Journal of Pediatric Surgery 52 (2017) 1930–1933

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

The efficacy of resection of intrahepatic bile duct stenosis-causing membrane or septum for preventing hepatolithiasis after choledochal cyst excision

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

Department of Pediatric Surgery, Nagoya University Graduate School of Medicine, Nagoya, Japan

ARTICLE INFO

Article history:
Received 16 August 2017
Accepted 28 August 2017

Key words:
Choledochal cyst
Intrahepatic bile duct stenosis
Hepatolithiasis
Late complication

ABSTRACT

Background/purpose: We previously found that many patients who developed hepatolithiasis after choledochal cyst excisions had intrahepatic bile duct stenosis (IHBDS). In 1992, we started resection of the membrane or septum which was found at the site of IHBDS during choledochal cyst excisions. Since intrahepatic stones usually take years to form, the efficacy of this procedure has not been proved.

Methods: The records of patients who had IHBDS-causing membrane or septum and underwent choledochal cyst excision with Roux-Y hepaticojejunostomy between January 1979 and December 2006 were retrospectively analyzed. The patients who underwent surgical treatment for IHBDS-causing membrane or septum were compared with those who did not undergo the procedure.

Results: Sixty-nine patients met the criteria, and seven patients who were followed up for less than 5 years were excluded from the study. Thirty-three patients underwent surgical treatment for IHBDS, and three of them developed intrahepatic stones. Meanwhile, 10 of 29 patients who did not undergo the procedure developed intrahepatic stones. A statistically significant difference in intrahepatic stone formation was observed between the two groups in a log-rank test ($P = 0.016$).

Conclusions: Meticulous probing and excision of the IHBDS-causing membrane or septum are effective for preventing hepatolithiasis after choledochal cyst excisions.

Type of study: Retrospective Comparative Study.

Level of evidence: Level III.

We previously found that many patients who developed hepatolithiasis after choledochal cyst excisions had intrahepatic bile duct stenosis (IHBDS) and noticed that the membrane or septum could be found at the IHBDS sites [1,2]. Then, in 1992, we started routine resection of the IHBDS-causing membrane or septum during choledochal cyst excisions. Intrahepatic stones usually take years to form, and the average period for intrahepatic stone formation was 7 years as determined in our previous work [2]. Thus, the efficacy of resection of the IHBDS-causing membrane or septum could be evaluated only after a long-term follow-up, and there has been no report that certified the efficacy of this procedure.

In this study, we compared the rate of hepatolithiasis after choledochal cyst excision between the patients with and without surgical treatments for IHBDS-causing membrane or septum after an average of 15.4 years of follow-up.

* Corresponding author at: Nagoya University Graduate School of Medicine, Department of Pediatric Surgery, 65 Tsurumai-cho, Showa-ku, Nagoya, 466-8550, Japan. Tel.: +81 52 744 2959; fax: +81 52 744 2980.
E-mail address: ytanaka-tky@umin.ac.jp (Y. Tanaka).

http://dx.doi.org/10.1016/j.jpedsurg.2017.08.056
0022-3468/

1. Materials and methods

This study was approved by the ethics committee of Nagoya University and conforms to the provisions of the Declaration of Helsinki. We retrospectively reviewed the demographics, and the outcomes of patients who had IHBDS-causing membrane or septum associated with choledochal cysts and underwent choledochal cyst excision with Roux-Y hepaticojejunostomy between January 1979 and December 2006. The patients who underwent surgical treatment for IHBDS-causing membrane or septum were compared with those who did not undergo the procedure.

1.1. Detection of IHBDS-causing membrane or septum

IHBDS was defined as a narrowing of the lumen of the intrahepatic bile duct compared with the more peripheral bile duct, or as the blocking of contrast medium in the intrahepatic ducts, as detected by preoperative endoscopic retrograde cholangiopancreatography (ERCP), percutaneous transhepatic cholangiography, or intraoperative

cholangiography. IHBDS cases were meticulously scrutinized from the hepatic duct to the segmental bile ducts, or more interior to detect IHBDS-causing membrane or septum by cholangioscopy and direct vision during surgery [1–5].

1.2. Operative procedure

All operations were performed through the right upper quadrant laparotomy. 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 membrane or septum at the IHBDS site was routinely resected since 1992. Because most stenoses occurred near the hepatic hilum, the stenosis-causing membrane or septum could be hooked by the right-angled forceps or lacrimal duct bougie, and resected through the hepatic side cut-end of the choledochal cyst under direct vision (Fig. 1a–f). The cut surface of the membrane or septum was then sutured with 5-0 absorbable material to achieve hemostasis and to prevent cicatricial stricture. After resecting the stenosis-causing membrane or septum, a fine retrograde transhepatic biliary drainage (RTBD) tube was inserted in the common hepatic duct, passing through the left hepatic duct, and penetrated the liver parenchyma to be brought outside from the anterior abdominal wall. Biliary reconstruction was carried out using a Roux-en-Y end-to-side hepaticojejunostomy with 5-0 absorbable sutures in a single layer interrupted manner [6]. The elimination of the stenosis was confirmed by cholangiography through the RTBD tube. The RTBD tube was removed within 4 weeks after the operation.

1.3. Postoperative follow-up

The intrahepatic bile ducts were examined by ultrasonography at every arrival to the outpatient office, and also by computed tomography when needed. All patients underwent a liver function test and ultrasonography every 3 months for the first postoperative year and every 6 months thereafter. Because most intrahepatic stone formation is detected within 20 years, the maximal follow-up period for each patient was made to be 20 years in this study [7–11].

1.4. Statistical analysis

Mann–Whitney U test, chi-square test, Student's t-test and log-rank test were used to compare data. *P-values* < 0.05 were considered statistically significant.

2. Results

2.1. Demographics

Of the 160 patients (84 patients before 1992, and 76 patients in or after 1992) who underwent operation for choledochal cysts during the study period, sixty-nine patients had IHBDS-causing membrane or

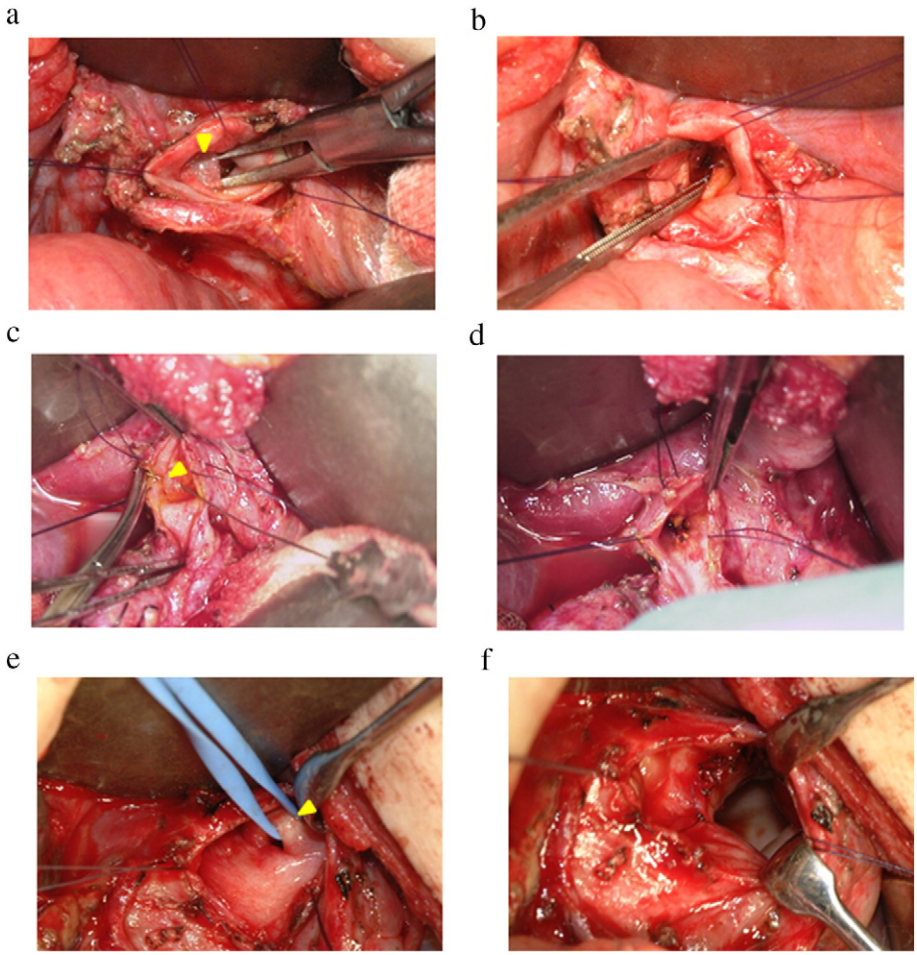

Fig. 1. a, b Intraoperative pictures (before (a) and after (b) surgical treatment) of a patient with membranous stenosis in the right hepatic duct. The arrow head in (a) points to the membrane hooked by a pair of forceps. **c, d** Intraoperative pictures (before (c) and after (d) surgical treatment) of a patient with septal stenosis in the right hepatic duct. The arrow head in (c) points to the septum hooked by a lacrimal duct bougie. **e, f** Intraoperative pictures (before (e) and after (f) surgical treatment) of a patient with septal stenosis in the left hepatic duct. The arrow head in (e) points to the septum pulled up by a silicon tape.