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Duodenogastric regurgitation in hepaticoduodenostomy after excision of congenital biliary dilatation (choledochal cyst)

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ABSTRACT

Purpose: We examined the clinical significance of duodenogastric regurgitation (DGR) as a late complication in the long-term follow-up after hepaticoduodenostomy (HD) as a reconstruction surgery for congenital biliary dilatation (CBD).
Methods: Seventeen patients with CBD were retrospectively analyzed for late complications (mean follow-up, 16.8 years). All patients had undergone total resection of the extrahepatic bile duct followed by HD. DGR was identified using endoscopic examination, intraluminal bile monitoring, and liver scanning.
Results: DGR was found in all 17 patients by endoscopic examination and intraluminal bile monitoring. Fourteen of the 17 (82.4%) patients with DGR had experienced abdominal symptoms since a mean of 6.9 years postoperatively. Liver scanning also revealed apparent DGR in all 14 symptomatic patients. We converted 7 of the 14 patients to hepaticojejunostomy reconstruction at a mean of 13.0 years after the initial excisional surgery. Their symptoms were completely relieved postoperatively.
Conclusions: DGR is an important complication after HD. Examination of patients for the development of DGR is an essential part of long-term follow-up in patients with CBD who have undergone HD as a reconstruction surgery. Conversion surgery is recommended in patients with DGR accompanied by long-term abdominal symptoms.
Levels of evidence: Level IV.

The 2015 Diagnostic Criteria for Congenital Biliary Dilatation were recently established by the Japanese Study Group on Pancreaticobiliary Maljunction, representing the first establishment of such guidelines worldwide [1]. In these diagnostic criteria, the medical term "congenital biliary dilatation" (CBD) was adopted instead of the more widely used term "choledochal cyst." This term is used to ensure more correct expression and universally accurate understanding of this entity and therefore better treatment results worldwide.

Primary cyst excision followed by biliary reconstruction is the fundamental treatment for CBD. With respect to the type of biliary reconstruction, hepaticojejunostomy (HJ) has been widely performed in many instances, and many reports have addressed the long-term outcomes [2,3]. Hepaticoduodenostomy (HD) has been performed as an alternative method of reconstruction [4]. HD was recently reported to have better outcomes than HJ [5], and similar results have been reported in laparoscopic excisional surgery [6–8]. However, these reports are concerning with respect to short- and intermediate-term outcomes such as the operative time, recovery of bowel function, and complications requiring reoperation. Few reports on the long-term (>10-year) outcomes after HD have been published [2].

We previously reported our preliminary observations that excessive duodenogastric regurgitation (DGR) is an important postoperative complication [9] that results in enhanced cellular proliferative activity of the gastric mucosa [10] in patients who have undergone HD as a reconstruction surgery for CBD. A meta-analysis [11] showed that patients who underwent HD had a higher incidence of postoperative reflux/gastritis than did patients who underwent HJ. In the present study, we investigated DGR as a late complication among long-term outcomes and assessed the clinical significance of conversion to HJ reconstruction.

1. Patients and methods

From 1977 to 2015, we encountered 83 pediatric patients with CBD in our department. These 83 patients included 25 who underwent HD from 1988 to 2003. The other 58 patients underwent HJ. Todani et al. [4] has advocated HD with a wide stoma at the hepatic hilum (HDH) as the most effective reconstruction method. They reported lower rates of postoperative biliary complications than after HD below the hepatic hilum because this method allows for free drainage of bile and

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prevents anastomotic stricture-induced cholangitis [12]. We have adopted HDH or HD as a reconstruction surgery for CBD to minimize anastomotic stricture formation.

HD was constructed approximately 2 to 3 cm distal to the pyloric ring using interrupted 5-0 absorbable sutures. In patients with Todani type IV-A CBD, HDH [4] was also performed to allow for a large stoma. We reviewed these 25 patients with special respect to the development of DGR as a late complication occurring ≥10 years postoperatively.

DGR was identified by intraluminal bile monitoring using the Bilitec device (Medtronic, Denmark) and endoscopic examination, which has been previously described [9]. Patients with DGR were further examined by liver scanning using ^{99m}Tc-PMT scintigraphy. Magnetic resonance cholangiopancreatography or drip infusion cholangiography with computed tomography was also performed to reveal the whole anatomy of the biliary system, including anastomotic conditions, as a follow-up study [13,14].

2. Results

As shown in Fig. 1, 17 of 25 patients who underwent HD were capable of undergoing DGR testing such as intraluminal bile monitoring and endoscopic examination. The other eight patients did not undergo DGR evaluation because they were lost to follow-up. The mean age at the time of the initial excisional surgery of the 17 patients (14 female, 3 male) was 4.2 years (range, 11 days to 18 years), and the mean follow-up duration was 16.8 years (range, 14–22 years). All patients were confirmed to have DGR at an average of 5.4 years (range, 3–11 years) postoperatively. They were also revealed to have gastritis with a higher proliferative activity of the gastric mucosa as indicated by the proliferating cell nuclear antigen (PCNA) labeling index (data not shown), as previously reported in a preliminary study of 11 patients who underwent HD [10]. Hepatic dysfunction was not observed in any patients with DGR. In contrast, all patients who underwent HJ, including five patients in our previous report [9], showed no evidence of DGR (data not shown).

Liver scans also revealed apparent DGR in 16 of the 17 patients. In the 14 patients with abdominal symptoms, a strong to moderate hot spot was observed in the stomach 30 to 120 min after injection of the radionuclear tracer (Fig. 2). In contrast, among the remaining three patients without abdominal symptoms, a weak hot spot of short duration was observed in two and no hot spot was observed in one. Abdominal symptoms tended to be correlated with both the duration of the time and strength of the hot spot in the stomach.

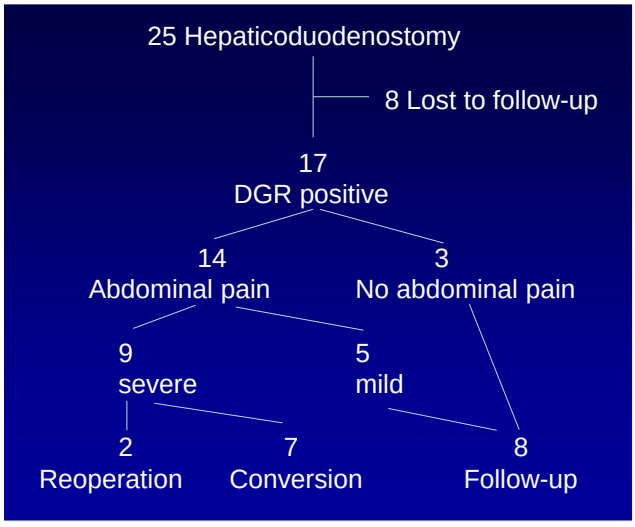

Fig. 1. Decision tree of 25 patients with hepaticoduodenostomy.

Abdominal symptoms such as epigastric pain, heartburn, and abdominal discomfort were present in 14 (82.4%) of the 17 patients with DGR. Three patients showed no abdominal pain but were nevertheless shown to have DGR. Their mean follow-up period was 17.0 years, and their symptoms had appeared at a mean of 6.9 years (range, 5–11 years) postoperatively. These 14 patients included 5 patients with mild abdominal pain that did not require medication and 9 patients with severe abdominal pain undergoing treatment with medications such as camostat mesilate, famotidine, and teprenone. Five patients with mild abdominal pain were regularly followed up without medication in the outpatient clinic.

Two of the nine patients with severe abdominal pain had previously undergone reoperations for an anastomotic stricture 5 years after HD (1 patient) and congenital intrahepatic bile duct stricture in segment IV 6 years after HD (1 patient). Resection of the HD anastomosis followed by conversion to HJ reconstruction was simultaneously performed. Their cholangitis- and DGR-related symptoms were relieved soon after surgery.

We recommended conversion to HJ reconstruction surgery for the remaining seven patients with severe abdominal pain (Table 1). Conversion surgery was performed by resection of the HD or HDH anastomosis followed by either HJ or HJH (HJ at the hepatic hilum). Four of the seven patients had Todani type IV-A CBD at the time of the initial surgery. The patients' age at the initial surgery ranged from 11 days to 18 years. HD was performed in four patients, including two with Todani type IV-A CBD. HDH was performed in three patients, including two with concomitant malrotation. Conversion surgery was performed at an average age of 15.4 years (range, 9–28 years), and the mean interval between the initial surgery and conversion surgery was 12.6 years (range, 8–16 years). The patients' abdominal symptoms disappeared soon after the conversion surgery, and the average 3.7-year follow-up (range, 3 months to 6 years) after the conversion surgery showed a good clinical course with no gastritis by postoperative follow-up endoscopic examination.

With respect to the relationship between abdominal symptoms and Todani's classification, 8 of 17 patients with test-proven DGR had Todani type IV-A CBD, and these 8 patients included 4 of the 7 patients with severe abdominal pain who underwent conversion surgery. In contrast, all three patients with no abdominal pain and four of five patients in whom conservative treatment could be achieved had Todani type Ia or Ic CBD. In addition, two of four patients who underwent HDH (high level) showed DGR with severe abdominal pain. The 13 patients who underwent HD (low level) included 7 patients with severe abdominal pain, 3 patients with mild abdominal pain, and 3 patients with no pain.

3. Discussion

We previously reported that excessive DGR occurs several years after HD as confirmed by intraluminal bile monitoring and endoscopic examination [9]. DGR-related changes were further confirmed by the enhanced cellular proliferative activity of the gastric mucosa as shown by the PCNA index [10]. DGR may be implicated in gastric and esophageal carcinogenesis, but little is currently known about how DGR is associated with gastric carcinogenesis [15]. This prompted us to examine the long-term outcomes of patients with CBD who had undergone HD, with special attention to the development of DGR.

The presence of DGR is difficult to determine only by the patient's complaints; specific examination techniques are needed to diagnose DGR. We used both endoscopy and a special Bilitec device to reveal bile within the gastric contents as the initial examination. Liver scans are also useful for detection of gastric regurgitation of bile along with the time course of bile flow and can be performed on an outpatient basis without sedation or general anesthesia. DGR was observed in all 17 patients who had undergone HD in whom endoscopy and intraluminal bile monitoring were performed at a mean of 5.4 years