a pacemaker[20]. One patient had an ectopic pregnancy history. Also, one patient had undergone LC before, but residual gallbladder and CBD stones were found six months later[17]. The presentation in four of these patients was jaundice[15,19,21], while four patients developed cholecystitis[8,15,19]. Additional high-resolution imaging was used to assess the abdominal anatomy in six patients: Two patients underwent abdominal CT only[16], magnetic resonance cholangiopancreatography (MRCP) was used in two cases[18,19], and two patients underwent both CT and MRCP[8,21].

Due to the left-right presentation of SI, a timely diagnosis of acute abdominal diseases in SI patients is difficult for emergency physicians[34,35]. Liu *et al*[8] reported a 15-d delay diagnosis of acute cholangitis



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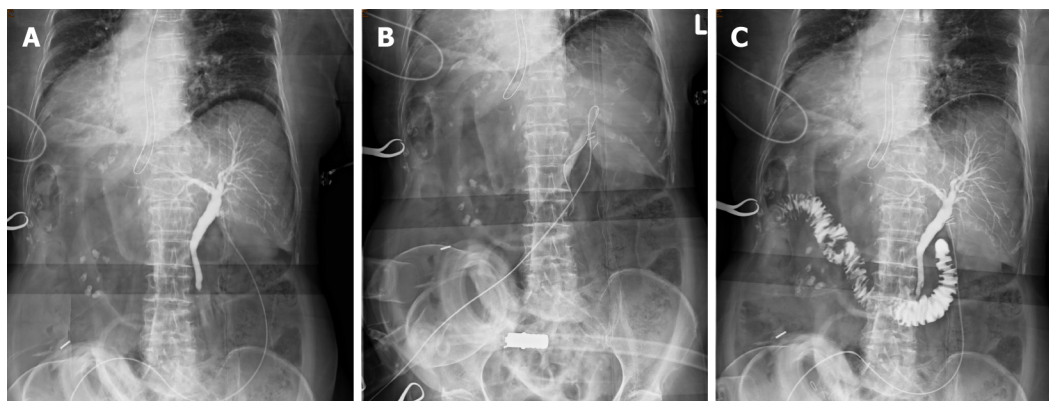
Figure 1 Preoperative computed tomography showed situs inversus, stones in the gallbladder and the common bile duct, and associated anomalies. A: A gallbladder stone (blue arrows), a common bile duct stone (orange arrows) and a diverticulum at the 3rd portion of the duodenum (yellow arrows); B: Right upper ureter of the duplication (orange arrows) and lower pelvis of the duplication (yellow arrows); C: Right upper ureter (orange arrows) and right lower ureter (yellow arrows).



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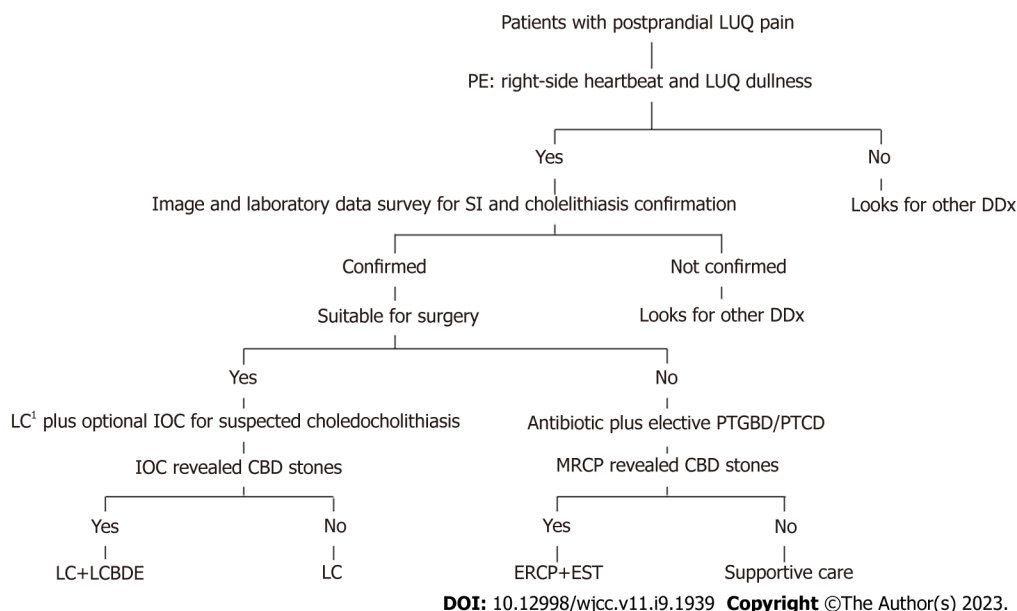
Figure 2 Intraoperative photos during single-incision laparoscopic transcystic common bile duct exploration in a patient with situs inversus. A: Dissection of the hepatocystic triangle; B: An opened retrieval basket in a 6 French feeding tube; C: Cystic duct cannulation for intraoperative cholangiography; D: Completion of choledocholithotripsy and cholecystectomy.

[8]. For patients with postprandial left upper abdominal pain, physical examination is needed to reveal a heartbeat in the right hemithorax and hepatic dullness in the left upper abdomen so that cholelithiasis associated with SI can be considered in addition to acute myocardial infarction or a peptic ulcer. In patients with uncertain features, a careful physical examination with radiological investigations plays a significant role in SI diagnosis[36,37]. Here, we propose a practical algorithm for the diagnosis and management of cholelithiasis and choledocholithiasis in SI patients (Figure 4).



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Figure 3 Intraoperative cholangiograms during transcystic choledocholithotripsy using a modified “basket in catheter” technique. A: The diagnostic cholangiogram showed an obstructive biliary tree; B: Transcystic basket trawling; C: The completion cholangiogram revealed stone clearance and a patent distal common bile duct.



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Figure 4 A practical algorithm for diagnosis and management to treat cholelithiasis and choledocholithiasis in situs inversus patients.

¹Laparoscopic cholecystectomy can be performed with four ports or a single port based on the surgeon's discretion. LUQ: Left upper quadrant; PE: Physical examination; SI: Situs inversus; DDx: Differential diagnosis; LC: Laparoscopic cholecystectomy; IOC: Intraoperative cholangiography; MRCP: Magnetic resonance cholangiopancreatography; PTGBD: Percutaneous transhepatic gallbladder drainage; PTCD: Percutaneous transhepatic cholangiography and drainage; CBD: Common bile duct; LCBDE: Laparoscopic common bile duct exploration; ERCP: Endoscopic retrograde cholangiopancreatography; EST: Endoscopic sphincterotomy.

Endoscopic retrograde cholangiopancreatography vs laparoscopic common bile duct exploration

Patients with SI also pose significant challenges to endoscopists and surgeons[38-42]. ERCP is one of the most challenging procedures in SI patients. Given the reversed anatomy, a 180-degree clockwise rotation in the duodenum is often required[43-47]. Furthermore, cannulation to the bile duct is difficult because of the lack of visualization and the ectopic location of the ampulla of Vater in SI patients[7]. In 2022, Ding *et al*[48] published a case series containing 14 patients with SI undergoing ERCP. The rate of successful cannulation was 85.7% (12/14), while difficult cannulation occurred in 71.4% (10/14) of those patients[48]. Although the literature review until 2021 revealed a 100% cannulation success of 41 patients[48], one of five cases reported in 2022 failed[49-52]. Plus there were 5 failed cases in our literature review, and the overall success rate was 87.7%. Compared to ERCP, LCBDE provides full intraperitoneal visualization. In the five case reports of our literature review, all the patients had successful LCBDE after failed ERCP, and the former might be a better option to treat choledocholithiasis in SI patients.

LAPAROSCOPIC COMMON BILE DUCT EXPLORATION IN SITUS INVERSUS PATIENTS

Outcome

In addition to our case, the operative time was only shown in two more patients: 240 min[21] and 129 min[16]. Postoperative hospital stay was also recorded in two other patients as five days[8] and seven days[17]. While we spent 152 min to complete the SILCBDE, the postoperative hospital stay was only three days. Of these patients, no conversion, complication, or mortality was recorded. The detailed operative results are presented in Table 3.

Trocar positioning

Conducting laparoscopic surgery in SI patients is also difficult owing to the transposed organs[53-55]. The first consideration is trocar positioning. There are essentially two types of laparoscopic port placements in SI patients: The "American mirror technique" and the "French mirror technique"[9,10]. Surgeons can choose either of them depending on their discretion. In most of the case reports in our review, the "American mirror technique" was used, while some authors ambiguously described it as a "conventional technique" that could be the "American mirror technique" or "French mirror technique". The handedness of the surgeons is the second problem[56,57]. Surgeons use their left hand for dissection *via* the epigastric port and use the right hand for the midclavicular port. As most surgeons are right-handed, using the left hand will not be precise and may pose danger. In using the right hand, however, the surgeon will have to cross the patient's body to perform the dissection[10]. To address this dilemma, a systemic review showed that left-handed surgeons yielded shorter intervention times than right-handed surgeons during LC of SI patients[9]. However, there were only seven left-handed surgeons in 121 cases in this study. While most surgical procedures are designed for right-handed surgeons, it is not uncommon for left-handed surgeons to use their right hand as the working hand in daily practice[58, 59]. Using a nondominant hand might not have much impact on the surgical outcome.

In our literature review of LCBDE in SI patients (Table 2), although some of the patients ambiguously described their operative technique as "conventional"[8,15,20], most of them reported a four-port technique with the American mirror style[18,19,21]. Our patient is the only case treated by SILCBDE.

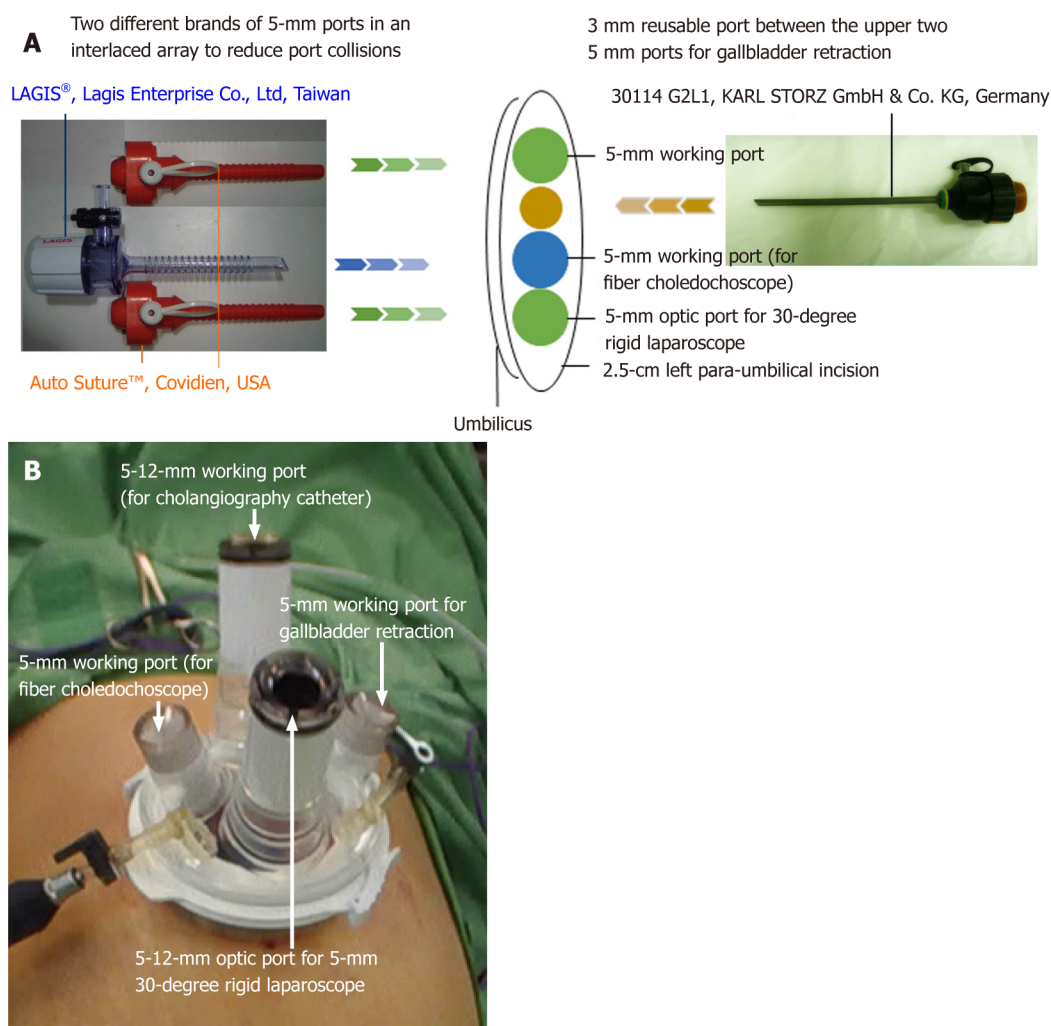
Single-incision laparoscopic common bile duct exploration

Compared with the difficulty in LCBDE for SI patients, SILCBDE seems to have some advantages. We developed this novel technique in July 2012, and it soon became our standard of care for choledocholithiasis, including difficult situations such as Mirizzi syndrome Csendes type II-IV[22,23,60]. In addition to a 5-mm 50-cm-long 30-degree laparoscope and a 5-mm flexible fiber choledochoscope set, only conventional straight laparoscopic instruments were needed. Either the single-incision multiple-port longitudinal-array technique[61] or a commercial multichannel port could be used at the surgeon's discretion (Figure 5). This procedure is indicated for every patient with choledocholithiasis who can tolerate regular laparoscopic surgery. While SILCBDE is selected for SI patients, the mirror position of trocars is unnecessary because only one single port was used. The handedness problem is invalid, as dissection can be performed by the right hand, and the left hand can be used for gallbladder traction[62-65]. It has little adverse effect in our technique as well, while gallbladder traction is usually carried out by the assistant[22,23,66]. Additionally, SILCBDE decreases incisional trauma and postoperative pain, speeds recovery and provides favorable cosmesis. The only modification needed is the positions of operators and assistants, which should be moved from the patient's left side to the right side. Since our surgeon is an experienced surgeon who has performed more than 100 SILCBDEs[22], the risk of major complications is minimal. Our patient also attained the shortest postoperative stay in this review.

Choledochotomy and transcystic approach

Another issue in SI undergoing LCBDE is the approach route. The laparoscopic approach for CBD stones can be categorized into transcystic exploration and choledochotomy[67]. The location, number, and size of the CBD stones along with the anatomy of the cystic duct and the CBD influence the choice between these two techniques[68]. For example, small distal stones (≤ 6 mm diameter) are more suitable for the transcystic approach; choledochotomy can be considered if the CBD is larger than 7 mm or intrahepatic duct stones exist[67]. In our literature review, choledochoscopy was used in three patients to conduct bile duct exploration[16,18,21]. Five patients underwent choledochotomy that closed by one T-tube drainage[21], one patient had primary closure[18], two patients had choledochoduodenostomies[17,19], and the remaining one patient had an unmentioned repair[16]. The only transcystic approach was applied in our case. Balloon and saline sweeping was used in one case[17], and we used a basket in a catheter to remove the bile duct stone (Table 2).

Although many studies have reported that transcystic approaches have shorter postoperative hospital stays and fewer bile leaks than choledochotomies[69-72], in most of the cases in our review, the latter was used. Senthilnathan *et al*[17] mentioned that a residual gallbladder containing small calculi was found six months after LC in an SI patient[17]. CBD stones were also exhibited at the same time, which might have migrated from the cystic duct. In this situation, a transcystic approach might be a better option to detect and remove retained cystic duct stones that could be missed by choledochotomy. Our



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Figure 5 Port design in single-incision laparoscopic common bile duct exploration. A: The single-incision multiple-port longitudinal-array technique; B: A commercial multichannel port.

patient is the only case in which a transcystic approach was applied in this review. We showed that SILCBDE with a transcystic approach can be performed successfully in an SI patient. In addition, we used a modified technique named BIC[24]. As choledochoscopy is a delicate device and could be under repair at times, BIC can be performed easily by using a small feeding tube and a stone basket. In our experience, it could achieve stone clearance in nearly half of simple CBD stone cases (data unpublished). With these novel techniques, an SI patient with choledocholithiasis can experience the least invasive LCBDE approach. In our case, SILCBDE *via* a transcystic approach using a modified BIC technique took 152 min to complete, similar to other cases in this review. After a 24.5-mo follow-up, no late complications or stone recurrence were identified.

Limitation

There are several limitations to our study that must be acknowledged. First, due to the rarity of such cases, the number of included cases is very limited. The conclusion might be considered expert opinions, relatively low-quality evidence. Second, cases included in our review were published between 2004 and 2022. Bias could occur because of the advances in techniques and instruments used, which results in consistent cohort heterogeneity. Finally, missing data were encountered in most of the patients, and therefore, some important issues were difficult to address. For example, information on the postoperative length of hospital stay and complications was only available for three (33.3%) and five (55.6%) patients, respectively.

CONCLUSION

Both ERCP and LCBDE are valid options to treat choledocholithiasis by experienced endoscopists and

surgeons. Although there are no related comparative studies, LCBDE seems to be superior to ERCP for SI patients in terms of better intraperitoneal visualization and a lower failure rate. In SI patients, some difficulties of conventional multiport laparoscopic surgical techniques could be overcome by SILCBDE. The transcystic approach and a modified BIC technique are also feasible and safe with many benefits, as we described in our report.

FOOTNOTES

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