CASE REPORT

Endoscopic Management of Acute Cholecystitis and Cholangitis Caused by Limy Bile

Sang Heon Lee, Jong Ho Moon, Hyun Jong Choi, Hyung Ki Kim, Young Deok Cho, Moon Sung Lee, and Chan Sup Shim

Digestive Disease Center, Department of Internal Medicine, Soon Chun Hyang University School of Medicine, Bucheon and Seoul, Korea

Limy bile is a relatively rare condition in which a radiopaque material is visible in the gallbladder, extending rarely into the bile duct, on plain radiography. Acute cholangitis or cholecystitis caused by limy bile is a very rare condition. There are no definite treatment guidelines for limy bile, but in most cases with cholangitis or cholecystitis, laparoscopic cholecystectomy has been the preferred treatment. We report a case of limy bile with biliary symptoms that was treated only with an endoscopic procedure. (**Gut and Liver 2009;3:349-351**)

Key Words: Limy bile; Cholangitis, Cholecystitis; Endoscopic transpapillary gallbladder drainage

INTRODUCTION

Limy bile is a rare disorder in which the gallbladder (GB) is filled with a thick paste-like radiopaque material, which extends rarely into the common bile duct (CBD).^{1,2} Since Churchman's description of this syndrome in 1911, more than 300 cases have been reported in the literature, including 7% with limy bile in both the GB and CBD.³ Tsukamoto *et al.*⁴ reported 26 cases in which obstructive jaundice was accompanied by limy bile. In cases with biliary symptoms accompanying limy bile, laparoscopic cholecystectomy is a less invasive option than open surgery, and its safety has been established.⁵ Nevertheless, patients still incur the risks associated with general anesthesia and longer hospital admission period.

We experienced a case of limy bile with acute cholangitis and cholecystitis that was treated with endoscopy

without surgical intervention.

CASE REPORT

A 45-year-old female presented in the outpatient department with right upper quadrant pain, fever, and chills for 3 days duration. She did not have a specific medical treatment history. Laboratory findings were as follows: white blood cell count, 13,200/mm³; total/direct bilirubin, 7.73/5.52 mg/dL; gamma-glutamyl transferase, 797 IU/L; alkaline phosphatase, 232 IU/L; AST/ALT, 348/759 IU/L; and amylase/lipase, 49/32 IU/L.

Computed tomography showed high-density material in a dependent position of the GB and CBD. CBD dilatation was about 1.3 mm (Fig. 1). The patient was diagnosed with obstructive jaundice due to choledocholithiasis, which entails limy bile.

A duodenoscopy revealed diffusely enlarged major papilla. An endoscopic retrograde cholangiography (ERCP) revealed a 14-mm filling defect at the distal CBD. After an endoscopic sphincterectomy (EST), limy bile materials were washed out by sweeping with a balloon catheter (Fig. 2), and we inserted endoscopic nasobiliary drainage (ENBD). After then, clinical finding of the patient was improved dramatically, and surgical treatment such as a laparoscopic cholecystectomy was postponed. Four days after the first ERCP, we repeated the ERCP and again found a filling defect in the distal CBD. After completely removing limy bile from the CBD, we inserted a guide wire into the GB through the cystic duct, and a 7 Fr double-pigtail catheter (length, 15 cm) was introduced to the

Correspondence to: Jong Ho Moon

Digestive Disease Center, Soon Chun Hyang University Bucheon Hospital, Soon Chun Hyang University School of Medicine, 1174, Jung-dong, Wonmi-gu, Bucheon 420-767, Korea

Tel: +82-32-621-5094, Fax: +82-32-621-5080, E-mail: jhmoon@schbc.ac.kr

Received on April 23, 2009. Accepted on May 21, 2009.

DOI: 10.5009/gnl.2009.3.4.349

Fig. 1. (A, B) Computed tomography showing high-density material in the dependent position of the gallbladder and common bile duct.

Fig. 2. Extraction of limy bile from the common bile duct after endoscopic sphincterotomy.

Fig. 3. Endoscopic transpapillary gallbladder drainage for the treatment of acute cholecystitis.

Fig. 4. (A, B) Follow-up computed tomography at 6 months showing the complete absence of limy bile in the gallbladder and common bile duct.

GB to evacuate limy bile (Fig. 3). The patient recovered completely after the endoscopic procedure.

Six months later, she was admitted for follow-up examination. During the follow up of six months, she was administered daily medication consisting of 600 mg

of UDCA (Ursa[®]; Daewoong, Seoul, Korea) and 300 mg of a terpene preparation (Rowachol[®]; Rowa Pharma, Cork, Ireland). Laboratory findings were within normal ranges, and follow-up studies, including a CT, showed the complete evacuation of limy bile from the biliary system

and GB (Fig. 4). The double-pigtail stent inserted to the GB was removed.

DISCUSSION

Limy bile is an uncommon condition in which the gallbladder and bile duct are filled with radiopaque material, readily noted on plain radiographs.^{1,2} Since the first description of this syndrome in 1911 by Churchman, more than 300 cases of limy bile have been reported.³

The presence of limy bile in the CBD is very rare, with only a few reported cases. Thus, when obstructive jaundice occurs with limy bile syndrome, there are no definite treatment guidelines. The most frequently used treatment to date is cholecystectomy; cholecystectomy is performed for limy bile in the GB, and cholecystectomy plus insertion of a T-tube into the CBD after removal of limy bile are performed for limy bile in the GB and the CBD.⁶ Recently, laparoscopic cholecystectomy for focal limy bile in the gallbladder has been reported,⁵ and a combined application of EST and laparoscopic cholecystectomy was used to treat limy bile in both the GB and CBD.⁷

In the present case, we performed EST to treat the cholangitis caused by biliary lithiasis. In most cases of limy bile syndrome, laparoscopic cholecystectomy is a safe procedure for treatment of acute cholecystitis. However, in patients at increased surgical risk, endoscopic drainage such as endoscopic transpapillary gallbladder drainage (ETGD) is an alternative treatment for acute cholecystitis.^{8,9} Itoi *et al.*⁹ reported that among 36 acute cholecystitis patients who received ETGD, 35 showed clinically favorable response without any serious procedure-related complications.

Given that limy bile is a movable precipitant of calcium carbonate combined with cholesterol,¹⁰ we postulated that

ETGD could be used to drain limy bile and performed ETGD in this patient. Six months after the procedure, we observed complete evacuation of limy bile from the GB. Though surgery is the mainstay of the treatment for limy bile, endoscopic treatment such as EST and/or ETGD may be considered in selective patients, especially a high-risk candidate for operation.

REFERENCES

1. Naryshkin S, Trotman BW, Raffensperger EC. Milk of calcium bile. Evidence that gallbladder stasis is a key factor. *Dig Dis Sci* 1987;32:1051-1055.
2. Ballas KD, Alatsakis MB, Rafailidis SF, Psarras K, Sakadamis AK. Limy bile syndrome: review of seven cases. *ANZ J Surg* 2005;75:787-789.
3. Sava G, Millot P, Becmeur F, Vaxman F, Grenier JF. Limy bile syndrome. Study of a case with double localization in the gallbladder and common bile duct. *Gastroenterol Clin Biol* 1988;12:156-159.
4. Tsukamoto T, Ohta Y, Shuto T, et al. Limy bile: a case of obstructive jaundice and review of 26 cases. *Osaka City Med J* 2003;49:67-70.
5. Onghena T, De Waele JJ, Vereecken L, Van Loon C. Limy bile and laparoscopic cholecystectomy. *Acta Chir Belg* 2001;101:31-34.
6. Moreaux J, Roux JM. Limy bile. A surgical experience in 16 patients. *Gastroenterol Clin Biol* 1994;18:550-555.
7. Takatori Y, Yamauchi K, Negoro Y, et al. Limy bile syndrome complicated with primary hyperparathyroidism. *Intern Med* 2003;42:44-47.
8. Kjaer DW, Kruse A, Funch-Jensen P. Endoscopic gallbladder drainage of patients with acute cholecystitis. *Endoscopy* 2007;39:304-308.
9. Itoi T, Sofuni A, Itokawa F, et al. Endoscopic transpapillary gallbladder drainage in patients with acute cholecystitis in whom percutaneous transhepatic approach is contraindicated or anatomically impossible (with video). *Gastrointest Endosc* 2008;68:455-460.
10. Cooke M. Limy bile. *Proc R Soc Med* 1968;61:1110-1112.