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【 CASE REPORT 】

Endoscopic Ultrasound-guided Hepaticogastrostomy in a Seven-year-old Girl

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Abstract:
Endoscopic ultrasound-guided hepaticogastrostomy (EUS-HGS) is an effective biliary drainage procedure in adult cases with difficult biliary access. However, there have been no reports on this procedure being used in pediatric cases. We successfully performed EUS-HGS in a pediatric case with a surgically altered anatomy. A standard convex-type echoendoscope and standard devices were used, and there were no device-related complications. The benefit of EUS-HGS for pediatric patients was avoidance of a percutaneous tube, which is difficult to maintain in active children. The accumulation of further cases and performance of a prospective study are warranted to standardize and expand the experience with this procedure.

Key words: EUS-HGS, intervention EUS, child

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Introduction

Endoscopic ultrasound (EUS) is widely used for both diagnostic and therapeutic purposes for pancreaticobiliary disease in adults (1). Currently, EUS-guided hepaticogastrostomy (EUS-HGS) is performed as a salvage drainage technique, instead of percutaneous transhepatic biliary drainage (PTBD), for cases with difficult/failed biliary access. However, there are few reports of therapeutic EUS in children (2, 3), and none for pediatric EUS-HGS.

We herein report a pediatric patient who underwent EUS-HGS for recurrent cholangitis.

Case Report

A seven-year-old girl underwent extrahepatic bile duct resection and hepaticojejunostomy for congenital biliary dilation with pancreatobiliary maljunction at three months old. Cholangitis occurred repeatedly because of a choledochojejunal anastomotic stricture (CJAS) beginning three months after the operation. The patient was treated conservatively, but her cholangitis recurred. Surgical re-anastomosis was performed at two years old, and no symptoms recurred until five years old. However, cholangitis recurred.

Computed tomography (CT) revealed dilatation of the intrahepatic bile duct (IHBD), and CJAS recurrence was suspected (Fig. 1a). Reoperation or a PTBD approach was considered, but the family and pediatric surgeon requested minimally invasive endoscopic treatment to minimize the patient's psychological and physical burden. However, conventional endoscopic retrograde cholangiopancreatography (ERCP) was difficult because of the surgically altered anatomy, and enteroscope-assisted ERCP was expected to be difficult and risky due to severe adhesions. After multidisciplinary discussions, we decided to perform EUS-HGS to dilate the stricture.

The patient (height 108 cm and weight 17.8 kg) was sedated with ketamine, midazolam and pentazocine without tracheal intubation by a pediatric surgeon. EUS-HGS was performed using a conventional curved linear array echoendoscope (EG 580 UT; Fujifilm, Tokyo, Japan). The B3

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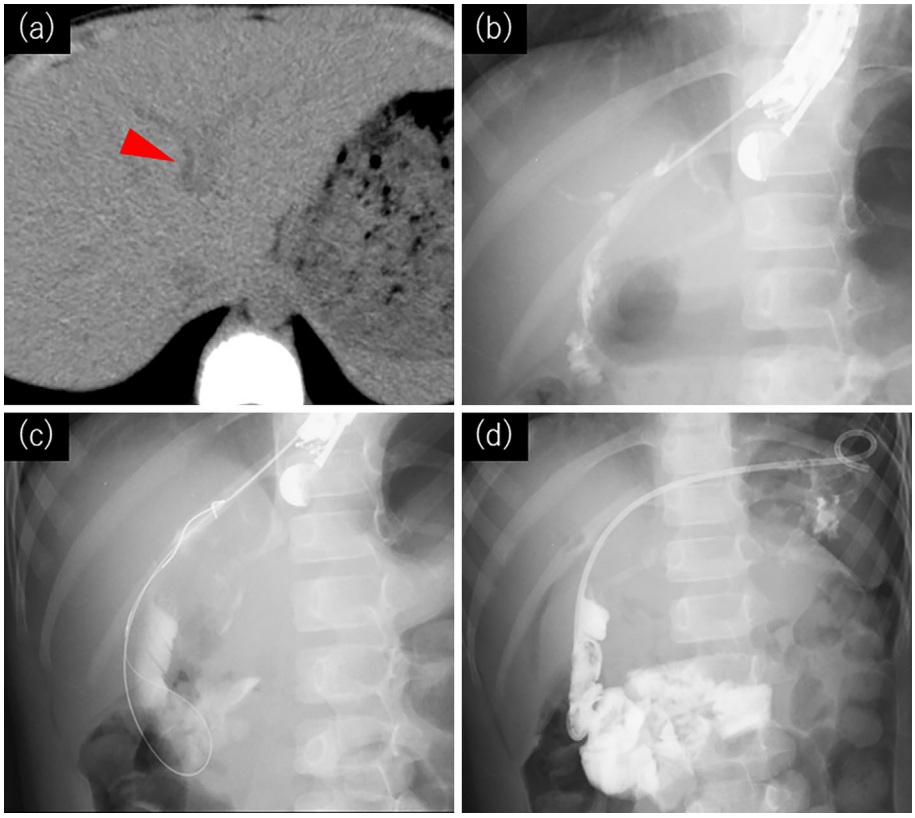

Figure 1. **CT findings and EUS procedures. (a) CT revealed dilatation of the intrahepatic bile duct (arrowhead). (b) The B3 branch was punctured by a 19-gauge needle under EUS guidance, and contrast medium was injected. (c) The guidewire was passed through the CJAS to the intestinal tract. (d) A 7-Fr double-pigtail stent was placed across the CJAS.**

branch was punctured via the stomach using a 19-gauge needle under EUS guidance (Fig. 1b). After injection of contrast medium, a guidewire (0.025 in VisiGlide2; Olympus Medical Systems, Tokyo, Japan) was advanced into the IHBD. We manipulated the guidewire gently and succeeded in passing the CJAS (Fig. 1c). Dilation of the puncture route and stricture was performed using an ultra-tapered mechanical dilator (ES dilator; Zeon Medical, Tokyo, Japan) and a 4-mm-diameter dilating balloon (REN; Kaneka Medix, Osaka, Japan), and finally a 7-Fr double pigtail stent (DPS) (Zimmon; Cook Medical Japan, Tokyo, Japan) was placed across the CJAS (Fig. 1d).

Two DPSs were placed two months after the initial treatment to dilate both the CJAS and the fistula created by EUS-HGS. In the third session, four months after the initial treatment, a per oral cholangioscope (POCS) was inserted, and there was no evidence of malignancy at the anastomotic site (Fig. 2a). The DPSs were changed every three to four months to maintain the fistula and dilate the CJAS. The CJAS gradually improved on cholangiography (Fig. 2b-d). The DPSs were removed 22 months after the initial procedure, and cholangitis did not recur thereafter.

Discussion

EUS-HGS has been reported as an effective biliary drainage procedure for adult patients with difficult/failed biliary access. However, severe adverse events, such as bleeding, perforation and peritonitis, have been reported, and only specialists can perform interventional EUS (1, 4). The use of interventional EUS for pediatric patients has been delayed because it is considered a more difficult and risky procedure for children. Therapeutic use of EUS in children has been mainly for drainage of pancreatic fluid collection and abdominal abscess (5), and there have been no reports of HGS in pediatric cases. Major obstacles to the expansion of EUS in children have been the unavailability of appropriate-size scopes and the small number of pediatric gastroenterologists with expertise in advanced endoscopic procedures (6). A dedicated duodenoscope (outer diameter of 7.5 mm) for ERCP has been developed for children, but a dedicated EUS scope is not available for children and must be the same size as that for an adult. Using a standard EUS scope (11-14 mm) may increase the risk of cervical esophageal perforation in children (6). However, we successfully performed EUS-HGS using a standard convex-type echoendoscope, and there were no scope-related complications. Standard devices for pancreatobiliary endotherapy were used and were effective, without any device-related complications. One difference in the EUS-HGS procedure between pediatric and adult cases is the puncture point. We puncture closer to the hilum in our pediatric patients than in adult patients because of in-

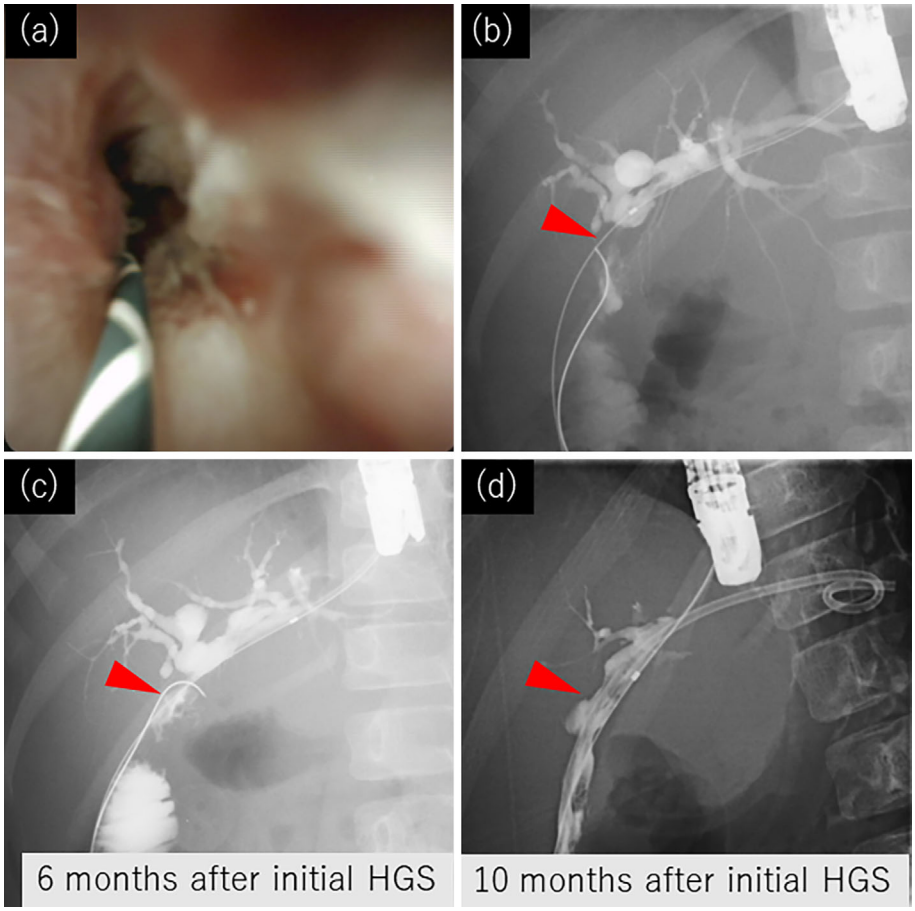

Figure 2. **Cholangiogram and X-ray findings of the CJAS. (a) Per-oral cholangioscopic findings of the CJAS suggested fibrotic stricture. (b) A cholangiogram of the initial drainage procedure revealed the CJAS. (c) The CJAS improved by six months after EUS-HGS. (d) A cholangiogram at 10 months after the initial procedure showed that the CJAS had almost completely resolved.**

sufficient dilation of the peripheral intrahepatic bile duct. Close attention should thus be paid to the puncture point when performing HGS in children.

Previously, PTBD was the first choice after failure of ERCP, but the quality of life was significantly impaired because of external fistula (7, 8). For pediatric patients, it is difficult to keep a PTBD tube in place for a long time, and there is a high risk of accidental removal during childhood activities. Furthermore, long-term tube placement can cause keloid formation on the body surface, which is also a major cosmetic disadvantage. EUS-HGS does not require restriction of exercise and does not damage the body surface. In addition, because fistula dilation requires prolonged treatment, long-term maintenance of EUS-HGS with plastic stents benefits the quality of life more than PTBD. In this regard, EUS-HGS is superior to PTBD because of the internal fistula.

Although the long-term prognosis remains unclear, we believe that the benefits of EUS-HGS over PTBD are obvious, particularly in the pediatric population. In EUS-HGS, the fistula closes spontaneously after the stents are removed. Therefore, the decision to remove stents should be made carefully, and there is no consensus on how long they should remain *in situ*. This is, to our knowledge, the first case of fistula dilation by EUS-HGS, and the stent was removed after 22 months in this patient. However, there have been several similar cases in adults, and in our experience, the stent can be removed approximately one year after the fistula dilation.

The main limitations of this procedure, to date, include experience in only a few cases and the unclear long-term prognosis. A lack of dedicated devices is also a limitation; however, the use of standard devices for pancreatobiliary endotherapy in pediatric patients has been demonstrated in our experience.

In conclusion, EUS-HGS and subsequent procedures through the matured fistula were both feasible and effective for treating pediatric patients. The main benefit of EUS-HGS for pediatric patients is the avoidance of the need for a percutaneous tube, which is difficult to maintain in active children. However, because the long-term results of EUS-HGS for children are unclear, the accumulation of further cases and performance of a prospective study are warranted to standardize and expand our experience with this procedure.

The authors state that they have no Conflict of Interest (COI).

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