Case Report

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Primary Sclerosing Cholangitis Associated with Limy Bile and Acute Pancreatitis

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Key Words
Sclerosing cholangitis, primary · Pancreatitis, acute · Bile, limy

Abstract
Primary sclerosing cholangitis (PSC) as the cause of acute pancreatitis (AP) is a rare phenomenon. Here, we report the first case of PSC associated with limy bile (LB) as well as AP. In this case, spontaneous outflow of the LB occurred, and the AP resolved on its own with conservative management. In addition to the administration of ursodeoxycholic acid, endoscopic intervention was undertaken and the patient remains symptom free to date. This report describes a unique case in which PSC, LB, and AP occurred in the same patient. The association of these rare conditions is also discussed.

Introduction

Primary sclerosing cholangitis (PSC) as the cause of acute pancreatitis (AP) is a rare phenomenon [1]. Limy bile (LB) is also a rare condition caused by calcium carbonate precipitation in the bile [2]. PSC complicated with both LB and AP has not previously been reported. Spontaneous flow of LB from the gallbladder to the common bile duct and from the common bile duct to the duodenum has been reported [3, 4] and can be a cause of transient obstructive jaundice and AP [4]. Regarding the pathogenesis of AP, it has been shown by a prospective study that biliary sludge is an underestimated cause of idiopathic AP [5].

Case Report

A 34-year-old male was diagnosed as having PSC with elevated biliary enzyme in December 1987. Ursodeoxycholic acid (UDCA) had been administered since then. His first episode of AP occurred in January 1997, and required hospital admission. He was hospitalized with AP again in July 1998. The etiology of these two attacks of AP was thought to be unknown. After that, he repeatedly experienced epigastralgia and spontaneous relief. In February 2000, he was readmitted with severe epigastralgia due to AP. He did not consume alcohol and was on no medication other than ursodeoxycholic acid. He had no family history of AP, and there was no history of metabolic disorders such as hypertriglyceridemia or hypercalcemia. The laboratory data on readmission disclosed the following values (the normal reference range is given in parentheses): leukocyte count, 12,880/mm^3; serum amylase, 3,966 IU/l (33–120 IU/l); serum lipase, 3,651 IU/l (13–49 IU/l); serum aspirate aminotransferase, 92 IU/l (13–30 IU/l); alanine aminotransferase, 90 IU/l (10–40 IU/l); serum alkaline phosphatase, 474 IU/l (100–

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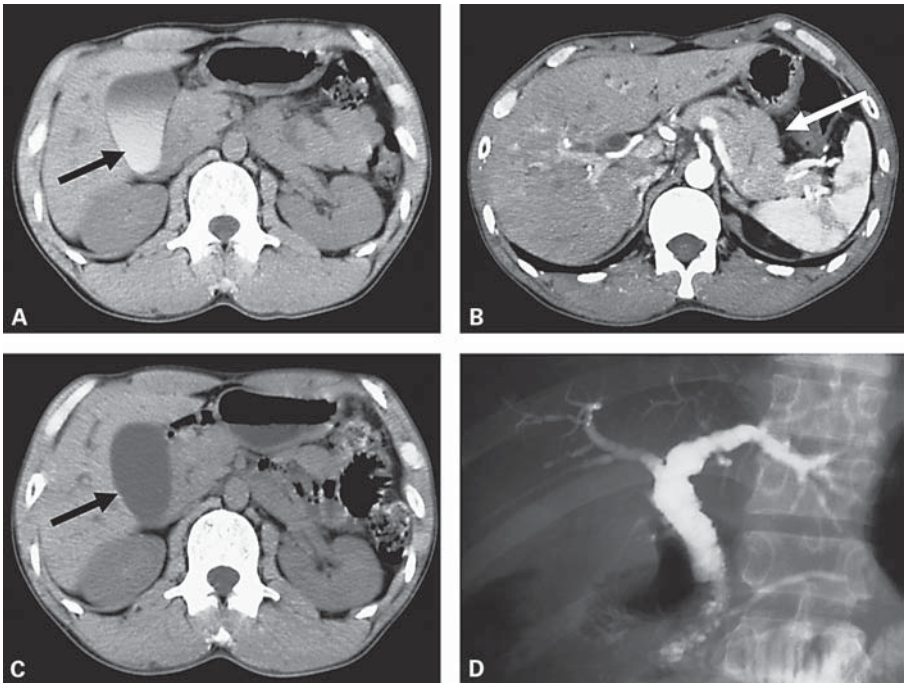

Fig. 1. A Plain CT scan showing limy bile in the gallbladder (black arrow). **B** Contrast-enhanced CT scan showing swollen pancreas with homogeneous parenchyma (white arrow). **C** Plain CT scan showing complete disappearance of limy bile (black arrow). **D** Endoscopic retrograde cholangiogram showing biliary strictures and diverticulum-like outpouching. No gallstones were detected in the choledochus.

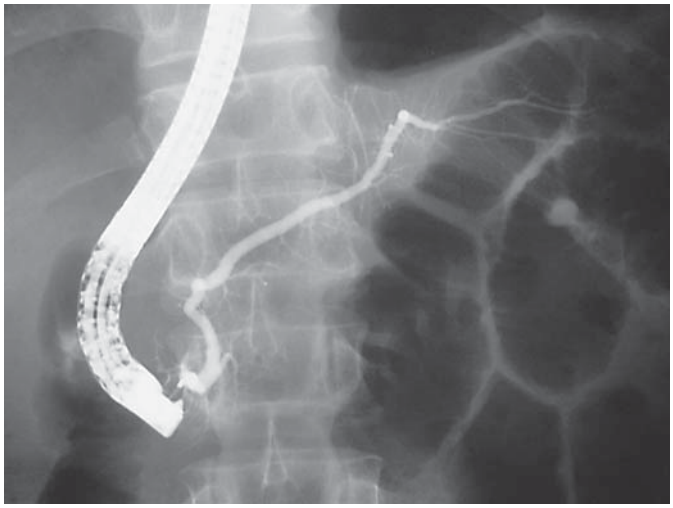

Fig. 2. Endoscopic retrograde pancreatogram showing no morphological abnormalities of the pancreatic duct.

320 IU/l); serum γ-glutamyl transpeptidase, 218 IU/l (16–73 IU/l); serum triglyceride, 58 mg/dl (40–150 mg/dl); and serum calcium 8.7 mg/dl (8.5–10.5 mg/dl). Computed tomography on readmission showed LB (fig. 1A) and a swollen pancreas with homogeneous density of the parenchyma (fig. 1B). This was the first appearance of LB in this case. Symptoms of AP and LB disappeared spontaneously with conservative management (fig. 1C). After recovery from AP (18 days after the last AP episode), endoscopic retrograde cholangiopancreatogram showed biliary strictures and diverticulum-like outpouching. No gallstones were detected in the choledochus (fig. 1D) and no morphological changes of the pancreatic duct were recognized (fig. 2). A rapid decrease of serum pancreatic enzymes (fig. 3) and LB suggested that the AP was of biliary origin, in other words, caused by transient mechanical blockage of the ampulla of Vater. Therefore, 25 days after the last AP episode, endoscopic sphincterotomy was performed to prevent ampullary mechanical blockage as a possible cause of recurrent AP. In consideration of a possible association with ulcerative colitis, total colonoscopy was performed, which revealed no definite evidence of ulcerative colitis but findings of apthoid colitis. The patient has remained well at 4-year follow-up, with no further episodes of AP.

Discussion

The pathogenesis of biliary pancreatitis has not yet been fully clarified. However, Opie's 'common channel theory', which supposed that AP is due to reflux of bile into the pancreatic duct, has been supported for a long time. A report that gallstones were found in the feces of 94% of patients with AP [6] supports Opie's theory. Moreover, a prospective study showed biliary sludge to be an underestimated cause of idiopathic AP [5].

In this case, gallstones were not detected, but LB appearance was recognized. LB is a rare condition caused