

Obstructive Jaundice and Acute Cholangitis Due to Papillary Stenosis

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Abstract

Papillary stenosis is characterized by fixed fibrosis leading to structural outflow obstruction and it is usually secondary to inflammation and fibrosis from the chronic passage of gallstones, episodes of acute pancreatitis, chronic pancreatitis, sclerosing cholangitis, peptic ulcer disease, and cholesterosis. However, obstructive jaundice with or without acute cholangitis which leads the physician to suspect the presence of malignancy as a cause is a rare manifestation of papillary stenosis. We report here a case of papillary stenosis presenting with obstructive jaundice and acute cholangitis. The lesion was so difficult to exclude the presence of malignancy preoperatively and intraoperatively that a pylorus-preserving pancreaticoduodenectomy was performed. Histologic examination of the resected specimen revealed fibrosis, adenomatoid ductal hyperplasia, and mild chronic inflammation of the papilla of Vater and distal common bile duct.

Key Words: Papillary stenosis, obstructive jaundice, pancreaticoduodenectomy

INTRODUCTION

Papillary stenosis is characterized by fixed fibrosis leading to structural outflow obstruction.¹ According to the Geenen-Hogan classification, type I patients have biliary-type pain, abnormal liver tests on two or more occasions, delayed drainage of ERCP contrast, and a dilated bile duct greater than 12 mm.¹ The etiology of this group of patients is presumed to be papillary stenosis.² The term stenosing papillitis also refers to an anatomic deformity of the papilla of Vater that is characterized by narrowing of the lower end of the bile duct and the proximal end of the duct of Wirsung.³ The defect is said to be secondary to inflammation and fibrosis from the chronic passage of gallstones, episodes of acute pancreatitis, chronic pancreatitis, sclerosing cholangitis, peptic ulcer disease, and cholesterosis.³ According to Moody et al's review,³ the typical patient with stenosing papillitis is a middle-aged female presenting with recurrent epi-

sodes of severe upper abdominal pain several years after cholecystectomy. Their experience revealed that only about 20% of patients with stenosing papillitis had an elevation of liver or pancreatic enzymes associated with their painful episodes and jaundice never occurred in the absence of gallstones, pancreatitis, or neoplasm.³

We report here a case of papillary stenosis presenting with obstructive jaundice and acute cholangitis which are unusual manifestations of papillary stenosis. The lesion was so difficult to exclude the presence of malignancy preoperatively and intraoperatively that a pylorus-preserving pancreaticoduodenectomy was performed. Histologic examination of the resected specimen revealed fibrosis, adenomatoid ductal hyperplasia, and mild chronic inflammation of the papilla of Vater and distal common bile duct (CBD).

CASE REPORT

A 61-year-old man was admitted to the hospital on October 16th, 1994 due to epigastric pain for one day. His past medical history was unremarkable except for tuberculous pleurisy about 40 years previously. On admission, the patient appeared acutely ill and was febrile (39.0°C). The sclerae were slightly icteric and examination of the abdomen revealed direct tenderness on the right upper quadrant without

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organomegaly. Remarkable laboratory test results were as follows: the white blood cell counts 15,400/mm³ (polymorphs 83%), total bilirubin 4.9 mg/dl, alkaline phosphatase 543 IU/L, aspartic transaminase 543 IU/L, and alanine transaminase 391 IU/L. Serum CEA and CA19-9 were within the normal range.

An abdominal ultrasonography and a subsequent computed tomography (CT) revealed dilated intra- and extrahepatic bile ducts and multiple small calcified densities in the gallbladder. However, no mass or lymphadenopathy was delineated around the peripapillary region. An endoscopic retrograde cholangiopancreatography (ERCP) was tried but aban-

doned due to excessive nausea and retching of the patient. A subsequent percutaneous transhepatic cholangiogram showed complete obstruction at the distal CBD (Fig. 1). A percutaneous transhepatic biliary drainage (PTBD) was performed. Bacterial culture of the bile juice grew *Xanthomonas maltophilia*. On the 8th hospital day, an ERCP was tried again and showed a normal papilla and pancreatic ducts, but

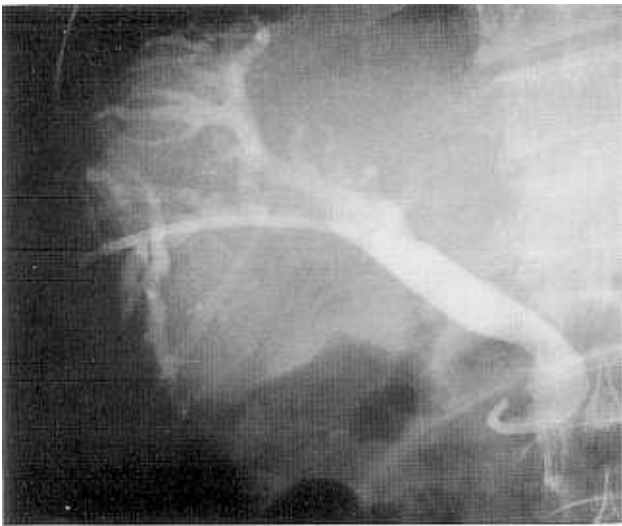


Fig. 1. A cholangiogram through a percutaneous transhepatic biliary drainage tube shows a complete obstruction at the distal common bile duct.

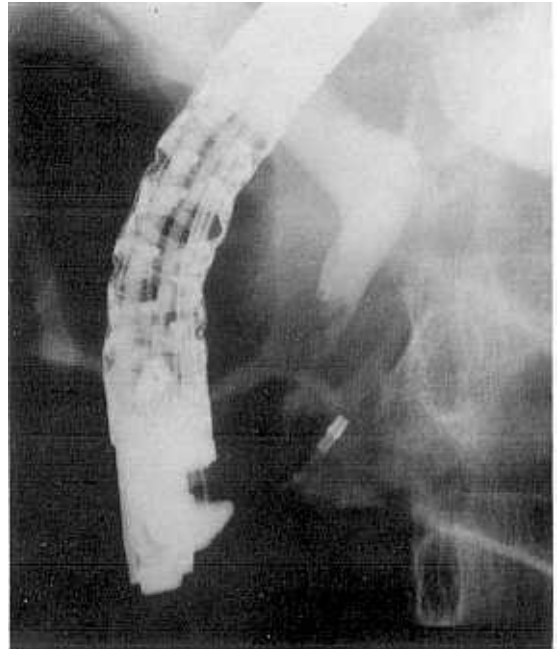


Fig. 2. A simultaneous cholangiogram via a percutaneous transhepatic biliary drainage tube with a cannula for endoscopic retrograde cholangiopancreatography placed at the ampulla of Vater shows a 13 mm long segment of obstruction and a suspicious irregular shaped filling defect proximal to the stenosis.

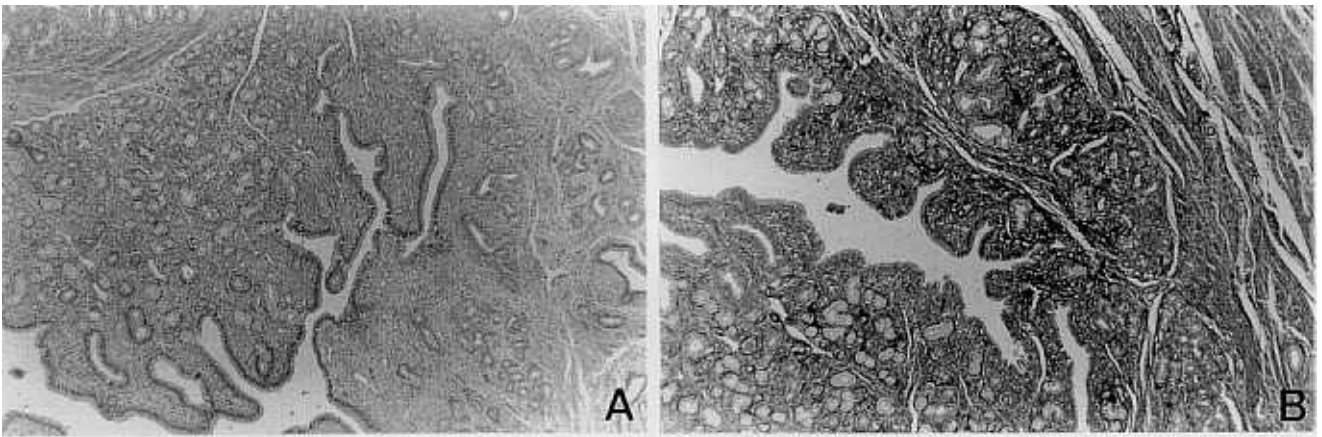


Fig. 3. Histologic examination of the stenotic portion of the papilla and distal common bile duct shows fibrosis, adenomatoid ductal hyperplasia, and mild chronic inflammation (A, H & E, $\times 40$; B, Masson's trichrome, $\times 100$).

failed to cannulate the CBD. A simultaneous cholangiogram via a PTBD tube with an ERCP cannula placed on the opening of the ampulla of Vater showed a 13 mm-long segment of obstruction and a suspicious irregular-shaped filling defect proximal to the stenosis (Fig. 2).

On the 17th hospital day, an exploratory laparotomy was carried out. A 2.5 × 2.0 cm sized firm mass was palpated around the papilla without enlarged lymph nodes. Although an intraoperative biopsy of the lesion revealed only reactive hyperplasia, the surgeon performed a pylorus-preserving pancreaticoduodenectomy.

Grossly, no mass was found at the papilla and distal CBD and no stones were found in the gallbladder. However, the papilla and distal CBD were stenotic. Microscopic examination of the stenotic portion revealed fibrosis, adenomatoid ductal hyperplasia, and mild chronic inflammation of the papilla and distal CBD without evidence of malignancy (Fig. 3).

The patient recovered uneventfully and has been doing well up to now (June, 1998).

DISCUSSION

Although this case deserves the diagnosis of papillary stenosis or stenosing papillitis, clinical manifestation is quite unusual in that jaundice and acute cholangitis occurred due to complete obstruction of the papilla of Vater. Papillary stenosis in the present case might have been attributed to chronic passage of gallstones. However, since initially found echogenic densities by ultrasonography and calcified densities by CT were not found in the removed gallbladder, it is possible to conclude that these small stones or sludges were developed due to obstruction and removed by PTBD.

We found similar cases to ours in the literature. In 1970, Nardi reported a case of acute suppurative cholangitis due to ampullary fibrosis which was asserted to be the first report on acute cholangitis as a complication of papillary stenosis.⁴ This patient was explored on an emergency basis without cholangiographic and CT examinations and underwent sphincteroplasty. Biopsy of the sphincter of Oddi demonstrated fibrosis with chronic inflammation. One of the two cases (a 37-year-old female) that appeared in the Spanish literature under the term benign idiopathic stenosis of the choledochus involved the papilla of Vater and underwent a pancreaticodu-

denectomy because an obstructing lesion at the distal CBD was considered malignant.⁵ In the Korean literature,⁶ a similar case was found under the term of adenofibromatous hyperplasia of the papilla of Vater, which refers to glandular proliferation and increased fibrosis. We do not think this is a totally different entity from papillary stenosis. The degree of glandular proliferation and fibrosis may vary according to the stage of the disease process. This 42-year-old male patient also underwent a pancreaticoduodenectomy due to the difficulty of excluding malignancy.

If this kind of lesion is confirmed before operation, we can simply treat the patients by using various endoscopic approaches such as endoscopic sphincterotomy, balloon dilatation, or temporary placement of a stent or less invasive surgical approach. However, despite the advent of techniques for diagnosing pancreaticobiliary diseases, it does not seem to be easy to confirm that an obstructing lesion is benign. It is not likely that negative study with endoscopic ultrasonography excludes malignancy. Negative histologic examination of the biopsied specimen which might be obtained by endoscopic sphincterotomy or cholangioscopy would not preclude exploration in appropriate patients.⁷ Furthermore, intraoperative findings are suggestive but nonspecific and in some cases, completely inaccurate.⁷ In the present case, despite negative histologic examination of the intraoperative frozen biopsy, the surgeon performed a pylorus-preserving pancreaticoduodenectomy because the lesion was palpated like a mass.

The incidence of malignancy among pancreaticoduodenectomies for suspected but unproven malignancy was reported to be about 45% (9 out of 20 patients),⁸ which is not low enough to risk losing the option of potentially curative surgery if the patient has a malignancy. Fortunately, the morbidity and mortality of the pancreaticoduodenectomy have remarkably decreased in experienced hands and several reports support performing this procedure in benign conditions. These data justify a continued aggressive approach to suspected pancreatic and periampullary malignancy.⁷⁻¹⁰

In conclusion, it should be emphasized that a short segment stricture at or near the papilla of Vater causing obstructive jaundice and/or acute cholangitis, even without a history of gallstone disease, pancreatitis, sclerosing cholangitis or peptic ulcer disease might be due to benign papillary stenosis, which can be managed by less invasive treatment options.

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