Limy Bile Syndrome Complicated with Primary Hyperparathyroidism

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Abstract

Limy bile is a rare condition characterized by excessive precipitation of calcium carbonate in the gallbladder. Cases of complicated hyperparathyroidism are extremely rare. There is only one reported case, where serum and urine calcium levels were high. On the other hand, the presence of limy bile in the common bile duct is also very rare, and has been reported in only 20 cases. We report a patient with obstructive jaundice in whom the initial abdominal radiography showed limy bile in the gallbladder and common bile duct, and laboratory values showed a high serum calcium level and highly sensitive PTH (HS-PTH) level.

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Key words: limy bile syndrome, hyperparathyroidism, hypercalcemia, obstructive jaundice

Introduction

Limy bile is a rare condition in which a radiopaque gallbladder and/or bile ducts are noted on plain abdominal radiography. Limy bile is caused by calcium carbonate precipitation in the bile and is usually associated with distal biliary tract obstruction. The etiology of limy bile is unclear. We report the clinical course of a man, who experienced limy bile syndrome complicated with hyperparathyroidism.

For editorial comment, see p 1.

Case Report

A 64-year-old man, presented with mild epigastric pain for 10 days accompanied by jaundice and vomiting. He had mild tenderness in the epigastrium and right upper quadrant. Laboratory findings were as follows: aspartate transaminase (AST): 426 IU/l (normal, 8 to 40 IU/l); alanine transaminase (ALT): 641 IU/l (normal, 5 to 35 IU/l); ALP: 1,400 IU/l (normal, 104 to 338 IU/l); total bilirubin: 6.3 mg/dl (normal, 0.2 to 1.2 mg/dl); conjugated bilirubin: 4.9 mg/dl (normal, 0.0 to 0.4 mg/dl); serum calcium: 10.9 mg/dl (normal, 8.0 to 10.0 mg/dl) and highly sensitive PTH (HS-PTH): 850 pg/ml (normal, 160 to 520 pg/ml) (Table 1). Plain abdominal radiography on admission showed a visible gallbladder and common bile duct, with a calcium density in them (Fig. 1). Ultrasound also showed an acoustic shadow consistent with gallstones and common bile duct dilatation (Fig. 3). Since the patient had never received radiopaque agents, diagnosis of limy bile was made. The patient was diagnosed as having obstructive jaundice due to choledocholithiasis which entails limy bile, and therefore, laparoscopic cholecystectomy would be done after improving inflammation. An endoscopic retrograde cholangiography (ERC) and endoscopic sphincterotomy (EST) were done. We showed the dilated common bile duct within some stones and limy bile (Fig. 4) as well as excretion of the limy bile with bile after EST (Fig. 5). Analysis of limy bile showed a composition of 98% calcium carbonate. The plain abdominal radiography taken on the next day demonstrated the radiopacity of the gallbladder as showing no remarkable change, however, the common bile duct almost disappeared and limy bile had moved around the rectum (Fig. 6). On plain abdominal radiography, limy bile in the gallbladder showed no significant change on the 11th day after EST, however these findings disappeared 1 month later. His condition gradually improved, with serum bilirubin and transaminase levels only mildly elevated on the 19th day. However, he experienced recurring abdominal pain in addition to elevation of serum bilirubin and transaminase...
levels on the 23rd day. The symptoms disappeared the next day and serum bilirubin and transaminase levels gradually improved again, so it was thought that the clinical course occurred by the strangulation of the CBD stone, and it improved spontaneously. Preventing the recurrence of cholesterolithiasis and cholecystitis, laparoscopic cholecystectomy was operated 2 months later. There were three stones without limy bile in the gallbladder, of which the biggest one was 10 mm in diameter. Figure 3 shows ultrasound of the gallbladder and common bile duct with gallstones and common bile duct dilatation (1.0 cm).

**Table 1. Laboratory Findings on Admission**

<table>
<thead>
<tr>
<th>Test</th>
<th>Value</th>
<th>Test</th>
<th>Value</th>
<th>Test</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>WBC</td>
<td>6,400/μl</td>
<td>CRP</td>
<td>0.56 mg/dl</td>
<td>HS-PTH</td>
<td>850 pg/ml</td>
</tr>
<tr>
<td>RBC</td>
<td>536×10⁶/μl</td>
<td>AST</td>
<td>426 IU/l</td>
<td>HbAC</td>
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<tr>
<td>Hb</td>
<td>17.1 g/dl</td>
<td>ALT</td>
<td>641 IU/l</td>
<td>CEA</td>
<td>9.4 ng/ml</td>
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<tr>
<td>Ht</td>
<td>49.6%</td>
<td>LDH</td>
<td>308 IU/l</td>
<td>CA19-9</td>
<td>1,390 U/ml</td>
</tr>
<tr>
<td>PLT</td>
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<td>ALP</td>
<td>1,400 IU/l</td>
<td>Gastrin</td>
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<tr>
<td>T.P</td>
<td>7.5 g/dl</td>
<td>LAP</td>
<td>412 IU/l</td>
<td>Calcitonin</td>
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<tr>
<td>Alb</td>
<td>4.4 g/dl</td>
<td>γ-GTP</td>
<td>1,705 IU/l</td>
<td>Thyroglobulin</td>
<td>14 ng/ml</td>
</tr>
<tr>
<td>BUN</td>
<td>19.6 mg/dl</td>
<td>T.Bil</td>
<td>6.3 mg/dl</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cr</td>
<td>1.04 mg/dl</td>
<td>D.Bil</td>
<td>4.9 mg/dl</td>
<td></td>
<td></td>
</tr>
<tr>
<td>UA</td>
<td>5.3 mg/dl</td>
<td>I.Bil</td>
<td>1.4 mg/dl</td>
<td>Protein</td>
<td>(4+)</td>
</tr>
<tr>
<td>Na</td>
<td>135 mEq/l</td>
<td>ZTT</td>
<td>2.0 KU</td>
<td>Glucose</td>
<td>(4+)</td>
</tr>
<tr>
<td>K</td>
<td>4.4 mEq/l</td>
<td>TTT</td>
<td>1.0 KU</td>
<td>Occult blood</td>
<td>(+)</td>
</tr>
<tr>
<td>Cl</td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ca</td>
<td>10.9 mg/dl</td>
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</tr>
</tbody>
</table>

**Figure 1.** Plain abdominal radiography on admission showing limy bile in the gallbladder and common bile duct.

**Figure 2.** Abdominal CT showing radiopaque material filled gallbladder and common bile duct.

**Figure 3.** Ultrasound showed an acoustic shadow consistent with gallstones and common bile duct dilatation (1.0 cm).
Figure 4. Plain abdominal radiography before ERC showing limy bile in the gallbladder and common bile duct, and stone in the lower end of the common bile duct (left). After ERC it is more clear.

Figure 5. On EST, limy bile flowed from the orifice.

Figure 6. Plain abdominal radiography after EST showing disappearance of limy bile from the common bile duct and migration around the rectum.

Figure 7. Three stones, of which the largest one was 10 mm in diameter, were seen in the opened gallbladder.

Discussion

Lumpy bile is an uncommon condition in which the gallbladder and bile ducts are filled with radiopaque material readily noted on plain radiographs. Since the first description of this syndrome in 1911 by Churchman (2), over 300 cases of limy bile have been reported (3).

This syndrome is considered as 1–2% of cholecystolithiasis. Due to improvement of the diagnosis technique and recognition recently, such reports show a tendency of increasing. As to the etiology of limy bile, many theories, clinical or experimental, have been suggested in the past.
Obstruction or narrowing of the cystic duct

Okamura et al (4) reported that obstruction of the cystic duct is present in 93%.

Inflammation of the gallbladder wall

McWhirter (5) suggested that low-grade inflammation of the gallbladder is needed for the deposition of calcium.

Change in pH of the bile

It has been supposed that the obstruction at the cystic duct as well as infection or inflammation of the gallbladder may cause an alkaline swing of the pH of the gallbladder bile. In addition, when the pH is greater than 6.6 (6), calcium carbonate is crystallized from the solution.

Other systemic factors

Green reported a case whose serum and urine calcium levels were high, and he suggested the presence of an anomaly in the calcium metabolism. However, none of these cases have been hitherto reported, because no careful studies have been made on calcium metabolism. Therefore, the present case was extremely rare to be associated with primary hyperparathyroidism, and because serum Ca and PTH levels were high. However obstruction of the cystic duct and inflammation of the gallbladder wall seemed to exist in this case, and it was thought that if serum calcium levels were high for hyperparathyroidism, calcium would be easily deposited and would consist of limy bile.

The presence of limy bile in the common bile duct is very rare, and has been reported in some cases. According to Save (3), only 20 cases among 300 limy bile reports were reported as limy bile located in both the gallbladder and common bile duct. Only 9 cases were reported in Japan (7). The double localization always occurred in patients with limy bile and gallstone. This association suggests that limy bile in the common bile duct is due to migration of small impacted stones from the cystic duct or the neck of the gallbladder, and migration of the calcareous material from the gallbladder to the common bile duct (8).

In patients with limy bile extending to the common bile duct and having obstructive jaundice, evacuation of the impacted stone is necessary. Spontaneous evacuation of the stone through the papilla of Vater probably occurred. A few cases of spontaneous disappearance of limy bile have been reported (9, 10). This spontaneous evacuation of the calcareous material through the papilla may be explained by the small size of the migrating stones and the creamy consistency of limy bile mixed with normal bile. When spontaneous clearance does not occur, clearance of the common bile duct through endoscopic naso-biliary drainage (ENBD) or EST is the treatment of choice. Afterwards, laparoscopic cholecystectomy is a good indication for preventing the recurrence of choledocholithiasis. In the present case, EST was extremely effective.

Conclusions

We had the rare opportunity of encountering limy bile syndrome complicated with hyperparathyroidism. This case illustrates the effectiveness of EST and laparoscopic cholecystectomy.

References