Introduction of resection of intrahepatic bile duct stenosis-causing membrane or septum into laparoscopic choledochal cyst excision

Yujiro Tanaka,[a] Takahisa Tainaka,[a] Wataru Sumida,[a] Akinari Hinoki ,[a] Chiyoe Shirota,[a] Naruhiko Murase,[a] Kazuo Oshima,[a] Ryo Shirotsuki,[a] Kosuke Chiba,[a] and Hiroo Uchida [a]

[a]Department of Pediatric Surgery, Nagoya University Graduate School of Medicine, Nagoya 466-8550, Japan

Address correspondence to Yujiro Tanaka

65 Tsurumai-cho, Showa-ku, Nagoya, 466-8550, Japan

Tel.: +81-52-744-2959

Fax: +81-52-744-2980

E-mail: ytanaka-tky@umin.ac.jp

Abstract

Purpose: We previously showed that meticulous probing and resection of the intrahepatic bile duct stenosis (IHBDS)-causing membrane or septum was effective in preventing hepatolithiasis after choledochal cyst excisions in open surgeries. Then, we introduced this maneuver into laparoscopic choledochal cyst excisions in 2014 and performed routine resections since then. The aim of this study was to show the feasibility of this method in laparoscopic surgery.

Methods: We retrospectively reviewed the demographics and outcomes of patients who underwent laparoscopic choledochal cyst excisions at our hospital between January 2014 and December 2017. The patients who underwent surgical treatment for IHBDS-causing membrane or septum were compared with those who did not undergo the procedure. The outcomes of the patients with IHBDS were also compared between patients who were ≥ 3 years of age and those < 3 years at operation.

Results: Seventeen of 35 patients underwent laparoscopic resection of IHBDS-causing membrane or septum. There were no complications related to the procedure although the operative time and intraoperative bleeding amount increased in the patients with IHBDS who were ≥ 3 years of age.

Conclusions: Meticulous probing and excision of the IHBDS-causing membrane or septum is safe and feasible during laparoscopic choledochal cyst excision.

Keywords: laparoscopic surgery, choledochal cyst, bile duct stenosis, ductoplasty

Introduction

We previously showed that meticulous probing and excision of the intrahepatic bile duct stenosis (IHBDS)-causing membrane or septum was effective in preventing hepatolithiasis after choledochal cyst excisions in open surgeries [1]. In our method, IHBDS cases were meticulously scrutinized from the hepatic duct to the segmental bile ducts or more interiorly to detect IHBDS-causing membrane or septum [2]. The cut surface of the membrane or septum was then sutured with 5-0 absorbable material to achieve hemostasis and prevent cicatricial stricture. Although there are some reports concerning laparoscopic ductoplasty for intrahepatic bile duct dilation, only a wide hilar hepaticojejunostomy by cutting the anterior or lateral wall of the hepatic ducts was performed in most cases [3-7]. Then, we introduced our method into laparoscopic choledochal cyst excisions in 2014. The aim of this study was to show the feasibility of this method in laparoscopic surgery.

Patients and Methods

This study was approved by the ethics committee of our hospital and conforms to the provisions of the Declaration of Helsinki. We retrospectively reviewed the demographics and outcomes of the patients who underwent laparoscopic choledochal cyst excision with Roux-en-Y hepaticojejunostomy between January 2014 and December 2017. The data of the patients who had resection of IHBDS-causing membrane or septum associated with choledochal cysts were compared with those of the patients who did

not undergo the procedure. To assess the influence of age at operation, the patients were divided into two groups based on the median age at operation and the outcomes were compared between the older group and the younger group.

IHBDS was defined as a narrowing of the lumen of the intrahepatic bile duct compared with the more peripheral bile duct, as detected by magnetic resonance cholangiopancreatography (MRCP) or preoperative endoscopic retrograde cholangiopancreatography (ERCP). IHBDS cases were meticulously scrutinized from the hepatic duct to the segmental bile ducts or more interiorly to detect IHBDS-causing membrane or septum by cholangioscopy through the umbilical port using rigid endoscope with normal saline irrigation or laparoscopy.

All operations were performed with 5 ports at the umbilicus (inserting 2 ports), right upper and middle flanks, and left upper abdomen by using 8–10 mmHg artificial pneumoperitoneum. The root of the teres hepatis ligament was sutured to the abdominal wall and raised to expose the hepatic hilum. For excision of the choledochal cyst, the common hepatic duct was usually cut at 0.5 cm below the confluence point of the right and left hepatic ducts and 0.5 cm above the joint point to the pancreatic duct. The joint point of the common hepatic duct and the pancreatic duct was confirmed by intraoperative cholangiography. When there was an IHBDS-causing membrane or septum, it was routinely resected. The lateral wall of the hepatic duct was incised to make a wide hilar anastomosis when necessary. Because most stenoses occurred near the hepatic hilum, the IHBDS-causing membrane or septum could be hooked

by the right-angled forceps or a hook-shaped laparoscopic electrocautery and resected through the hepatic side cut-end of the choledochal cyst under laparoscope using scissors, Bipolar Micro Forceps (Aesculap, PA, USA), electrocautery, and Surgitron Surgical RF knife (Ellman, NY, USA) with 4.0-MHz high-frequency technology (Fig. 1a–c). The cut surface of the membrane or septum was then sutured with 5-0 absorbable material as far as possible to achieve hemostasis and prevent cicatricial stricture (Fig. 1d). For the site where suturing was not possible, electrocautery or Surgitron Surgical RF knife was used with care making least damage to the surrounding mucosa (Fig. 1e, f). Then, the proximal jejunum was exteriorized through the umbilical incision to form a Roux loop of 25 cm. The Roux limb was then introduced to the hilum through the retrocolic route, and a Roux-en-Y end-to-side hepaticojejunostomy was established with 5-0 absorbable sutures in a single-layer interrupted manner. The intrahepatic bile ducts were examined regularly by ultrasonography and MRCP at the outpatient's office.

Fisher's exact test and the Mann-Whitney U test were used to compare clinical data. *P*-values < 0.05 were considered statistically significant.

Results

In total, 35 patients underwent laparoscopic choledochal cyst resection during the period. The median age at operation was 2.60 years, and median operative time was 354 min. There was no conversion to open surgery. Seventeen patients had IHBDS-causing membrane or septum and resected

laparoscopically. Among them, IHBDS was relieved in 8 patients after a three-month follow-up, and none had their IHBDS worsen after surgery. Five patients were prenatally diagnosed as having a subhepatic cyst, and all of them had IHBDS.

We compared the demographic data and outcomes between the patients who had resection of IHBDS-causing membrane or septum and those who did not (Table 1). There was no difference concerning age at operation, sex, operative time, intraoperative bleeding amount, the period needed for recovery of bowel movement, and hospital stay after surgery. On the other hand, the demographics showed that cystic type was significantly more frequent in the patients with IHBDS-causing membrane or septum, and fusiform type was more frequent in the patients without IHBDS-causing membrane or septum. As a result, the maximal diameter of the common bile duct was significantly larger in the patients with IHBDS-causing membrane or septum. Concerning complications, minor leakage of the hepaticojejunostomy, ileus, chylous ascites, Petersen hernia, and hepatolithiasis developed. Minor leakage was determined as having bilious discharge from the drainage tube settling near the hepaticojejunostomy which ceased within a week. Petersen hernia developed in a patient with IHBDS six months after surgery and needed reoperation. A small intrahepatic stone was seen in a patient who underwent resection of the IHBDS-causing membrane and septum, and the stone was formed at the more peripheral IHBDS site, which could not be treated during surgery. All the other complications were treated conservatively, and no complications occurred due to resection of IHBDS-causing membrane or septum. No stricture of the

hepaticojejunostomy developed. There was no difference in the complication rate between the patients who had resection of IHBDS-causing membrane or septum and those who did not.

We also investigated the difference in the outcomes with age at operation. Since the median age at operation was 2.60 years, we divided the patients into the older group (≥ 3 years of age at operation) and the younger group (< 3 year of age at operation). The outcomes of the patients with IHBDS-causing membrane or septum were compared between the two groups (Table 2). There was no difference concerning sex, type of choledochal cyst, the period needed for recovery of bowel movement, hospital stay after surgery, and complication rate. However, the operative time was longer and intraoperative bleeding amount was greater in the patients who were ≥ 3 years of age.

On the contrary, there was no difference in the operative time or intraoperative bleeding amount between the patients who were ≥ 3 years of age (n = 7) and those < 3 years (n = 11) at operation in the patients without IHBDS-causing membrane or septum (356 min vs. 332 min in median, $P = 0.75$, and 56 ml vs. 36 ml in median, $P = 0.47$) (Table 3). This implies that laparoscopic resection of the choledochal cysts with IHBDS-causing membrane or septum would become more difficult with age.

Discussion

IHBDS-causing membrane or septum was found in 17/35 (48.6%) patients and compatible to the rate that we found in open surgeries (43.1%, $P = 0.58$) [1]. A 5-mm laparoscope could be used as a

cholangioscope to inspect the intrahepatic ducts. A 3.5-mm cystoscope was also helpful in detecting IHBDS-causing membrane or septum in the segmental bile ducts or more interiorly. A good view of the lumen of the intrahepatic bile ducts could be achieved with saline irrigation. MRCP and virtual cholangioscopy were reported to depict biliary morphology and show IHBDS more accurately than intraoperative cholangiography [8]. This supports our using intraoperative cholangiography only for detecting the joint point of the common hepatic duct and the pancreatic duct and not for detecting IHBDS.

The difficulty in resection of the IHBDS-causing membrane or septum during laparoscopic choledochal cyst excision would come from the limitation of operating range of the laparoscopic instruments due to the site of the ports. In our experience, the right hepatic duct or its branches could be approached from the port at the left upper abdomen, and the left hepatic duct or its branches could be approached from the port at the right middle flank. The cut surface of the membrane or septum could be sutured using the port at the left upper abdomen. Since the lateral wall of the hepatic ducts was cut to make a wide hilar hepaticojejunostomy in most cases with IHBDS, most of the cut surfaces were laparoscopically sutured. Another point of concern was that by which instrument the IHBDS-causing membrane or septum should be resected. For the ones near the confluence of the hepatic ducts, scissors and Bipolar Micro Forceps (Aesculap, PA, USA) were useful. For the ones at the more peripheral site, a hook-shaped laparoscopic electrocautery was useful because the shaft of the instrument was insulated and would not damage the wall of the duct on the route that the electrocautery went through. However,

suturing the cut surface was not possible in the latter cases, and heat injury had to be made minimal. Therefore, we used the Surgitron Surgical RF System (Ellman, NY, USA) with 4.0-MHz high-frequency technology to minimize the thermal damage to the surrounding mucosa in these patients. During our study, there had been no complication due to the resection of the IHBDS-causing membrane or septum. IHBDS was relieved in 8 patients after a three-month follow-up in our study, and none had their IHBDS worsen after surgery. Taking into account that some dilation of the intrahepatic bile duct is reported to resolve within years after choledochal cyst excision, more IHBDS would be relieved with a longer follow-up period [9-11]. Thus, our method of meticulous probing and excision of the IHBDS-causing membrane or septum could be safely and effectively performed during laparoscopic surgery. However, a long-term follow-up is needed to assess the usefulness of this method for preventing hepatolithiasis [1].

In our study, almost all IHBDS-causing membranes or septa were found in the patients with cystic-type choledochal cysts. Notably, all choledochal cysts prenatally detected had IHBDS-causing membrane and were resected before one year of age without anastomotic complications. On the other hand, among the patients with IHBDS-causing membrane or septum, the operative time was longer and intraoperative bleeding amount was more in the patients $\geq$ 3 years of age than those < 3 years. Since the IHBDS-causing membrane or septum is usually thin and has normal mucosa with no histological evidence of inflammatory change, it is thought to be not an acquired one but congenital [12,13]. Therefore, the membranes or septa in the intrahepatic bile duct would make the operation more difficult

with age. Since the maximal diameter did not differ between the patients ≥ 3 years of age than those < 3 years, the difficulty might be due to the adhesions rather than the size of the lesion. Lilly's technique was useful for the cases with adhesions around the choledochal cysts [14,4,7].

With regard to complications, the rate of complications might be higher in our study [3,7]. Half of the complications were minor leakage from the hepaticojejunostomy, and 4 out of six developed in the cases with fusiform-type choledochal cysts. Wide hilar anastomosis was adopted for the cases with narrow hepatic duct, and the lateral wall of the hepatic ducts were cut leaving little margin for anastomosis. As shown in the previous studies, wide hilar anastomosis might have higher morbidity rate [15], and therefore, good technical skill would be needed for this procedure.

Conclusion

Meticulous probing and excision of the IHBDS-causing membrane or septum was safe and feasible during laparoscopic choledochal cyst excision. Although good technical skills would be needed for small infants, laparoscopic choledochal cyst excision should be performed shortly after IHBDS was detected.

Compliance with ethical considerations

Disclosure Statement

No competing financial interests exist.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in the study.

References

1. Tanaka Y, Tainaka T, Sumida W, Shirota C, Hinoki A, Murase N, Oshima K, Shirotsuki R, Chiba K, Uchida H (2017) The efficacy of resection of intrahepatic bile duct stenosis-causing membrane or septum for preventing hepatolithiasis after choledochal cyst excision. J Pediatr Surg 52:1930-1933. doi:10.1016/j.jpedsurg.2017.08.056
2. Ando H, Kaneko K, Ito F, Seo T, Ito T (1997) Operative treatment of congenital stenoses of the intrahepatic bile ducts in patients with choledochal cysts. Am J Surg 173:491-494
3. Li L, Liu SL, Hou WY, Cui L, Liu XL, Jun Z, Liu-Ming H, Gang L, Kamal NA (2008) Laparoscopic correction of biliary duct stenosis in choledochal cyst. J Pediatr Surg 43:644-646. doi:10.1016/j.jpedsurg.2007.10.064
4. Tang ST, Yang Y, Wang Y, Mao YZ, Li SW, Tong QS, Cao GQ, Zhao ZX (2011) Laparoscopic choledochal cyst excision, hepaticojejunostomy, and extracorporeal Roux-en-Y anastomosis: a technical skill and intermediate-term report in 62 cases. Surg Endosc 25:416-422. doi:10.1007/s00464-010-1183-y
5. Wang J, Zhang W, Sun D, Zhang Q, Liu H, Xi D, Li A (2012) Laparoscopic treatment for choledochal cysts with stenosis of the common hepatic duct. J Am Coll Surg 214:e47-52. doi:10.1016/j.jamcollsurg.2012.03.001
6. Diao M, Li L, Cheng W (2013) Role of laparoscopy in treatment of choledochal cysts in children. Pediatr Surg Int 29:317-326. doi:10.1007/s00383-013-3266-z
7. Li S, Wang W, Yu Z, Xu W (2014) Laparoscopically assisted extrahepatic bile duct excision with ductoplasty and a widened hepaticojejunostomy for complicated hepatobiliary dilatation. Pediatr Surg Int 30:593-598. doi:10.1007/s00383-014-3501-2
8. Saito T, Hishiki T, Terui K, Sato Y, Mitsunaga T, Terui E, Nakata M, Takenouchi A, Matsuura G, Yahata E, Ohno S, Sato H, Yanagawa N, Masuda Y, Yoshida H (2011) Use of preoperative, 3-dimensional magnetic resonance cholangiopancreatography in pediatric choledochal cysts. Surgery 149:569-575. doi:10.1016/j.surg.2010.11.004
9. Todani T, Watanabe Y, Urushihara N, Noda T, Morotomi Y (1995) Biliary complications after excisional procedure for choledochal cyst. J Pediatr Surg 30:478-481
10. Stringer MD (2007) Wide hilar hepaticojejunostomy: the optimum method of reconstruction after choledochal cyst excision. Pediatr Surg Int 23:529-532. doi:10.1007/s00383-007-1929-3
11. Ono S, Fumino S, Shimadera S, Iwai N (2010) Long-term outcomes after hepaticojejunostomy for choledochal cyst: a 10- to 27-year follow-up. J Pediatr Surg 45:376-378. doi:10.1016/j.jpedsurg.2009.10.078
12. Ando H, Ito T, Kaneko K, Seo T (1995) Congenital stenosis of the intrahepatic bile duct associated with choledochal cysts. J Am Coll Surg 181:426-430

13. Kemmotsu H, Mouri T, Muraji T (2009) Congenital stenosis of the hepatic duct at the porta hepatis in children with choledochal cyst. J Pediatr Surg 44:512-516. doi:10.1016/j.jpedsurg.2008.06.011
14. Lilly JR (1978) Total excision of choledochal cyst. Surg Gynecol Obstet 146:254-256
15. Lal R, Agarwal S, Shivhare R, Kumar A, Sikora SS, Saxena R, Kapoor VK (2005) Type IV-A choledochal cysts: a challenge. J Hepatobiliary Pancreat Surg 12:129-134. doi:10.1007/s00534-004-0960-1

Figure legends

Fig. 1(a–d) Intraoperative pictures (before (a, b) and after (c, d) surgical treatment) of a patient with membranous stenosis in the right hepatic duct. The arrowhead in (a, b) points to the membrane. In (b), the membrane was hooked by a hook-shaped electrocautery. The cut surface was laparoscopically sutured in (d). The arrowhead in (d) points to the suture site.

(e, f) Intraoperative pictures (before (e) and after (f) surgical treatment) of a patient with membranous stenosis deep in the left hepatic duct. The arrowhead in (e) points to the membrane. The Surgitron Surgical RF System with 4.0-MHz high-frequency technology was used to cut the membrane. The cut surface could not be sutured.

Fig. 1

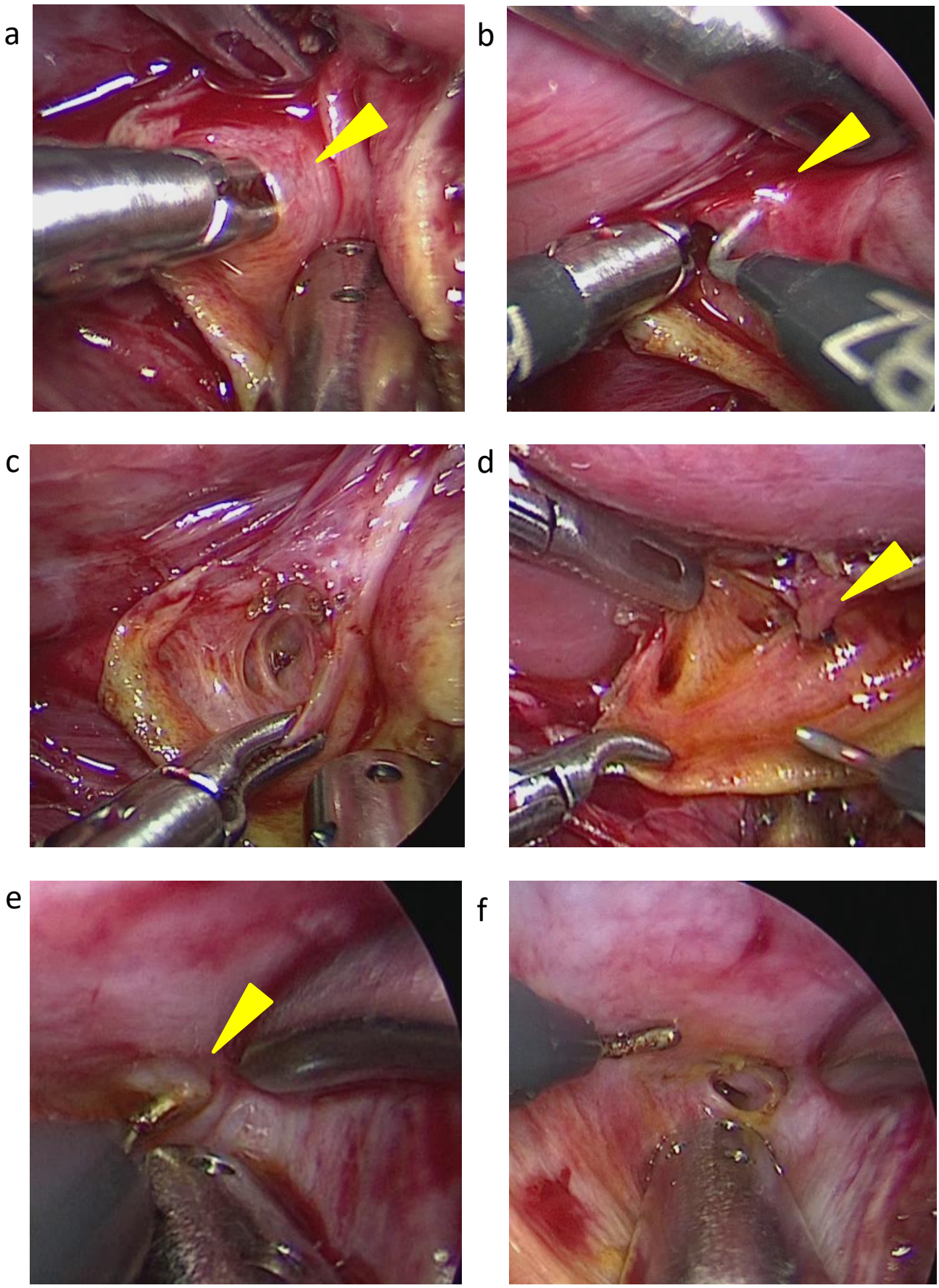

Table 1. Demographic data and outcomes of the patients with and without intrahepatic bile duct stenosis (IHBDS)-causing membrane or septum

	With IHBDS-causing membrane or septum (n = 17)	Without IHBDS-causing membrane or septum (n = 18)	*P*-value
Age at operation (years)	Median, 2.08 (range, 0.07–13.75)	Median, 2.68 (range, 1.14–11.87)	0.38
Male/female	3/14	4/14	1
Maximal diameter of the common bile duct (mm)	Median, 34 (range, 5–90)	Median, 14.5 (range, 4–47)	0.032
Cystic type/ fusiform type	16/1	10/8	0.018
Operative time (min)	Median, 363 (range, 274–557)	Median, 343 (range, 234–852)	0.29
Intraoperative bleeding amount (mL)	Median, 44 (range, 3–118)	Median, 36.5 (range, 2–1216)	0.97
Fasting period after surgery (days)	Median, 3 (range, 2–4)	Median, 3 (range, 2–7)	0.72
Hospital stay after surgery (days)	Median, 10 (range, 7–26)	Median, 10 (range, 7–15)	0.48
Complications	5/17 2 minor leakage 1 Petersen hernia 1 chylous ascites 1 hepatolithiasis	7/18 4 minor leakage 2 ileus 1 wound infection	0.72

The *P*-value was calculated using the Mann-Whitney *U* test and Fisher's exact test.

Table 2. Demographic data and outcomes of the patients with intrahepatic bile duct stenosis (IHBDS)-causing membrane or septum who were ≥ 3 years and < 3 years of age

	< 3 years (n = 10)	≥ 3 years (n = 7)	*P*-value
Age at operation (years)	Median, 0.5 (range, 0.07–2.7)	Median, 6 (range, 3.79–13.75)	
Male/female	3/7	0/7	0.23
Maximal diameter of the common bile duct (mm)	Median, 26.5 (range, 5–47)	Median, 34 (range, 12–90)	0.49
Cystic type/ fusiform type	9/1	7/0	1
Laterality of IHBDS			
Right hepatic duct or its branch	30 % (3/10)	0 %	0.23
Left hepatic duct or its branch	20 % (2/10)	57.1 % (4/7)	0.10
Both	50 % (5/10)	42.9 % (3/7)	0.65
Operative time (min)	Median, 335.5 (range, 274–432)	Median, 418 (range, 319–557)	0.045
Intraoperative bleeding amount (mL)	Median, 21 (range, 3–46)	Median, 82 (range, 42–118)	0.0039
Fasting period after surgery (days)	Median, 3 (range, 3–4)	Median, 3 (range, 2–4)	0.11
Hospital stay after surgery (days)	Median, 10 (range, 7–18)	Median, 9 (range, 7–17)	0.62
Complications	2/10	3/7	0.59

The *P*-value was calculated using the Mann-Whitney *U* test and Fisher's exact test.

Table 3. Demographic data and outcomes of the patients without intrahepatic bile duct stenosis (IHBDS)-causing membrane or septum who were ≥ 3 years and < 3 years of age

	< 3 years (n = 11)	≥ 3 years (n = 7)	*P*-value
Age at operation (years)	Median, 2.16 (range, 1.14–2.97)	Median, 7.84 (range, 3.26–11.87)	
Male/female	1/10	3/4	0.25
Maximal diameter of the common bile duct (mm)	Median, 21.0 (range, 5–47)	Median, 7.0 (range, 4–38)	0.19
Cystic type/ fusiform type	8/3	2/5	0.14
Operative time (min)	Median, 332 (range, 234–424)	Median, 356 (range, 290–852)	0.75
Intraoperative bleeding amount (mL)	Median, 36 (range, 5–112)	Median, 56 (range, 2–1216)	0.44
Fasting period after surgery (days)	Median, 3 (range, 2–7)	Median, 3 (range, 3–4)	0.48
Hospital stay after surgery (days)	Median, 8 (range, 7–15)	Median, 10 (range, 8–13)	0.29
Complications	4/11	3/7	1

The *P*-value was calculated using the Mann-Whitney *U* test and Fisher's exact test.