

The efficacy of double-balloon enteroscopy for intrahepatic bile duct stones after Roux-en-Y hepaticojejunostomy for choledochal cysts

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Abstract

Introduction Intrahepatic bile duct (IHBD) stones are one of the most complicated morbidities that occur after Roux-en-Y hepaticojejunostomy (RYH); however, the optimal therapeutic approach is controversial.

Methods Double-balloon enteroscopy (DBE) has been widely and frequently performed even in pediatric patients. We herein report the successful management of IHBD stones by biliary lithotripsy using DBE after RYH for a choledochal cyst (CC). DBE has made it possible to perform endoscopic therapeutic intervention, including balloon dilatation of an anastomotic stricture and removal of IHBD stones, without any major complications.

Conclusion DBE is a less invasive and safe treatment method for IHBD stones in pediatric patients, which is capable of reaching the bilioenteric anastomosis after RYH for CC.

Keywords Double-balloon enteroscopy ·
Intrahepatic bile duct stone · Choledochal cyst

Introduction

Choledochal cysts (CC) usually have a good prognosis when they are diagnosed early and when patients undergo

early surgical treatment, which typically consists of total excision of the extrahepatic bile duct and reconstruction using Roux-en-Y hepaticojejunostomy (RYH) [1]. However, we sometimes encounter some late complications including intrahepatic bile duct (IHBD) stones with persisted cholangitis. Moreover, IHBD stones and repeated cholangitis may lead to carcinogenesis of the biliary tract even after early diversion surgery in children [2]. Therefore, long-term surveillance remains essential in pediatric patients, especially if there is persistent dilatation of the intrahepatic bile duct; however, the optimal therapeutic approach for IHBD stones is controversial in pediatric patients.

Double-balloon enteroscopy (DBE) is a novel diagnostic and therapeutic modality reported by Yamamoto et al. [3]. DBE allows not only for a diagnosis to be made, but also for endoscopic therapeutic interventions, including hemostasis, polypectomy, stenting and balloon dilatation of the small intestine. DBE has also made it possible to approach the bilioenteric anastomosis via the leg of the Roux-en-Y reconstruction [4, 5].

We herein report the successful management of IHBD stones by biliary lithotripsy using DBE after hepaticojejunostomy for CC in pediatric patients.

Patients and methods

DBE was performed under general anesthesia in two pediatric patients with IHBD stones, which had developed 10 and 5 years, respectively, after RYH for CC.

All study participants provided informed consent, and the study design was approved by the appropriate ethics review boards.

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DBE system

DBE was performed using a Double-Balloon Endoscopy system (Fujinon, Saitama, Japan), which consisted of a double-balloon endoscope (EI-530B; Fujifilm, Tokyo, Japan), a flexible overtube with a balloon and a balloon controller to inflate or deflate the balloons. The basic structure of the endoscope for DBE is the same as that of ordinary endoscopes except that the DBE scope has an air channel for the balloon. Both the endoscope and overtube balloons are made from 0.1-mm thick latex, which is very soft and can be inflated or deflated by a dedicated balloon controller (PB-20; Fujinon, Saitama, Japan) with one-touch controls, while monitoring the air pressure. The balloons are inflated up to the size of the lumen of the intestine to grip effectively and safely regardless of its size [6].

Principle of DBE

Double balloon endoscopy uses a flexible overtube with a balloon at the tip to prevent stretching. The inflated balloon at the tip of the overtube holds the intestine and prevents the tip of the overtube from slipping. The overtube can thus bend flexibly but not stretch. The overtube does not stretch even in the presence of bends or loops, does the intestinal tract that is held by the balloon at the tip of the overtube. Consequently, insertion of the endoscope shaft does not stretch the intestine and force is effectively transmitted to the endoscope tip. The endoscope can thus be advanced into the distal portion with the balloon at the tip of the overtube as a fixed support. After the endoscope is advanced as deeply as possible, the balloon at the tip of the overtube is deflated, and the overtube is slid onto the endoscope. During this procedure, the balloon at the endoscope tip is inflated to grip the intestine from inside, preventing the endoscope tip from slipping. After sliding the overtube to its new distal position, the overtube balloon is inflated, and both the endoscope and overtube are withdrawn with the two balloons inflated. After these procedures, the intestinal tract is pleated over the overtube and shortened. The endoscope is inserted into the distal small intestine while the fixed support established by the balloon at the tip of the overtube is moved step by step [6].

Results

DBE has made it possible to perform endoscopic therapeutic intervention, including balloon dilatation of bilioenteric anastomotic stricture and removal of IHBD stones even in pediatric patients, without any major complications.

Case 1

An 11-year-old male was referred to our hospital for abdominal pain and vomiting due to IHBD stones. He had undergone a hepaticojejunostomy with resection of CC when he was one year old. The length of the leg of the Roux-en-Y reconstruction was 40 cm. His postoperative course had been uneventful, however, he developed abdominal pain and vomiting when he was 11 years old. He was revealed to have cystic dilatation of IHBD and IHBD stones by a CT scan (Fig. 1). He was treated with a course of antibiotics for repeated cholangitis. After the treatment of cholangitis, DBE was performed under general anesthesia. Endoscopic retrograde cholangiography (ERC) was performed through DBE, and showed a dilatation of intrahepatic bile duct and the filling defect of suspicious stones (Fig. 2). DBE showed a severe stricture of the bilioenteric anastomosis (Fig. 3a). DBE was also used to perform balloon dilatation of the anastomotic stricture (Fig. 3b) and biliary lithotripsy using a crusher catheter under direct observation (Fig. 3c). The total length of the operation was 155 min. The patient's postoperative course was uneventful except for transient hyperamylasemia. He was free from the persistent abdominal pain after the DBE intervention.

Case 2

A 9-year-old male was suffering from repeated abdominal pain and jaundice. He had undergone RYH with resection of CC diagnosed as Todani IV-A type when he was 4 years old. The length of the leg of the Roux-en-Y reconstruction was 50 cm. After the operation he had needed to be admitted to the hospital to receive treatment for abdominal pain with jaundice about once a year. On further



Fig. 1 Abdominal CT scan revealed cystic dilatation of intrahepatic bile duct and multiple intrahepatic bile duct stones in Case 1



Fig. 2 Endoscopic retrograde cholangiography showed dilatation of intrahepatic bile duct and filling defect of suspicious stones in Case 1

examination, including a CT scan and MRI, his clinical condition seemed to be attributable to IHBD stones. Therefore, he was admitted to our hospital for DBE. DBE was performed under general anesthesia and permitted us to reach the bilioenteric anastomosis via the leg of the Roux-en-Y reconstruction. DBE was removed leaving the overtube in place. Then ultraslim endoscope (EG-530NW; Fujifilm, Tokyo, Japan) with 5.9 mm outer diameter and 2.0 mm working channel was inserted smoothly through the overtube. Direct cholangioscopy to reach anterior segmental branch revealed multiple stones (Fig. 4). Cholangioscopy with DBE also allowed us to crush and remove the IHBD stones using a basket catheter (MTW, Wesel, Germany) through the overtube. The total length of the operation was 157 min. There were no intraoperative or postoperative complications. The patient’s postoperative course was good, with relief of his symptoms, including the abdominal pain and jaundice. Figures 5 and 6 show the schemas of the procedure using DBE.

Discussion

IHBD stones and postoperative cholangitis were reported as one of the few late complications after Roux-en-Y hepaticojejunostomy for choledochal cyst (CC). Postoperative bile stasis and infection of the biliary tract are speculated to cause the formation of IHBD stones. On the other hand, congenital stenosis of the intrahepatic bile duct is occasionally present, which might require special attention at the initial operation. In any case, the optimal therapeutic procedure to treat postoperative stricture of the IHBD and/or IHBD stones is controversial, but essential, especially in

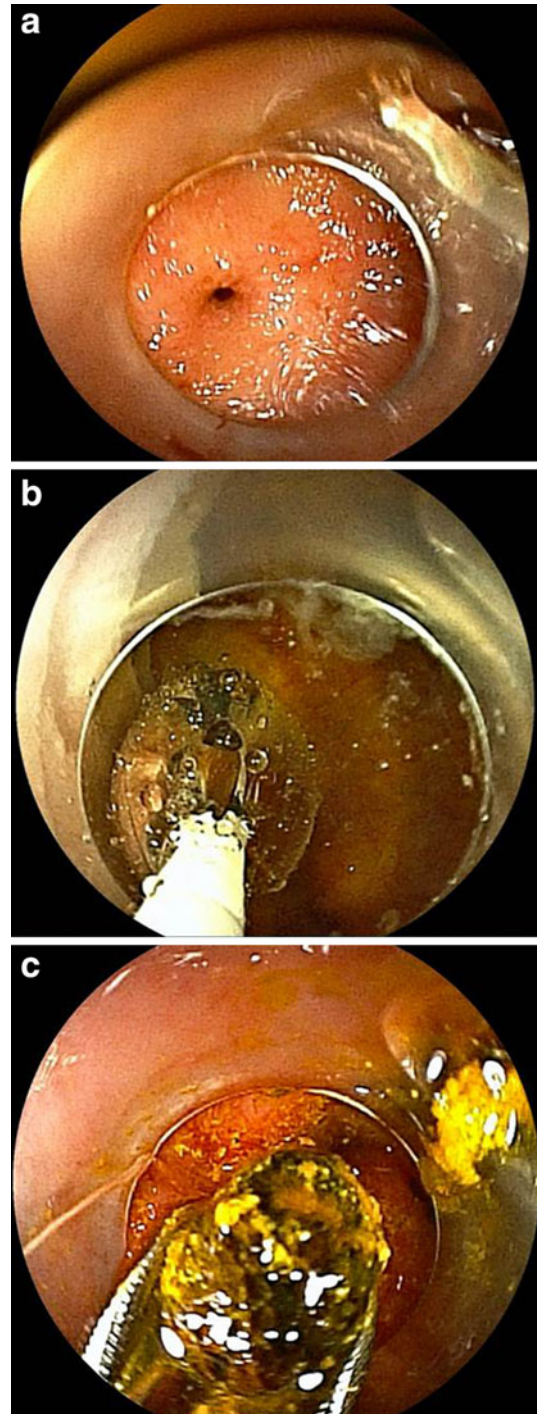


Fig. 3 **a** DBE showed stricture of the bilioenteric anastomosis in Case 1. **b** DBE allowed us to dilate the anastomotic stricture using balloon catheter in Case 1. **c** DBE allowed us to crush and remove the intrahepatic bile duct stones using a crusher catheter under direct observation in Case 1

pediatric patients. It is difficult to approach and reach the bilioenteric anastomosis through the Roux-en-Y reconstruction using a conventional endoscope. Moreover, the presence of postoperative intestinal adhesions can make it



Fig. 4 Direct cholangioscopy with DBE revealed multiple intrahepatic bile duct stones in Case 2

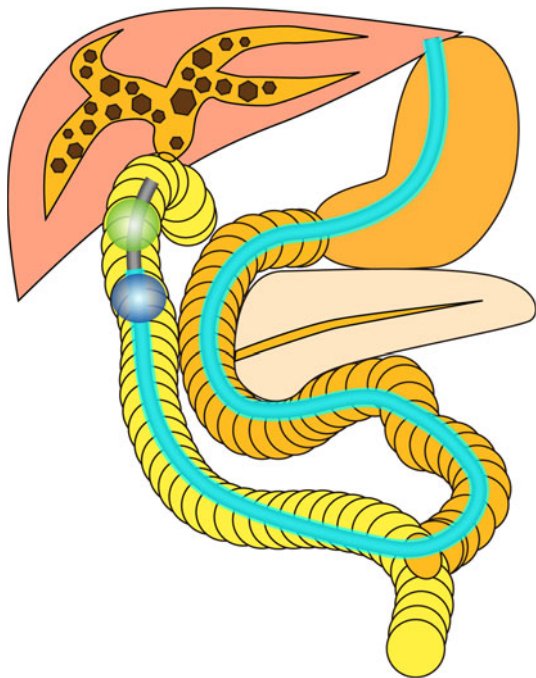


Fig. 5 The schema of DBE in a patient with intrahepatic bile duct stones following Roux-en Y reconstruction

more difficult in the cases with Roux-en-Y reconstruction. The surgical approach is also challenging, and necessarily includes the release of the intestinal adhesion, division of the bilioenteric anastomosis, removal of stones from the intrahepatic bile duct and re-anastomosis of the hepaticojejunostomy.

DBE was first reported in 2001 [1], and the device was released for sale in Japan, China and Europe in 2003, and has been widely used as a novel diagnostic and therapeutic method in patients with small bowel disease [7, 8]. Several studies recently reported its value and safety for

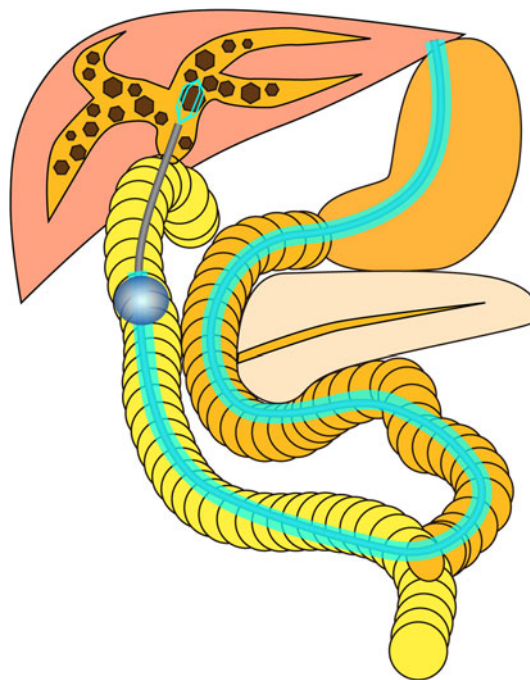


Fig. 6 The schema of biliary lithotripsy using direct cholangioscopy with DBE

applications in pediatric patients [9–12]. DBE in pediatric patients seems to be more difficult than in adult patients because the intestinal wall of pediatric patients is thin and the intestinal lumen is narrow. However, a more recent study with a large number of experiences described that the rate of major complications in pediatric patients was not significantly higher than that in adults [11].

We applied DBE as a therapeutic method for pediatric patients with IHBD stones after Roux-en-Y reconstruction. DBE has made it possible to reach the hepaticojejunal anastomosis endoscopically via the leg of the Roux-en-Y reconstruction, and to perform successful therapeutic interventions, including dilatation of the bilioenteric anastomotic stricture using balloon catheter through accessory channel and removal of IHBD stones using a crush catheter or basket catheter, without any major complications even in pediatric patients. These successful therapeutic managements using DBE for pediatric patients with IHBD stones help avoid the need for more invasive treatments, such as percutaneous transhepatic intervention and surgical procedures. Moreover, the present therapeutic procedure with DBE can be performed repeatedly. Based on our experiences, the youngest age at which we can perform DBE is three years.

In conclusion, although our experiences are limited, DBE is a less invasive and safe treatment modality for pediatric patients with IHBD stones. DBE is capable of reaching the bilioenteric anastomosis after RYH for CC, and is a useful therapeutic procedure in pediatric patients.

Conflict of interest Hironori Yamamoto has patents in Japan for the double-balloon system described in this article; he is a consultant to and received honoraria, grants and royalty from Fujifilm.

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