Early experience of laparoscopic choledochal cyst excision in children

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Purpose: Laparoscopic choledochal cyst excision with Roux-en-Y hepatico-jejunostomy (LCE) in children is being attempted more frequently around the world, and although it has been performed in Korea, no publication has been published on it. However, cholangitis and/or pancreatitis are limitations that make open conversion more likely. The aims of this study, through a retrospective clinical analysis, were to prove the efficacy of LCE in children and to validate that preoperative management expands its indications.

Methods: From May 2011 to November 2012, 13 pediatric LCEs were performed. Demographics, preoperative findings, management, operative and postoperative outcomes were reviewed.

Results: The mean age at operation was 48.5 months and mean bodyweight 19.0 kg. Ultrasonography was conducted in all patients followed by either magnetic resonance cholangiopancreatography (8 cases) or computed tomography (5 cases). The mean diameter of the cysts was 30.2 mm. Eight patients with cholangitis and/or pancreatitis were given antibiotics preoperatively. Four had their condition resolved by administration of antibiotics, 3 underwent additional endoscopic retrograde biliary drainage or percutaneous transhepatic biliary drainage, and one, due to aggravating tenderness, underwent surgery after 4 days of administrating antibiotics without improvement of the inflammation. Two faced open conversions, one because of a very narrow bile duct, and the other because of remnant inflammation after inadequate preoperative management already mentioned above. Patients were discharged on the eighth postoperative day. There were no complications.

Conclusion: Pediatric LCE is a feasible option for choledochal cyst. Proper preoperative management such as antibiotics and drainage procedures enhances its efficacy by broadening its indications, even with concomitant cholangitis and/or pancreatitis.

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INTRODUCTION

Choledochal cyst, a condition in which dilatations occur throughout the biliary tree, was first described by Vater [1] in 1723 and later classified by Todani et al. [2] in 1977. It is more common in Asians, with an incidence range of 1:10,000 in Western countries to as high as 1:1,000 in Japan, and about 3.5 times more likely to occur in women than in men [3]. Choledochal cysts are usually diagnosed in childhood and about 25% are detected during adult life [4]. Choledochal cyst has symptoms such as abdominal pain, jaundice and cholangitis and may eventually lead to malignant transformations. Thus, prompt surgical intervention is imperative.

The surgical treatment of choice has evolved starting from external drainage to complete excision and Roux-en-Y hepaticojejunostomy in open surgery, which had been the standard procedure for many years [5,6].

Since its first report by Farello et al. in 1995 [7], laparoscopic choledochal cyst excision with Roux-en-Y hepaticojejunostomy (LCE) in children has gained worldwide attention. Not only is LCE safe and effective, but it also has several advantages over its open counterpart including excellent visualization, shorter recovery time, prevention of adhesion, less pain and obviation of long subcostal incisions [8].

Despite of its worldwide dissemination, pediatric LCE is scarcely reported in Korea [9]. Furthermore, not all choledochal cyst children are eligible for LCE, largely because cholangitis and pancreatitis are considered to be its contraindications [10]. Through clinical analysis of our institution's early experience with pediatric LCE, this study seeks to demonstrate the feasibility of the procedure and the efficacy of preoperative management in reducing open conversion and in expanding the indications of LCE even in cases with severe cholangitis and/or pancreatitis.

METHODS

In our institution from May 2011 to November 2012, 13 patients received LCE from a single surgeon from which 2 patients faced open conversion.

All data were obtained retrospectively by reviewing the electronic medical records. The age at the time of the operation, body weight, chief complaint, diagnostic modality, Todani's classification, presence of anomalous pancreatico-biliary duct union (APBDU), cyst diameter, presence of gallstones and sludges, preoperative laboratory findings, and preoperative managements, operation time, estimated blood loss during operation by measuring the blood pooled in the suction bottle, starting day of oral diet, hospital stay and postoperative complications were reviewed.

Cholangitis was defined as any symptom of the Charcot triad (abdominal pain, jaundice, fever/chill) combined with an increase in preoperative total bilirubin, aspartate aminotransferase (AST), and alanine aminotransferase (ALT) levels above the normal range and pancreatitis was defined as a preoperative serum amylase or lipase level of more than threefold the normal upper limit. The normal range of the various laboratory results at our institution is as follows: total bilirubin (0.2–1.2 mg/dL), alkaline phosphatase (ALP, 60–300 IU/L), aspartate aminotransferase/alanine aminotransferase (AST/ALT, 0–40 IU/L), amylase (28–100 IU/L), and lipase (22–51 IU/L).

This study was conducted with the approval of the Institutional Review Board of our institution (IRB No: H–1208–052–421).

Operative procedure

The four-port technique was used. Three 5 mm ports were inserted through the umbilicus, right upper quadrant and left side of the umbilicus along the nipple line and a 12 mm camera port was inserted on the right side of the umbilicus along the nipple line. The patients were placed in the supine position. The peritoneum was inflated with CO_2 gas to maintain a pressure of 10–12 mmHg. The choledochal cyst was completely dissected using Harmonic scalpels (Johnson & Johnson, New Brunswick, NJ, USA) and Bovie coagulators. The cyst was excised down to the intrapancreatic portion and double clipped with Hem-o-lock clips (Teleflex Inc., Limerick, PA, USA). The upper part of the cyst was then transected just below the tributary of the right and left hepatic ducts. The hepaticojejunostomy was performed intracorporeally with Vicryl 5–0 (Johnson & Johnson) interrupted sutures, and the jejunojejunostomy was extracorporeally sutured by hand sewing, after which it was put in the peritoneum and checked for torsion of the bowel. The mesenteric defects were fixed and surgical drainage was inserted.

RESULTS

Among the 13 cases in which LCE was attempted, 4 were boys and 9 were girls (Table 1). The mean age of the children was 48.5 months (range, 3 months to 12 years) at the time of the operation. No one had any congenital anomalies or underlying medical conditions. The mean body weight was 19.0 kg (range, 6.9 to 66 kg).

Of the 13, ten patients presented with at least one of the three Charcot triad. Ten patients presented with abdominal pain, 6 with vomiting, 3 with fever, 2 with acholic stool, and 1 with jaundice.

The mean laboratory results for the patients at the time of admission were as follows: total bilirubin, 2.05 mg/dL (range, 0.3 to 8.7 mg/dL); alkaline phosphatase, 312 IU/L (range, 108 to 649 IU/L); AST, 97.15 IU/L (range, 23 to 392 IU/L); ALT, 154.85 IU/L (range, 4 to 899 IU/L); amylase, 331.27 IU/L (range, 11 to 837 IU/L); and lipase, 863.98 IU/L (range, 12 to 1,963 IU/L).

All 13 patients were screened with ultrasonography (US), and magnetic resonance cholangiopancreatography images were taken to further evaluate Todani classification, the presence and type of APBDU, cyst diameter, and the presence of gallstones in 8 patients. The other 5 patients had computed tomography (CT) images from other hospitals. There were 8 type IVa and 5 type Ic choledochal cysts according to Todani

Table 1. Summary of patients

No	Gender	Age at operation	Body weight (kg)	Todani classfication	APBDU	Cyst diameter (cm)	Cholangitis	Pancreatitis	Preoperative antibiotics	Preoperative drainage	Operative time (min)
1	F	23 mo	13.3	IVa	O	3.5	O	O	O	X	190
2	F	4 yr	15.4	Ic	O	1.2	X	X	X	X	275
3	F	18 mo	10.8	IVa	Indeterminable	1.0	X	O	X	X	290[a]
4	F	6 yr	26.2	Ic	Indeterminable	0.7	X	X	X	X	295
5	M	3 mo	6.9	IVa	Indeterminable	7.0	X	X	X	X	200
6	F	4 yr	15.05	Ic	O	2.5	X	X	X	X	270
7	M	9 yr	28.5	Ic	O	1.08	O	X	O	X	255
8	M	12 yr	66.0	IVa	Indeterminable	3.3	O	O	O	X	120[a]
9	F	5 yr	16.9	IVa	O	1.4	O	O	O	ERBD	235
10	F	33 mo	13.5	IVa	O	6.1	O	O	O	PTBD	230
11	F	26 mo	10.2	IVa	Indeterminable	7.2	O	O	O	PTBD	232
12	M	26 mo	13.1	Ic	Indeterminable	1.3	O	O	O	X	225
13	F	21 mo	11.1	IVa	O	3.1	O	O	O	X	200

APBDU, anomalous pancreatico-biliary duct union; ERBD, endoscopic retrograde biliary drainage; PTBD, percutaneous transhepatic biliary drainage.
[a]Open conversion.

classification. APBDU was found in 7 patients. The other 6 cases showed indeterminate or equivocal findings. Gallstones or sludges were found in 3 patients. The mean diameter of the cysts was 30.2 mm (range, 7 to 72 mm).

Nine patients out of the 13 had cholangitis and/or pancreatitis. Seven of them had cholangitis and pancreatitis concomitantly, while 1 patient had cholangitis and another had pancreatitis separately. Among them, only 1 patient with mild pancreatitis was resolved of her symptoms by a nil per os (NPO) period of 6 days without further intervention. Among the other 8 patients, 1 patient received surgery despite insufficient antibiotic administration because physical examination and preoperative laboratory results worsened. He faced open conversion. Four sufficed with a mean antibiotic treatment period of 3.6 days (range, 1 to 5 days) and 3 required an additional drainage procedure despite antibiotic administration. Of the 3 who received an additional drainage procedure, two of them had a percutaneous transhepatic biliary drainage (PTBD) inserted for 19 and 17 days, respectively, before the operation and the other one had an endoscopic retrograde biliary drainage (ERBD) inserted for 7 days preoperatively from another hospital before being admitted to our hospital.

Seven out of 8 patients who received antibiotics with or without a drainage procedure saw improvement in their symptoms and blood tests before receiving surgery. Their mean immediate preoperative laboratory results are: total bilirubin, 0.61 mg/dL (range, 0.4 to 1.0 mg/dL); alkaline phosphatase, 359.2 IU/L (range, 158 to 528 IU/L); AST, 68.3 IU/L (range, 31 to 142 IU/L); ALT, 121.8 IU/L (range, 10 to 313 IU/L); amylase, 107.75 IU/L (range, 61 to 194 IU/L); and lipase, 65.7 IU/L (range, 22 to 147 IU/L). There were no open conversion cases.

The mean estimated blood loss was 103 mL (range, 20 to 200 mL; the value for one patient could not be found) and the mean operation time was 237 minutes (range, 190 to 295 minutes) excluding open conversion. None of the patients received transfusions during or after the surgery. It took a mean period of 5 days (range, 5 to 6 days) to start oral feeding and 8 days (range, 6 to 11 days) to be discharged. There were no intraoperative and postoperative complications.

Of the 2 patients who underwent open conversion, the first case, who was the third patient in the study and 18 months old, had a bile duct diameter of about 5 mm at the site of anastomosis, a size too small to maneuver laparoscopically. The other open conversion case, who was already mentioned before, was treated with preoperative antibiotics only for 4 days due to aggravating symptoms and physical examination, before seeing improvement in the blood tests. Operative findings revealed severe remnant inflammation and a hepatic duct too fragile to anastomose laparoscopically which rendered open conversion necessary.

The mean follow-up period of the patients was 12.5 months (range, 5 to 23 months).

DISCUSSION

Pediatric LCE is gaining pronounced popularity around the world and has been acknowledged as an effective alternative treatment to its open counterpart in several large scale studies. Liem et al. [11], in their large scale study on pediatric LCE, showed that the procedure is feasible, with only 2 cases of open conversions. The same authors have also shown that laparoscopic surgery has similar results compared to open surgery for pediatric choledochal cysts regarding early postoperative complications, operative time, rate of reintervention and duration of postoperative stay [12]. Several authors also maintain in their studies on the comparison of pediatric LCE and open choledochal cyst excision cases that laparoscopic results are comparable or even superior to those of open surgery in intra and postoperative findings [13]. However, there are few reports on pediatric LCE in Korea [9]. Despite the small number of cases, the significance of our study stands out amidst the paucity of reports on pediatric LCE in Korean children

In other publications on the early experience of pediatric LCE, the operation times were as follows: 260 minutes (range, 210 to 450 minutes) [14], 310 minutes (range, 240 to 450 minutes) [15], and 439 minutes (range, 240 to 770 minutes) [16]; there were 35, 31, and 37 cases in their studies, respectively. Another study with a larger number of patients had a shorter mean operation time: 186 minutes (range, 90 to 340 minutes; 74 cases) [17]. The study with a larger number of cases showed a shorter mean operation time which shows that experience improves outcome. In our case, the mean operation time was 237 minutes (range, 190 to 295 minutes) excluding open conversion. The mean operation time of the former 5 cases and the latter 6 cases were 246 minutes and 229 minutes, respectively. Although such a decrease in the operation time did not have statistical significance according to the Kruskal–Wallis test (P = 0.446), the general trend was that the operation time decreased with increasing number of cases. The result of this study supports the observation made above from the other reports that more cases help overcome the learning curve.

Cholangitis and pancreatitis are known as contributing factors for open conversion by making tissues more fragile and inducing adhesion [10]. However, there have been reports that preoperative drainage serves as effective management leading to successful operations for choledochal cysts [18-20]. Kang et al. [19] reported a case of PTBD insertion of choledochal cyst in a patient with panperitonitis. They considered the intervention as a "bridge procedure" which alleviated the pressure on the impending choledochal cyst perforation so that open surgical treatment could be done electively 7 days later. Houben et al. [20] published an article on preoperative ERBD

in choledochal cyst children with a median interval of 10 days before performing open surgery in 3 patients and laparoscopic surgery in 4 patients without open conversion. We also used a drainage procedure as preoperative management to expand the indications of laparoscopic surgery by lowering the risk of open conversion in cases of cholangitis and pancreatitis. There were no open conversions in all the drainage cases which shows that biliary drainage has proven effective in expanding the indications for pediatric LCE. Moreover, the second open conversion case shows that the absence of biliary drainage in severe cholangitis and/or pancreatitis may result in severe inflammation and hence open conversion. According to Tsuchiya et al. [18], ERBD has some advantages over PTBD in children with choledochal cyst in that there is no need for external tube management and no scarring at the insertion site. Unfortunately, in our institution, ERBD was not available, hence PTBD was the only choice left.

The decision to undergo LCE should, however, be made with caution. As in our first open conversion case, small duct size renders laparoscopic maneuver extremely difficult and preoperative drainage procedure may not be effective. Therefore meticulous preoperative evaluation and/or intervention may obviate unnecessary open conversions in such cases by conducting the surgery in open fashion to begin with.

According to other studies on LCE, biliary leakage, sub-hepatic fluid collection, and pancreatic fistula were some of the few encountered complications. Lee et al. [16] reported 6 subhepatic fluid collections (16%), 4 port site infections (11%), 3 bile leakages (8%), and 2 prolonged ileus (5%) after 37 pediatric LCE cases. Liem et al. [11] reported 8 biliary leakages (2%), 4 pancreatic fistulas (1%) and one postoperative cholangitis case. There were no complications in our study. Three plausible explanations are possible. The first one is the prompt decision for open conversion early on in the operation if needed. The second one is the proper preoperative management including drainage procedures to optimize the condition of the patients, and the final one is the small number of cases.

The retrospective nature, the small number of samples, and a short follow-up period are the limitations of this study. Additional studies with further recruitment of cases with a longer observation period will probably produce more concrete results which will support the findings of this paper.

In conclusion, despite the small number of cases and a short follow-up period, this study shows that pediatric LCE could be a safe option for choledochal cyst in children. In addition, proper preoperative management, including antibiotics and biliary drainage procedures, may not only expand the indications but also lower the complication rates of this procedure.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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