## Journal of Paediatric



# Surgeons of Bangladesh

# **Original Article**

# THE OPTIMAL BILIARY RECONSTRUCTION FOR PANCREATICOBILIARY MALJUNCTION AND CONGENITAL BILIARY DILATATION: CLINICAL COMPARISON BETWEEN HEPATICOJEJUNOSTOMY AND HEPATICODUODENOSTOMY

MITANI Y1, KUBOTA A2

#### **Abstract**

Background: The most important factors determining the optimal biliary reconstruction procedure for pancreaticobiliary maljunction (PBM) and congenital biliary dilatation (CBD) include remote malignant potential and postoperative complications. The aim of this study is to clarify which is more optimal, hepaticojejunostomy (HJ) or hepaticoduodenostomy (HD).

Methods: Seventy-seven pediatric patients with PMD and CBD, treated in our institution between 1985 and 2009, were reviewed in terms of clinical outcomes and postoperative complications. Forty-three underwent HJ and 34 HD, 6 of whom underwent laparoscopy-assisted surgery. Results: Anastomotic stenosis developed in one case of each group, which was treated with percutaneous transhepatic balloon dilation in a case of HJ and endoscopic balloon dilation in a case of HD. Intestinal obstruction related with a Roux-en-Y loop in 4 cases of HJ, 3 of which necessitated laparotomy. Postoperative endoscopy was performed in 14 cases of HD, and demonstrated gastritis in one case of them. No malignancy or cholangitis developed in both groups during the follow-up period.

Conclusions: The possible indirect cause of biliary malignancy, intrahepatic bile-duct stenosis or cholangitis, was not encountered in either procedure, whereas postoperative intestinal obstruction was only in HJ. HD can

Address of corresponding author: Dr. Akio Kubota, Department of Pediatric Surgery, Osaka Medical Center and Research Institute for Maternal and Child Health, 840 Murodo-cho Izumi, Osaka, 594-1101, Japan, Tel: +81-725-56-1220, Fax: +81-725-56-5682; E-mail: kubota@wakayama-med.ac.jp

be more optimal as a biliary reconstruction procedure for PBM and CBD.

Key words: Pancreaticobiliary maljunction, congenital biliary dilatation, Roux-en-Y hepaticojejunostomy, hepaticoduodenostomy, intestinal obstruction, postoperative.

#### Introduction:

The most important factors involved in the determination of the optimal biliary reconstructive procedure for pancreaticobiliary maljunction (PBM) and congenital biliary dilatation (CBD) are remote malignant potential and postoperative complications. Although malignancy associated with PBM develops in relatively younger patients, it develops 1 to 19 years (average 9.0±5.5 years) after the reconstructive surgery [1], and anastomotic stenosis, one of the indirect causes of postoperative malignancy, develops 3 to 21 years after the surgery [2]. Therefore, we have to follow patients for a longer period to confirm the optimal reconstructive procedure in terms of the development of malignancy. In this study, we aimed to clarify which of hepaticojejunostomy (HJ) or hepaticoduodenostomy (HD) is optimal in terms of postoperative complications and the development of anastomotic stenosis and cholangitis, which are the most important indirect causes of biliary malignancy after reconstruction surgery in PBM.

### Materials and methods:

Demographic data was shown in table 1. Seventyseven pediatric patients, 8 boys and 69 girls, with

Yasuyuki Mitani, Akio Kubota, Department of Pediatric Surgery, Osaka Medical Center and Research Institute for Maternal and Child Health, Osaka, Japan

PBM and CBD, were referred to our institution between 1985 and 2009. All of them were treated at our institution by 6 pediatric surgeons, and reviewed in terms of clinical outcomes and postoperative complications. PBM or CBD was diagnosed when a common channel was 5mm or more, or the common bile duct was dilated on ERCP or MRCP, or ultrasonography. As biliary construction type, HJ was performed until 1996, and HD was performed basically after 1997. Forty-three underwent HJ and 34 underwent HD, and in 6 of the 34, HD was performed by laparoscopy-assisted surgery. The age at the time of the operation ranged from 1 month to 14 years, and the follow-up period of HJ group ranged from 3 months to 22 years, with median of 10.6 years, that of HD group ranged from 3 months to 21 years, with median of 4.5 years. All the patients visited the institution to undergo serum studies of biliary enzymes and ultrasonographic studies. Differences between the HJ and HD groups were compared using Mann-Whitney U-test or chi-square test. Statistical significance was set at P<0.01.

#### Results:

The operation time of HJ and HD was 6.3±1.8 hours and 6.4±1.9 hours, respectively. Blood loss in HJ and HD group was 154±95ml and 120±117 ml, respectively. There were no significant differences in the operation time and blood loss between the two groups. The operation time of conventional HD and laparoscopy-assisted HD was 6.1±1.6 and 7.3±2.8 hours, respectively, and blood loss was 140±117ml and 40±58ml, respectively. The blood loss of laparoscopy-assisted HD was lower than that of conventional HD, however the difference is not statistically significant. As an operative complication, injury of the pancreatic duct was encountered in one patient in the HJ group, who required pancreatic ojejunostomy. Early postoperative complications, defined as complications that occur within one year after the surgery, included one case of intraperitoneal abscess formation with HJ, one case of prolonged biliary leakage with HJ, three cases of moderate intestinal obstruction with HJ and one with HD, and one case of anastomotic stenosis in each of the two groups. Abscess and biliary leakage were treated with percutaneous drainage, and the intestinal obstruction recovered with conservative therapy. The anastomotic stenosis was treated with percutaneous transhepatic biliary drainage in case of HJ, and endoscopic balloon dilatation in case of HD (table2). Late postoperative complications, defined as

complications that occur one year or longer after the surgery were four cases of intestinal obstruction with HJ but none with HD, and pancreatic calculus with HJ. Intrahepatic stone developed in one case of each group, which was asymptmatic. The intestinal obstruction required laparotomy in three cases, which revealed that the obstruction was located in the Rouxen-Y loop. The Roux-en-Y loops had been made in antecolic and retrocolic fashion, respectively, in two cases each. Since the Roux-en-Y loop was obstructed, vomiting was not a symptom in any of the cases, but there was severe pain in the right hypochondrium. Abdominal plane x-ray performed in upright position did not show gas accumulation or a mirror image in the Roux-en-Y loop, so it took time to make the diagnosis of intestinal obstruction. CT showed marked intestinal tract dilatation on a subhepatic view (Figure 1), and ultrasonography showed pathognomonic to-and-fro movement of enteral fluid in the dilated intestinal tract. The pancreatic calculus did not present any symptoms or long-term sequels, and did not require treatment. In neither group, did cholangitis, hepatic or pancreatic dysfunction or biliary malignancy develop during the follow-up period (table-III).

**Table-I**Demographic data of the patients

	HJ (n=43)	HD (n=34)			
Median age, years (range)	2.2 (0.1-14)	2.8 (0.1-13.3)			
Sex male: female	4:39	4:30			
Туре	26	13			
Cystic	17	17			
Fusiform	0	4			
Nnn-dilated Follow	10.6 (0.3-22.6)	4.5 (0 3-21)			
up period, years (range)					

**Table-II**Early complication

	HJ	HD	Difference
	(n=43)	(n=34)	
Intraperitoneal abscess	1	0	n.s
Biliary leakage	1	0	n.s
Intestinal obstruction	3	1	n.s
Anastomatic stenosis	1	1	n.s
Total	6	2	n.s

Table-III
Late complication

	HJ	HD	Difference
	(n=43)	(n=34)	
Malignancy	0	0	n.s
Cholangitis	0	0	n.s
Pancreatitis	0	0	n.s
Intarahepatic stone	1	1	n.s
Intestinal obstruction	4	0	n.s
Pancreatic calculus	1	0	n.s
Gastritis	0	1	n.s
Total	6	2	n.s



**Fig.-1:** CT of a case of intestinal obstruction in HJ group, demonstrates a localized marked dilatation of intestinal tract on a subhepatic view. No gas accumulation nor a mirror image is shown. Ultrasonography showed pathognomonic to-and-fro movement of enteral fluid in the dilated intestinal tract.

In the latest 14 cases of HD, gastric endoscopy was performed to look for regurgitation gastritis at 12 to 19 months, with median 15.5 months, after surgery. In 9 cases, macroscopic bile regurgitation into the stomach was noticed, but neither gastritis nor erosion was identified. Histological examination revealed no pathological changes such as dysplasia or chronic gastritis in 13 cases, but in one case, mild lymphocytic infiltration was noticed. However, the patient did not have any symptoms of gastritis.

#### Discussion:

In general the postoperative course of reconstructive surgery for PBM and CBD, which are both benign disease, is good. Therefore, the optimal reconstructive procedure is determined by the development of malignancy and postoperative complications during long-term observation of the course. It has been considered that biliary malignancy that develops in association with PBM or CBD is caused by a mutagen that is produced when pancreatic fluid regurgitates into the bile duct to make a mixture of pancreatic fluid and bile<sup>3</sup>, and it was also experimentally shown that a mixture of pancreatic fluid and bile becomes mutagenic in the presence of enterokinase4. It has also been confirmed that in case of maljunction, extra-hepatic biliary epithelium has malignant potential even in cases without dilatation<sup>5,6</sup> and gallbladder mucosa has one even from pediatric age<sup>7,8</sup>. However, exposure to a mutagen per se rarely causes malignancy, so most cases of malignancy that develop in association with maljunction occur because such digestive fluids become stagnated in the bile duct. It can therefore be considered that the postoperative development of malignancy, like the development of cholangitis or the formation of intrahepatic calculus, is caused by intrahepatic bile duct or anastomotic stenosis. It has been reported that there is no difference in the incidence of postoperative cholangitis after reconstructive surgery between HD and HJ [9]. On the one hand, Watanabe et al1 reviewed the postoperative development of malignancy after reconstructive surgery in 23 cases of PBM and CBD in detail, and reported that malignancy occurred not only after HD but also after HJ, there was no evidence that postoperative malignancy occurred more frequently after HD than HJ, and that in most cases malignancy occurred at bile duct irrelevant to the regurgitation of pancreatic fluid, but at the site of biliary stagnation. They concluded that another mechanism than regurgitation of pancreatic fluid is involved in the development of malignancy of the intrahepatic bile duct. Thus, it is considered that another mechanism than contact with a mixture of bile and pancreatic fluid acts on the bile duct with malignant potential and causes malignancy even after the reconstructive surgery for PBM and CBD.

Serious complications for which laparotomy is indicated may occur after reconstruction surgery, and is an important factor in the determination of the optimal procedure. As mentioned above, a very important complication that can also cause malignancy is anastomotic stenosis. Todani et al.<sup>2</sup> stressed that the most important thing in preventing

anastomotic stenosis is anastomosis at the level of the bifurcation of the left and right hepatic ducts. This seems possible both with HJ and HD. However, in cases of anastomotic stenosis, if the procedure was HD. endoscopic dilatation is possible as shown in the present series<sup>10</sup>. In the present series, only one case of early anastomotic stenosis was found both in the HD and HJ groups, and it has been reported that anastomotic stenosis appears from 3 to 21 years postoperatively<sup>1</sup>, so long-term follow-up is essential. As an intestinal complication after reconstructive surgery, it has been reported that bilious gastritis due to regurgitation of bile shows a high incidence<sup>11</sup>, but in the present series no cases showed symptoms suggestive of gastritis at one year or longer postoperatively. In our latest 14 cases we performed gastric endoscopy at one year or longer after surgery to investigate the development of bilious gastritis histologically, and although regurgitation of bile into the stomach was found in 64% of the subjects, histologically only one showed lymphocytic infiltration into the lamina propria (gastritis).

It has been reported that re-laparotomy after reconstructive surgery for this disease is conducted in 6 to 10% of cases<sup>2,12,13</sup>. In the present series relaparotomy was conducted in 3 of 77 cases, namely, 4%. Almost all of the reported cases involved reanastomosis or dilatation procedures for anastomotic stenosis or intrahepatic bile duct stenosis, or lateral segmentectomy for intrahepatic bile duct calculus. The authors have not yet experienced cases that needed laparotomy for stenosis, but experienced 3 cases which needed laparotomy for intestinal obstruction. It has been reported that re-laparotomy for intestinal obstruction was performed in 3% of subjects in a large HJ series<sup>13</sup>. In the present series too, all cases of obstruction were seen in the HJ group, and since it was obstruction of the Roux-en-Y loop, no subjects showed vomiting, and pain in the right hypochondrium was the only symptom. On Xray too, it was difficult to make a diagnosis of intestinal obstruction, but the CT showed localized dilatation of the intestinal tract on a subhepatic view, and the to-and-fro movement of enteral fluid in the dilated intestinal tract on ultrasonography was pathognomonic (Figure 1). Care should be taken when making the diagnosis because the clinical picture differs from that of ordinary adhesive intestinal obstruction.

Concerning the development of malignancy, the most important factor to be considered in the determination of the optimal reconstructive procedure for this disease, although further long-term observation is necessary, at present there is no difference between HJ and HD. Concerning the other factor, complications for which re-laparotomy is necessary, since HD, which is a simpler procedure, seems to be superior. Fuanbiki [14] preferred HD because it is less invasive procedure and preserves natural bile follow into the intestinal tract. Therefore, the authors concluded that HD is the optimal reconstructive procedure after the surgery for this disease.

**Conflict of interest statement:** All authors have no conflict of interestÿ

#### References

- Watanabe Y, Toki A, Todani T, et al: Bile duct cancer developed after cyst excision for choledochal cyst. J Hepatobiliary Pancreat Surg1999;6:207-212.
- Todani T, Watanabe Y, Urushihara N, et al: Biliary complications after excisional procedure for choledochal cyst. J Pediatr Surg 1995;30:478-481.
- Funabiki T, Matsubara T, Miyasaka S, et al: Pancreaticobiliary maljunction and carcinogenesis to biliary and pancreatic malignancy. Langenbecks Arch Surg 2009; 394:159-169,.
- 4. Mizuno M, Kato T, Koyama K: An analysis of mutagens in the contents of the biliary tract in pancreaticobiliary maljunction. Surgery Today 1996; 26:597-602.
- Matsubara T, Sakurai Y, Zhi LZ, et al: K-ras and p53gene mutations in noncancerous biliary lesions of patients with pancreaticobiliary maljunction. J Hepatobiliary Pancreat Surg 2002; 9:312-321.
- Funabiki T: Pancreaticobiliary Maljunction focused o biliary carcinogenesis-. Jpn J Gastroenterol Surg 2000; 33:261-270.
- 7. Tokiwa K, Iwai N: Early mucosal changes of gallbladder in patients with anomalous arrangement of the pancreaticobiliary duct. Gastroenterology 1996;110:1614-1618.
- 8. Ono S, Tokiwa K, Iwai N: Cellular activity in the gallbladder of children with anomalous

- arrangement of the pancreaticobiliary duct. J Pediatr Surg 1999;34:962-966.
- Todani T, Watanabe Y, Toki A, et al: Biliary complications after excisional procedure for choledochal cyst. Ann Surg 1988;207:142-146.
- Kubota A, Kawahara K, Okuyama H, et al: Clinical outcome after reconstruction for pancreaticobiliary maljunction. Jpn J Pediatr Surg 2004; 36:501-506 (abstract in English).
- 11. Shimotakahara A, Yamataka A, Yanai T, et al: Roux-en-Y hepaticojejunostomy or hepaticoduodenostomy for biliary reconstruction during the surgical treatment of choledochal cyst: which is better? Pediatr Surg Int 2006;21:5-7.
- Ohi R, Yaoita S, Kamiyama T, et al: Surgical treatment of congenital dilatation of the bile duct with special reference to late complications after total excisional operation. J Pediatr Surg 1990;25:613-617.
- Yamataka A, Ohshiro K, Okada Y, et al: Complications after cyst excision with hepaticoenterostomy for choledochal cysts and their surgical management in children versus adults. J Pediatr Surg 1997; 32:1097-1102.
- Funabiki T: Pancreaticobiliary maljunction Focused on biliary carcinogenesis-. Jpn J Gastroenterol Surg 2000; 33:261-270.