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Idiopathic fibrosing pancreatitis associated with ulcerative colitis

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Abstract
Idiopathic fibrosing pancreatitis has been associated with Sjögren's syndrome, primary biliary cirrhosis and primary sclerosing cholangitis. This condition frequently develops in childhood and youth, and has also been related to ulcerative colitis and pericholangitis. Pancreatic complications have been rarely described as systemic complications of ulcerative colitis. A 25-year-old man presented with epigastric pain and jaundice. Abdominal ultrasonography, computed tomography (CT), magnetic resonance cholangiopancreatography (MRCP) and endoscopic retrograde cholangiopancreatography (ERCP) revealed a diffuse enlargement of the pancreas, filiform distal stenosis of the common bile duct and intrahepatic bile ducts, and pancreatic duct dilatation. At operation, a rock-hard and nodular pancreas was noted. Cholecystectomy and Roux-en-Y hepaticojejunostomy, with an access loop, was successfully performed. Idiopathic fibrosing pancreatitis should be considered in young patients with obstructive jaundice, especially those affected with chronic inflammatory or autoimmune diseases. Glucocorticoid therapy would be the first-line treatment, although many patients require operation.

Key Words: *Idiopathic fibrosing pancreatitis, autoimmune pancreatitis, chronic pancreatitis, ulcerative colitis*

Introduction

Extra-colonic complications of ulcerative colitis can involve many organs, including liver, biliary tract, joints, eyes and skin. We present a case of obstructive jaundice secondary to a fibrosing pancreatitis associated with ulcerative colitis; complete symptomatic recovery followed surgical treatment.

Case report

A 25-year-old man was admitted with epigastric pain and jaundice. He had a 4-year history of ulcerative colitis demonstrated by colonic biopsy. Acute attacks of colitis had been treated with 5-aminosalicylate. There was no family history of pancreatic disease, hepatic disorders or inflammatory intestinal disease. Laboratory data revealed a total bilirubin of 195 μmol/L (normal <18), an alkaline phosphatase of 1010 U/L (normal <129), a gamma-glutamyl transpeptidase of 214 U/L (normal <61), SGOT of 184 U/L (normal <37) and SGPT of 502 U/L (normal <40). Hepatitis serology was negative. IgG levels were normal.

Ultrasonography revealed dilatation of the main bile duct and pancreatic duct. An ill-defined nodule was seen at the pancreatic head. Abdominal CT revealed intrahepatic and extrahepatic dilatation of the upper biliary tract, plus pancreatic duct dilatation and a homogeneous enlargement of the head of pancreas (Figure 1). The patient underwent endoscopic retrograde cholangiopancreatography (ERCP) (Figure 2) and magnetic resonance cholangiopancreatography (MRCP) (Figure 3), which revealed a filiform distal

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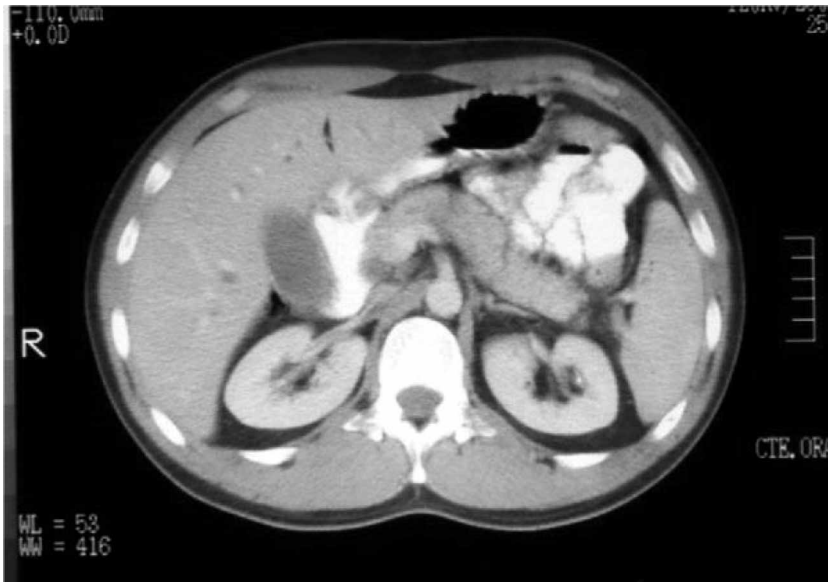

Figure 1. Abdominal CT. Intrahepatic and extrahepatic dilatation of the upper biliary tract. Pancreatic duct dilatation and homogeneous enlargement of the head of the pancreas are shown. No gallbladder stones were observed.

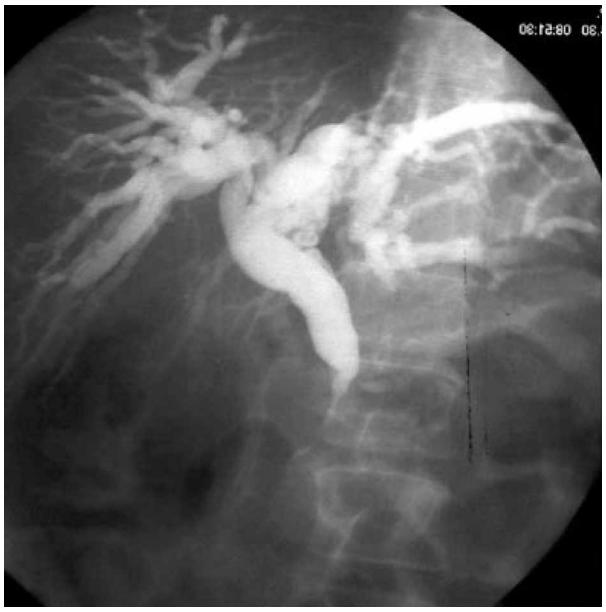

Figure 2. Endoscopic retrograde cholangiopancreatography (ERCP). Filiform distal stenosis of the common bile duct. There

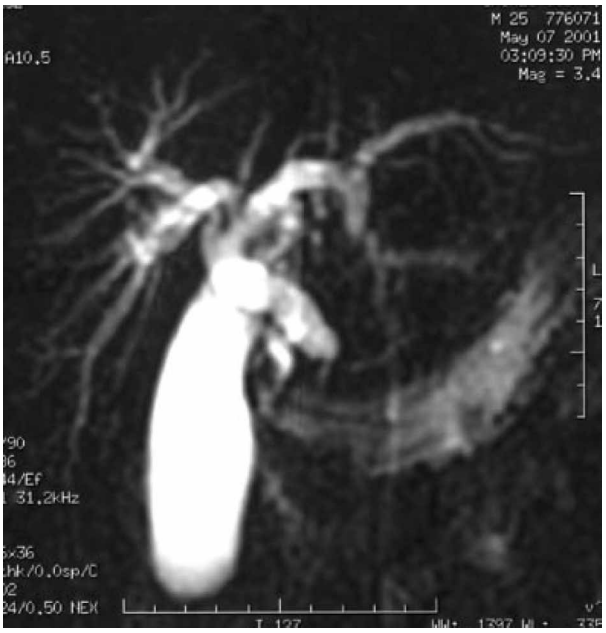

Figure 3. Magnetic resonance cholangiopancreatectomy (MRCP) showing distended gallbladder. There is intrahepatic and extrahepatic dilatation and distal stenosis of the common bile duct.

stenosis of the common bile duct with intrahepatic and extrahepatic biliary tract dilatation.

Preoperative diagnoses were a focal inflammatory process in the pancreas or sclerosing cholangitis. At laparotomy, a rock-hard and nodular pancreas was noted. Fine-needle aspiration biopsy of the pancreatic head was combined with peripancreatic node biopsy, bile cytology and choledochal brushings. Cholecystectomy and Roux-en-Y hepaticojejunostomy with an access loop were performed.

The histopathological report revealed a chronic pancreatitis with lymphoplasmacytic infiltration; the bile cytology and choledochal brushings were negative for malignant cells.

The patient is well without signs of recurrence 2 years postoperatively.

Discussion

Sarles et al. described the concept of an autoimmune aetiology in certain cases of chronic pancreatitis in 1961 [1]. In 1995, Yoshida et al. proposed that diseases with the following characteristics be referred to as'autoimmune pancreatitis': (1) increased serum gammaglobulin or IgG levels; (2) presence of auto-antibodies; (3) diffuse enlargement of the pancreas; (4) diffuse irregular narrowing of the main pancreatic duct on

ERCP; (5) fibrotic change with histopatho-logic lymphocytic infiltration; (6) absence of symptoms or only mild symptoms, usually with an absence of acute attacks of pancreatitis; (7) constriction of the common bile duct in the pancreas with proximal dilatation, and frequent cholestasis and hyperbilirubinaemia; (8) no pancreatic calcification; (9) no pancreatic cysts; (10) occasional association with other autoimmune diseases; and (11) effectiveness of steroid therapy [2]. This condition has also been named sclerosing pancreatitis and idiopathic fibrosing pancreatitis [3,4]. Autoimmune pancreatitis has been described in association with Sjögren syndrome, Crohn's disease and ulcerative colitis [5]. Its incidence in Japan is 5.93 patients per 100 000 inhabitants per year, with a prevalence of 4.78 patients per 100 000 inhabitants [6]. Sjögren syndrome was associated with 25% of the cases of autoimmune pancreatitis in a review of 40 institutions in Japan [7]. The second most common associated disease was primary sclerosing cholangitis (13%), followed by systemic lupus erithematodes.

In line with the Japanese criteria for the diagnosis of autoimmune pancreatitis, our patient had obstructive jaundice secondary to fibrosing pancreatitis, with a probable autoimmune aetiology associated with ulcerative colitis. In some patients with Crohn's disease and ulcerative colitis, this condition has been associated with antibodies against pancreatic juice [8]. Other authors have found high serum IgG4 concentrations, suggesting an autoimmune pathogenesis [9]. Glucocorticoid therapy induces clinical remission and would be the first-line treatment [6,9], although many patients require subsequent operation [3].

Autoimmune pancreatitis should be considered in young patients with obstructive jaundice [10], especially those affected with chronic inflammatory or autoimmune diseases [11].

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Variation of a variation: Case report of attenuated familial adenomatous polyposis

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Abstract

Background. First described in 1988, attenuated familial adenomatous polyposis (AFAP) is a rare autosomal dominant precancerous condition of the gastrointestinal tract. Few reports have described adenocarcinomatous change in the

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