

Inflammatory Bile Duct Obstruction during the Healing Stage of Severe Acute Pancreatitis

Akane Yamabe, Atsushi Irisawa, Goro Shibukawa, Ai Sato, Mariko Fujisawa, Noriyuki Arakawa, Yoshitsugu Yoshida, Ryo Igarashi, Takumi Maki, Shogo Yamamoto, Tsunehiko Ikeda, Yoko Abe and Koki Hoshi

Abstract

The patient was a 62-year-old woman with a history of severe acute pancreatitis complicated by walled-off necrosis (WON), who developed obstructive jaundice during the recovery phase from WON. Magnetic resonance cholangiopancreatography (MRCP) and endoscopic retrograde cholangiopancreatography (ERCP) revealed the complete obstruction of the distal bile duct, and endoscopic ultrasonography (EUS) revealed thickening of the duct wall, with a uniform distribution, and a relatively well-preserved layered structure. A cytopathological examination using ERCP showed no malignancy. The underlying etiology of this case may have been the formation of a cicatricial stricture during the resolution of WON, in addition to fibrosis caused by the spreading of inflammation from pancreatitis.

Key words: acute pancreatitis, walled-off necrosis, bile duct obstruction

(Intern Med 56: 1037-1040, 2017)

(DOI: 10.2169/internalmedicine.56.7859)

Introduction

When biliary stricture is found, both benign and malignant etiologies must be carefully considered as a variety of types of benign biliary stricture may mimic spreading hilar cholangiocarcinoma (CCA). Although up to 30% of biliary stricture cases can be benign (1), the vast majority are malignant, with the two major malignancies being pancreatic adenocarcinoma and CCA. The final determination of malignancy in biliary strictures can entail major surgery if a preoperative diagnosis of malignancy cannot be made. Several previous studies suggested that approximately 15-24% of patients undergoing surgical resection for suspected biliary malignancy have a benign etiology; however, there are no clinical or radiological features to reliably distinguish benign biliary strictures from malignant ones (2-6). The benign causes of distal bile duct obstruction are often inflammatory processes such as chronic pancreatitis, primary sclerosing cholangitis, autoimmune pancreatitis and immunoglobulin G4-related cholangitis, portal biliopathy, HIV-related cho-

langitis, parasitic infection, or idiopathic benign focal stricture. We sometimes encounter cases of bile ductal stenosis accompanying acute/chronic pancreatitis at an earlier stage of the disease. However, the occurrence of bile stenosis in the healing stage is a rare condition. We herein report a case of distal bile stenosis/obstruction in the healing stage of severe acute pancreatitis complicated by the formation of walled-off necrosis (WON).

Case Report

The patient was a 62-year-old woman who had previously been healthy. She visited hospital "A" for abdominal pain and was diagnosed with severe acute biliary pancreatitis. Treatment was initiated with a pancreatic enzyme inhibitor and antimicrobial agents, and the pancreatitis subsided. However, follow-up computed tomography (CT) revealed WON (Fig. 1). On hospital day 41, the patient was transferred to our hospital for to undergo treatment for WON. The patient's WON was non-infectious, and clinical symptoms, such as abdominal pain and gastrointestinal transit ab-



Figure 1. Follow-up CT after severe pancreatitis revealed huge WON on hospital day 41.

normalities were not present. In addition, the observations during follow-up showed that the WON was not increasing in size. Based on these factors, a conservative medical treatment approach was chosen, in accordance with the Atlanta classification system (7). Following the initiation of conservative treatment, the patient's WON gradually decreased in size (Fig. 2) and the clinical course showed no complications (such as infection or bleeding inside the WON). The patient was discharged on hospital day 137. However, on disease day 146, the patient complained of general malaise and was examined at the outpatient emergency department. A blood analysis revealed elevated hepatobiliary enzyme levels, and bile duct obstruction was suspected. Magnetic resonance cholangiopancreatography (MRCP) revealed a distal bile duct stricture (Fig. 3), and endoscopic ultrasonography (EUS) showed thickening of the entire circumference of the extrahepatic bile duct wall (Fig. 4). Because the wall thickening was uniformly distributed and the layered structure was relatively preserved, the findings were believed to be due to inflammatory changes. Endoscopic retrograde cholangiopancreatography (ERCP) was performed, and a relatively smooth stenosis of the distal bile duct was found, as observed with MRCP. Based on the morphology, it was suspected to be benign bile duct stricture. To treat the stricture, an indwelling plastic stent with a 7 Fr bilateral pigtail catheter was inserted. Bile was collected for a cytological examination, which did not reveal any malignancy. ERCP was repeated 54 days after the placement of the indwelling stent and showed that the stricture had improved (Fig. 5); stent reinsertion was not performed. There has been no recurrence of bile duct stenosis in 18 months of follow-up.

Discussion

With the exception of biliary calculi, malignant tumors are the most common cause of bile duct obstruction associated with obstructive jaundice; obstruction due to benign disease is rare. There have been only 2 reported cases involving the development of extrapancreatic bile duct stenosis during the recovery process after acute pancreatitis; these

were reported in 1995 by Delcenserie et al. (8). In both cases, jaundice developed following the resolution of effusion in the lesser sac with acute pancreatitis. Both patients were treated with hepatocholangiojejunostomy. The histological examination of the resected specimens revealed marked fibrosis at the sites of the bile duct stricture. In both cases, bile duct stricture was estimated to have been caused by inflammation of the bile duct serosa due to pancreatic enzymes. In fact, even in acute pancreatitis, bile duct stricture or obstruction can sometimes occur due to temporary edematous changes in the pancreas or the spreading of inflammation to the distal bile duct. In most cases, bile duct stricture or obstruction develops within a reasonably short period of time after the occurrence of acute pancreatitis. However, in the present case, extrahepatic bile duct obstruction occurred approximately 140 days after the onset of acute pancreatitis, during the so-called "healing phase." Thus, edema and the spreading of inflammation alone were unlikely to have caused the stricture; instead, the major causal factor may have been the occurrence of a cicatricial stricture in association with resolution of the WON, in addition to fibrosis due to the spreading of inflammation caused by acute pancreatitis.

In the presence of a benign bile duct stricture, malignancies need to be ruled out, and the diagnosis should be made based on comprehensive consideration of imaging studies and pathology findings. Benign strictures are commonly characterized by a continuous transition from the dilated portion of the bile duct to the site of the stricture, which typically has smooth margins and left-right symmetry. In addition to imaging tests, cytological examinations are widely used to determine whether the condition is benign or malignant. Although most reports have shown a specificity of nearly 100% (9), the negative predictive value is low, and malignancies cannot be completely ruled out in patients with negative cytology results. Thus, in some cases, surgical resection is eventually carried out when a malignant tumor is suspected on the basis of diagnostic imaging results, even when the cytology and biopsy results are negative. In the present case, the cytology was negative, and few of the imaging findings were suggestive of malignancy. In addition, WON was not complicated by infection and showed a tendency to decrease in size over time. Thus, immediate biliary drainage was performed with the placement of an indwelling plastic stent in order to help prevent an exacerbation of the fibrotic stricture. In our case, no signs or symptoms of recurrence were seen in the long-term follow-up after stent withdrawal. The reason for this is considered to be due to the fact that the placed plastic stent may have acted as a template, and the dilation of the stenotic bile duct could thus be maintained.

The differential diagnosis of bile duct stricture can be extremely difficult to determine at times. For this reason, the decision about the best method of treatment for each case should be made following a careful investigation and with sufficient consideration of all known factors. Although ex-

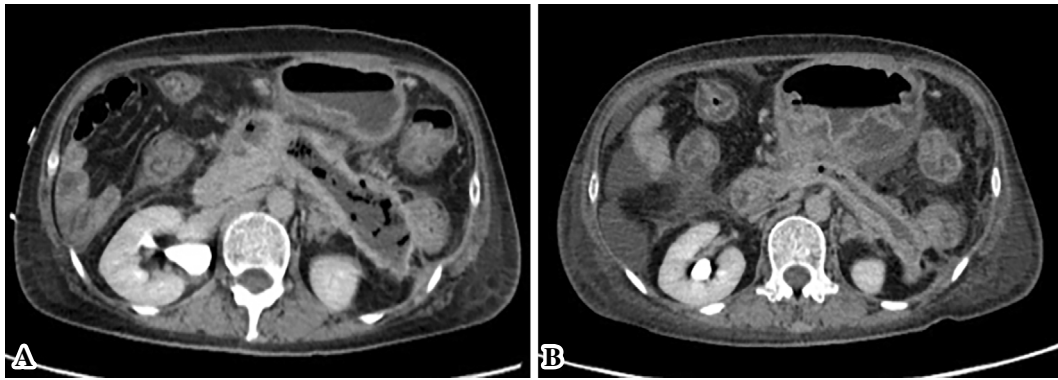


Figure 2. WON gradually decreased in size during recovery stage without intervention. A: Follow up CT in the hospital day 87. Follow up CT in the hospital day 130.

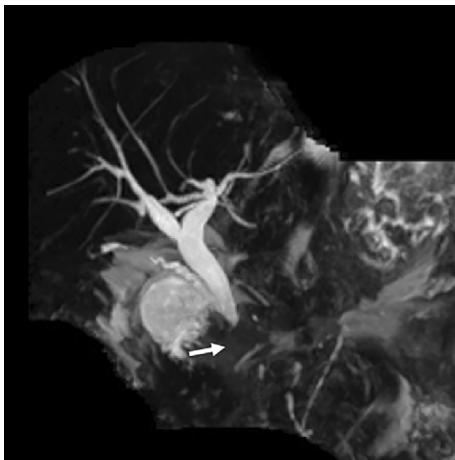


Figure 3. MRCP on disease day 146 revealed a distal bile duct stricture (arrow).



Figure 4. EUS showed thickening of the entire circumference of the extrahepatic bile duct wall (arrow).

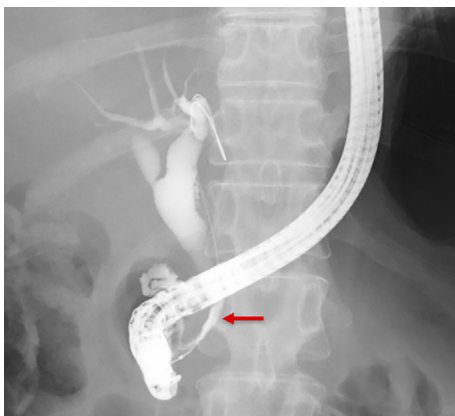


Figure 5. ERCP showed an improvement of distal bile duct stricture (arrow).

tremely rare, bile duct stricture like the one found in the case described in this report may likely occur, even during the healing process of acute pancreatitis. We therefore believe that bile duct stricture should be recognized as a late complication of acute pancreatitis.

The authors state that they have no Conflict of Interest (COI).

References

1. Tummala P, Munigala S, Eloubeidi MA, et al. Patients with obstructive jaundice and biliary stricture ± mass lesion on imaging: prevalence of malignancy and potential role of EUS/FNA. *J Clin Gastroenterol* **47**: 532-537, 2013.
2. Hayat JO, Loew CJ, Asress KN, et al. Contrasting liver function test patterns in obstructive jaundice due to biliary strictures and stones. *QJM* **98**: 35-40, 2005.
3. Clayton RAE, Clarke DL, Currie EJ, et al. Incidence of benign pathology in patients undergoing hepatic resection for suspected malignancy. *Surgeon* **1**: 32-38, 2003.
4. Gerhards MF, Vos P, Van Gulik TM, et al. Incidence of benign lesions in patients resected for suspicious hilar obstruction. *Br J Surg* **88**: 48-51, 2001.
5. Corvera CU, Blumgart LH, Darvishian F, et al. Clinical and pathologic features of proximal biliary strictures masquerading as hilar cholangiocarcinoma. *J Am Coll Surg* **201**: 862-869, 2005.
6. Wakai T, Shirai Y, Sakata J, et al. Clinicopathological features of benign biliary strictures masquerading as biliary malignancy. *Am Surg* **78**: 1388-1391, 2012.
7. Banks PA, Bollen TL, Dervenis C, et al; Acute Pancreatitis Classification Working Group. Classification of Acute Pancreatitis-2102: revision of the Atlanta classification and definitions by international consensus. *Gut* **62**: 102-111, 2013.
8. Delcenserie R, Joly JP, Lenne C, et al. Fibrotic stricture of the extrahepatic biliary tract: a new complication of acute pancreatitis. Two cases. *Pancreas* **10**: 100-103, 1995.
9. Volmar KE, Vollmer RT, Routbort MJ, Creager AJ. Pancreatic and

bile duct brushing cytology in 1000 cases: review of findings and comparison of preparation methods. *Cancer* **108**: 231-238, 2006.

The Internal Medicine is an Open Access article distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (<https://creativecommons.org/licenses/by-nc-nd/4.0/>).

© 2017 The Japanese Society of Internal Medicine
<http://www.naika.or.jp/imonline/index.html>