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Utility of hepatobiliary scintigraphy for recurrent reflux cholangitis following choledochojejunostomy: A case report



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ABSTRACT

INTRODUCTION: Reflux cholangitis is a frequent complication of Roux-en-Y choledochojejunostomy.

PRESENTATION OF CASE: A 68-year-old male underwent left lobectomy of the liver, bile duct resection and choledochojejunostomy for intrahepatic cholangiocarcinoma located in Segment 2 of the liver, 40 mm in diameter with a lymph node metastasis 5 years ago. He had frequent recurrences of postoperative reflux cholangitis and hepatic abscesses and was treated with antibiotics each time. Postoperative adjuvant chemotherapy was scheduled, but due to recurrent cholangitis it was difficult. Although double balloon endoscopy for endoscopic retrograde cholangiography was performed, no stenosis was found in the choledochojejunostomy anastomosis, and no defect suspected of calculus and stenosis were found by contrast. Antibiotics had to be administered for a long time because it recurred when antibiotics were discontinued. This time, a tumor 2.0 cm in diameter was detected in segment 7 of the liver on follow-up computed tomography. The preoperative diagnosis was recurrent Intrahepatic cholangiocarcinoma. Hepatobiliary scintigraphy was carried out in preparation for concomitant treatment of his reflux cholangitis. Retention in the blind loop of the choledochojejunostomy was retarded, and the excretion was delayed. Therefore, hepatectomy and resection of the blind loop were performed. We confirmed improvement of stasis in the blind loop on postoperative hepatobiliary scintigraphy. The postoperative course was uneventful, and antibiotics were not required.

DISCUSSION: Hepatobiliary scintigraphy may be able to clarify the mechanism underlying reflux cholangitis.

CONCLUSION: Hepatobiliary scintigraphy was useful for the treatment of recurrent reflux cholangitis in this case.

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1. Introduction

Reflux cholangitis is the most common complication of Roux-en-Y choledochojejunostomy [1]. Sugawara et al. [2] recently reported that approximately 70% of patients who underwent hepatobiliary resection with choledochojejunostomy had a positive bile culture. Hence, cholangitis resulting from reflux of intestinal contents across the choledochojejunostomy is common, occurring in approximately 10% of patients [3]. Reflux cholangitis may result in

anastomotic strictures, stone recurrence, and liver abscesses [4]. Long-term reflux cholangitis could also increase the risk of tumorigenesis [5].

Abnormalities of the biliary tree resulting in stasis, reflux, or obstruction predispose the patient to symptomatic infection. Treatment for acute cases is based on supportive care and antibiotics, which are frequently recommend. Definitive treatment for stone disease, strictures, and other anatomic abnormalities requires endoscopic or surgical intervention. However, a clinically useful examination to determine the need for surgical intervention has not been established.

This case report has been reported in line with the SCARE criteria [6].

2. Presentation of case

A 68-year old male underwent left lobectomy of the liver with bile duct resection and choledochojejunostomy for the tumor

Abbreviations: ICC, intrahepatic cholangiocarcinoma; CT, computed tomography; S7, segment 7.

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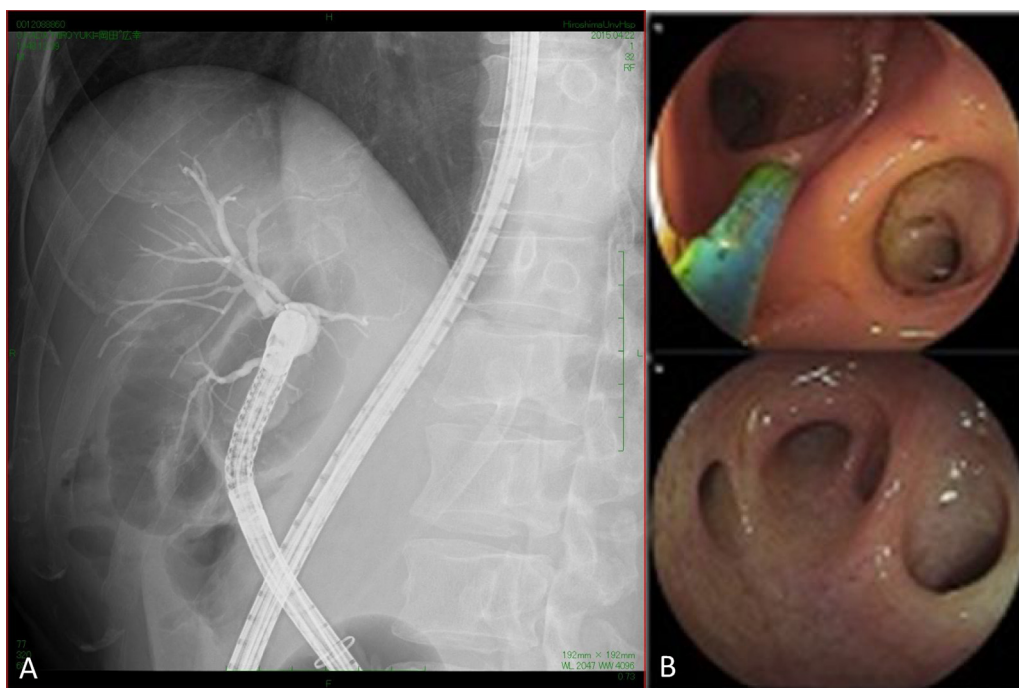


Fig. 1. Double-balloon endoscopy for endoscopic retrograde cholangiography. A. There were no defects suspected of calculus and no stenosis. B. Cannulation was performed into the bile duct.

Table 1
Representative febrile episodes and infectious work-up.

Time from postoperation (month)	Symptoms	diagnosis	Bile cultures	Treatment
2	Fever	Reflux cholangitis, Liver abscess	Klebsiella pneumoniae	MEPM → LVFX
3	Fever	Reflux cholangitis, Liver abscess	Enterococcus faecalis	MEPM → LVFX
4	Fever	Reflux cholangitis	Undetectable	MEPM
12	Fever	Reflux cholangitis	Undetectable	SBT/CPZ → SBT/ABPC
16	Fever	Reflux cholangitis	Undetectable	CPFX
25	Fever	Reflux cholangitis	Undetectable	CPFX
33	Fever	Reflux cholangitis	Undetectable	CPFX
36	Fever	Reflux cholangitis	Enterococcus faecalis	CPFX
48	Fever	Reflux cholangitis	Citrobacter freundii	MEPM

MEPM, meropenem; LVFX, levofloxacin; CPFX, ciprofloxacin SBT/CPZ, sulbactam/cefoperazone; SBT/ABPC, sulbactam/ampicillin.

located in Segment 2 of the liver, 40 mm in diameter with a lymph node metastasis 5 years ago. The pathological diagnosis was intrahepatic cholangiocarcinoma (ICC) and there was one metastasis to the lymph node. He was hepatitis B positive and administered and was administered entecavir. Although the ICC has not recurred so far, he has had frequent recurrences of postoperative reflux cholangitis and hepatic abscess and was treated with antibiotics each time. Postoperative adjuvant chemotherapy was scheduled, but due to recurrent cholangitis it was difficult. Every time reflux cholangitis occurred, double-balloon endoscopy for endoscopic retrograde cholangiography was performed. There is no stenosis in the choledochojunostomy anastomosis. The anterior segment and posterior segment bile duct is visualized by contrast. There were no defects suspected of calculus and no stenosis (Fig. 1A). Cannulation was performed into the bile duct (Fig. 1B), bile was collected and cultured. During the 5 years, he experienced several febrile episodes caused by the reflux cholangitis. He was admitted to our hospital with sudden fever and recovered with antibiotics determined by bile culture results (Table 1). After discharging, he continuously administered the antibiotics for about few weeks, but cholangitis recurred in a week after he stopped the antibiotics. As the cholangitis recurred every time the antibiotics were stopped, he was prescribed levofloxacin or ciprofloxacin on long-term antibiotic prophylaxis.

A tumor 2.0 cm in diameter was detected in segment 7 (S7) of the liver on follow – up computed tomography (CT). ICC recurrence was suspected and he was admitted to our hospital for surgical treatment of the hepatic tumor. The patient was asymptomatic and had no abnormal findings on physical examination. Laboratory findings on admission included white blood cell and platelet counts of 4040/ μ L and 162000/ μ L, respectively; hemoglobin, albumin, and total bilirubin levels of 13.4 g/dL, 4.2 g/dL, and 0.6 mg/dL, respectively; and aspartate and alanine aminotransferase, alkaline phosphatase, and gamma-glutamyl transpeptidase concentrations of 23 U/L, 17 U/L, 234 U/L, and 18 U/L, respectively. The patient had a prothrombin time (percent) of 91%. His Child-Pugh grade was A. The indocyanine green retention rate at 15 min was 10.8%. The results for hepatitis B virus antibody were positive. Serum duke pancreatic monoclonal antigen type 2 was elevated (4500.0 U/mL). Serum carcinoembryonic antigen (1.8 ng/mL), carbohydrate antigen 19-9 (17 U/mL), alpha-fetoprotein (2.1 ng/mL), protein induced by the vitamin K antagonist (22 mAU/mL), and s-pancreas-1 antigen (14.0 U/mL) levels were within normal reference limits. Abdominal CT scans revealed heterogeneous internal enhancement of the tumor in the arterial and venous phases. There was no lymph node enlargement (Fig. 2). Abdominal magnetic resonance imaging revealed low signal intensity on T1-weighted images, high signal intensity on T2-weighted images, and high signal intensity on dif-

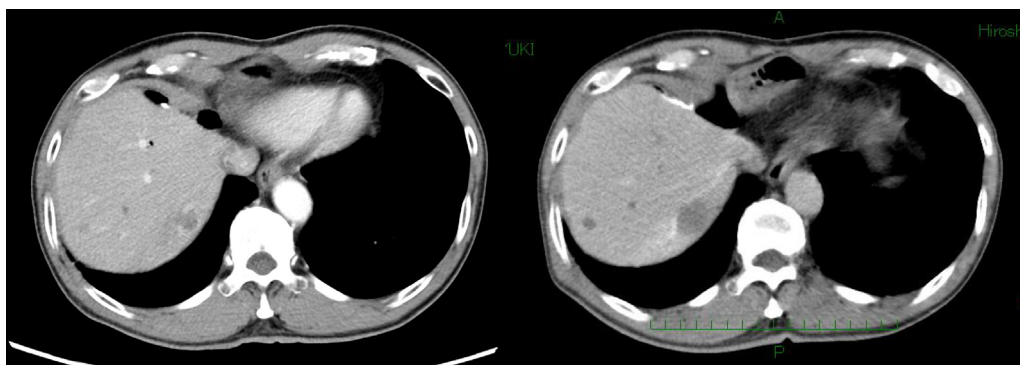


Fig. 2. Abdominal computed tomography scans.

The tumor in S7 was 2.0cm in diameter and showed heterogeneous internal enhancement in both the arterial and venous phases.

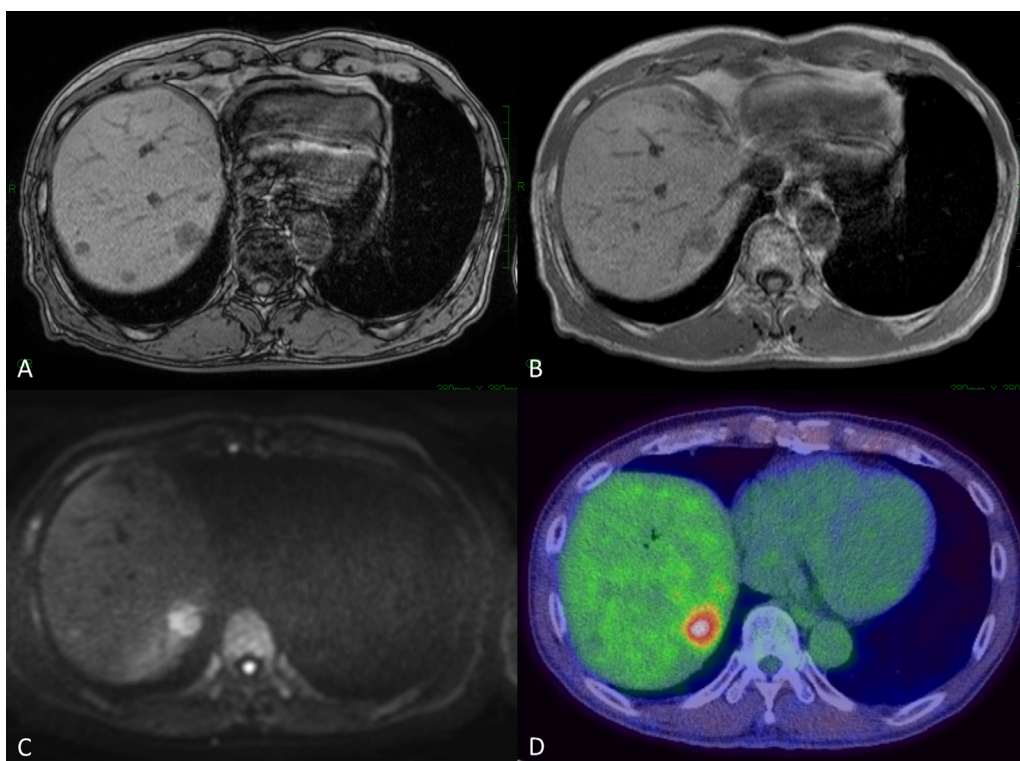


Fig. 3. Abdominal magnetic resonance imaging and Positron emission tomography.

A. The tumor exhibited low signal intensity on T1-weighted images. B. The tumor exhibited high signal intensity on T2-weighted images. C. The tumor exhibited high signal intensity on diffusion weighted images. D. Positron emission tomography revealed a maximum standardized uptake value of 5.4 in the tumor.

fusion weighted images (Fig. 3A–C). Positron emission tomography revealed a maximum standardized uptake value of 5.4 in the tumor (Fig. 3D). A preoperative diagnosis of recurrent ICC was made. In addition, the patient had recurrent reflux cholangitis. Therefore, hepatobiliary scintigraphy was carried out in consideration of concomitant treatment of the reflux cholangitis. Hepatobiliary scintigraphy revealed that there was no delayed excretion in the liver, but retention in the blind loop of the choledochojejunostomy was retarded, and the excretion was delayed (Fig. 4A). Due to the long blind loop, it was thought that bacteria proliferated at the blind loop and caused reflux cholangitis.

Therefore, we decided to treat the ICC and reflux cholangitis at the same time. Partial resection of S7 of the liver and blind loop resection were performed. The blind loop was not expanded or twisted, but its length was 10 cm. It was resected and shortened to 1 cm in length. Histopathological examination revealed that the tumor was a combined hepatocellular and cholangiocarcinoma.

Postoperative hepatobiliary scintigraphy was performed to confirm improvement of stasis in the blind loop (Fig. 4B). The postoperative course was uneventful and antibiotics were not required. The patient was discharged on the 21st postoperative day. And half a year has passed, but antibiotics were not required.

3. Discussion

We report the utility of hepatobiliary scintigraphy for recurrent reflux cholangitis following Roux-en-Y choledochojejunostomy. We performed blind loop resection and partial resection of the liver in a patient who developed multiple episodes of cholangitis several years after Roux-en-Y choledochojejunostomy and was found to have a combined hepatocellular and cholangiocarcinoma. We performed hepatobiliary scintigraphy and detected that the blind loop of Roux-en-Y choledochojejunostomy caused stasis and cholangitis.

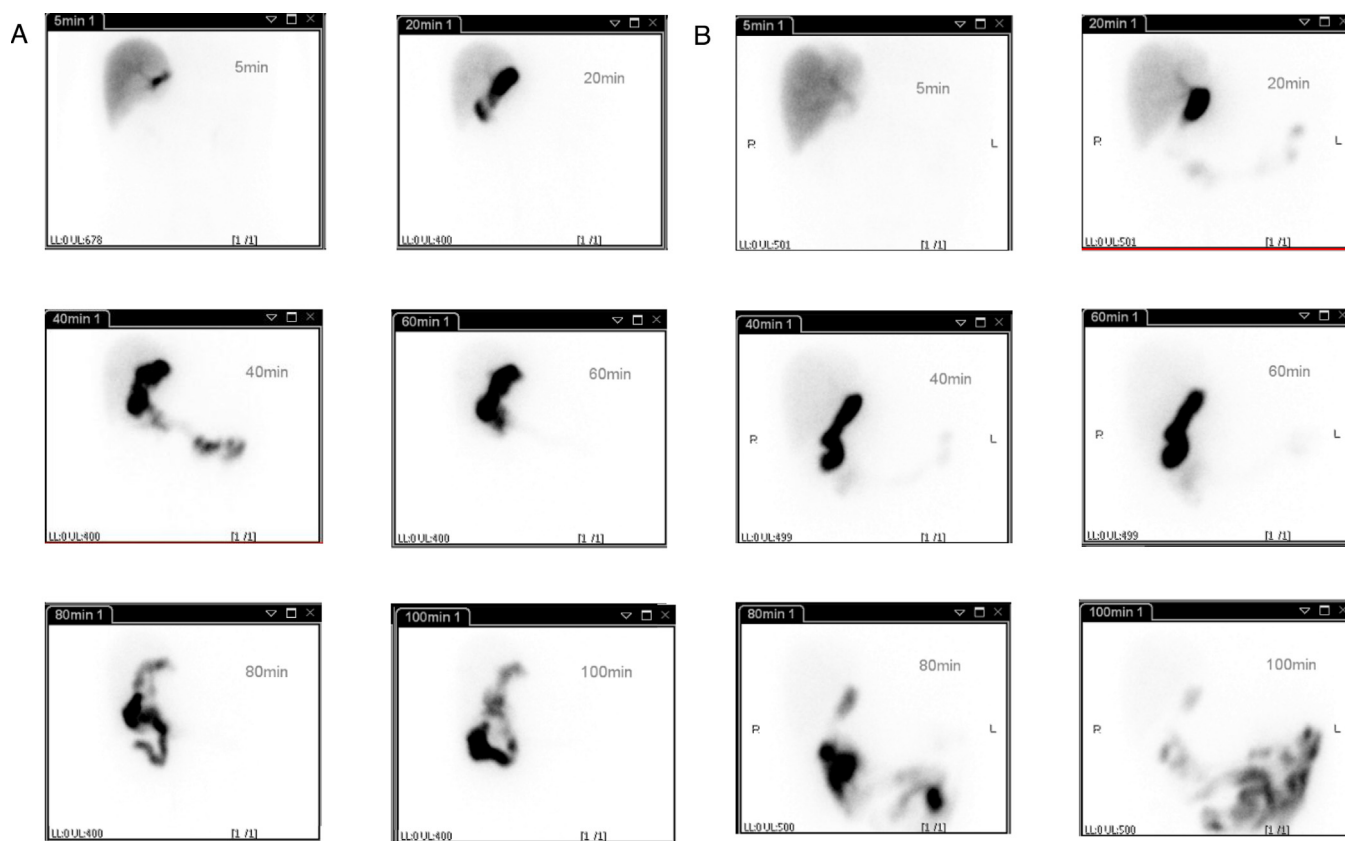


Fig. 4. Hepatobiliary scintigraphy.

A. There was no delayed excretion in the liver, but retention in the blind loop of the choledochojejunostomy was retarded and the excretion was delayed. B. Stasis in the blind loop was improved.

There are several mechanisms that may underlie reflux cholangitis. One such mechanism is that, in Roux-en-Y choledochojejunostomy, the proximal jejunal loop is usually long and has an inverted C shape. Contents in the proximal jejunum easily enter the distal jejunum upon anastomosis and then move in a retrograde direction to reach the bile duct, a risk factor associated with postoperative reflux cholangitis. Another mechanism is that the once the jejunum is transected, as is commonly performed, its motility and electrical activity are greatly affected. This can cause reverse peristalsis [7], and as a result, the intestinal contents can become stagnant, thus favoring bacterial colonization and leading to postoperative cholangitis and abnormal liver function [8]. Finally, a motility disorder might arise in the mobilized Roux-en-Y loop, allowing the intestinal contents to reflux [9]. Antireflux procedures may prevent further infections. Similar consequences of dysmotility were observed following gastrointestinal reconstructions [10].

We performed double-balloon endoscopic retrograde cholangiography first, but there were no calculus in the bile duct and no stenosis in choledochojejunostomy anastomosis which caused reflux cholangitis. Hepatobiliary scintigraphy was performed to clarify the mechanism of reflux cholangitis in our patient. Hepatobiliary scintigraphy has been shown to be an accurate, noninvasive method to diagnose biliary complications such as obstruction or leak in patients undergoing choledochojejunostomy [11]. We have often noticed a focus of persistent retention in the region near the biliary anastomosis. Defining the typical features of this retention, as well as delayed imaging, may be useful in discriminating this normal variant from pathological stasis.

We proposed a mechanism to explain the recurrent reflux cholangitis. We hypothesized that stasis in the blind loop promoted

bacterial overgrowth resulting in repeated episodes of infection. Resection of the blind loop prevented further episodes of cholangitis.

In the present case, medical therapies resulted in recurrent infections and antibiotic resistance in a patient with cholangitis following choledochojejunostomy. Resection of the blind loop should be considered for the treatment of recurrent cholangitis occurring after choledochojejunostomy. Since hepatobiliary scintigraphy can evaluate physiological bile flow, even in a Roux-en-Y choledochojejunostomy that seems normal (without expansion or twist) on CT, it was very useful to accurately detect stasis in the blind loop that had been causing the reflux cholangitis.

4. Conclusion

Hepatobiliary scintigraphy was useful for the diagnosis of recurrent reflux cholangitis in this case. It could be adjunct to other examination in diagnosing reflux cholangitis in similar patients.

Conflicts of interest

None.

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Ethical approval

In the Hiroshima University Ethical Committee, the Hiroshima University “Medical Research for Human” regulations stipulates

that it is necessary to apply for research if it is a research or intervention involving invasion. Because our report is not included in this regulation, ethical approval has been exempted.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Author contributions

All authors contributed to this work, and approved the final manuscript.

Registration of research studies

None.

Guarantor

Hiroyuki Tahara.

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