Formosan Journal of Surgery (2015) 48, 68–71

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CASE REPORT

Unusual presentation of a low-grade intraductal papillary neoplasm of the bile duct

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Received 10 July 2014; received in revised form 29 September 2014; accepted 19 November 2014
Available online 31 March 2015

KEYWORDS
cholangitis;
hemobilia;
intraductal bile duct neoplasm

Summary Intraductal papillary neoplasm of the bile duct usually manifests as abdominal pain, jaundice, and cholangitis but rarely presents as hemobilia. In this article, we present the case of a 74-year-old man with a 2-year history of repeated hemobilia without a definite diagnosis. A cholangiogram revealed a left intrahepatic duct dilatation with a filling defect, and computed tomography revealed a hyperdense lesion in the left lateral liver segment, which was subsequently resected. Histopathological examination revealed focal low-grade intraductal papillary neoplasm of the bile duct and markedly dilated bile ducts with a ruptured blood vessel. The postoperative course was uneventful. Clinicians should comprehensively evaluate cases of unusual and recurrent gastrointestinal bleeding by considering this diagnosis.

1. Introduction

Intraductal papillary neoplasm of the bile duct (IPNB) has been defined as a "biliary epithelial tumor with exophytic nature exhibiting papillary mass within the bile duct lumen and with prominent intraductal growth pattern"[1]. The most

Conflicts of interest: The authors do not have any conflicts of interest.

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http://dx.doi.org/10.1016/j.fjs.2014.11.003

common manifestations are abdominal pain, jaundice, and cholangitis; however, hemobilia has occasionally been observed.[2,3] Definitive clinical and radiological diagnosis is difficult in IPNB. Because malignant components are present in over half of all IPNB cases, a complete surgical resection is typically necessary.[2,3] In this article, we report the surgical treatment of an IPNB presenting as recurrent hemobilia.

2. Case report

A 74-year-old man with a 2-year history of recurrent upper gastrointestinal (GI) bleeding and cholangitis presented with epigastric pain, jaundice, and tarry stool. On examination, he had icteric sclera, pale conjunctiva, and local epigastric tenderness. Laboratory tests revealed that the serum bilirubin and hemoglobin levels were 3 mg/dL and 10.2 g/dL, respectively. An upper GI endoscopy revealed a large blood clot in the second portion of the duodenum near the papilla of Vater. Angiography did not show any vascular lesion from the branches of the celiac trunk and superior mesenteric artery. Abdominal ultrasonography and contrast-enhanced computed tomography (CT) revealed bilateral intrahepatic biliary tree dilatation with no intrahepatic lesion. Endoscopic retrograde cholangiopancreatography revealed bilateral intrahepatic biliary tree dilatation with filling defects in the distal common bile duct (CBD); although a few stone fragments were extracted, the filling defects remained. Choledochoscopy revealed minimal blood clotting in the CBD and some blood flow from the left intrahepatic duct; a T-tube was placed in the CBD after removing the clot, but after 1 month, a filling defect was observed again in the left intrahepatic duct (Fig. 1). Contrast-enhanced CT revealed a hyperdense lesion in the left lateral liver segment (Fig. 2),

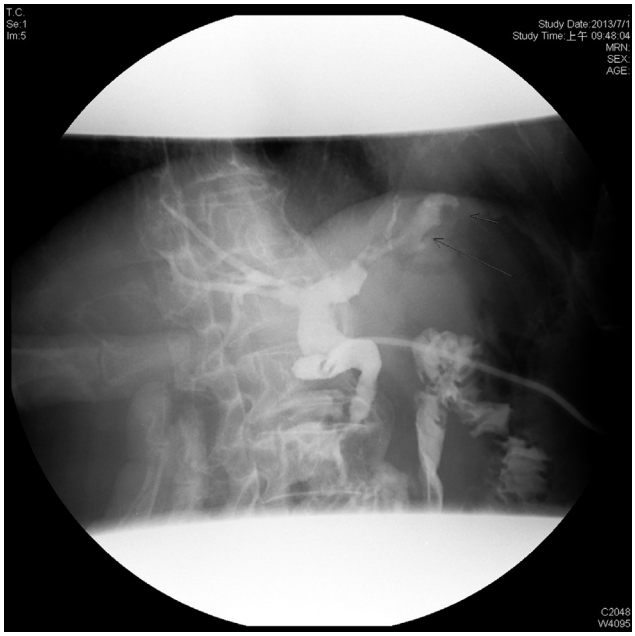

Figure 1 Cholangiogram demonstrating the biliary tract dilatation and the left intrahepatic duct-filling defect.

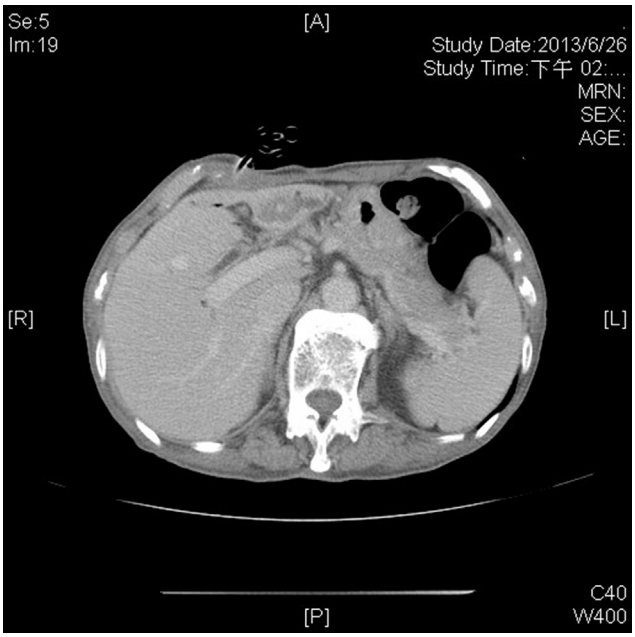

Figure 2 Contrast-enhanced computed tomography revealed a hyperdense lesion in the left lateral liver segment.

which was subsequently resected. Grossly, intrahepatic bile ducts were markedly dilated and a papillary tumor measuring 1.0 cm × 0.9 cm × 0.8 cm was observed along with an atrophic and fibrotic appearance of periductal liver parenchyma (Fig. 3). Microscopically, a focal low-grade IPNB (Fig. 4A) was evident along with markedly dilated bile ducts, granulation tissue, and vessel rupture (Fig. 4B). The histopathological diagnosis was pancreaticobiliary-type IPNB (Fig. 4C). The postoperative course was uneventful, and the patient was discharged on postoperative Day 7; no recurrence was observed at the 1-year follow-up examination.

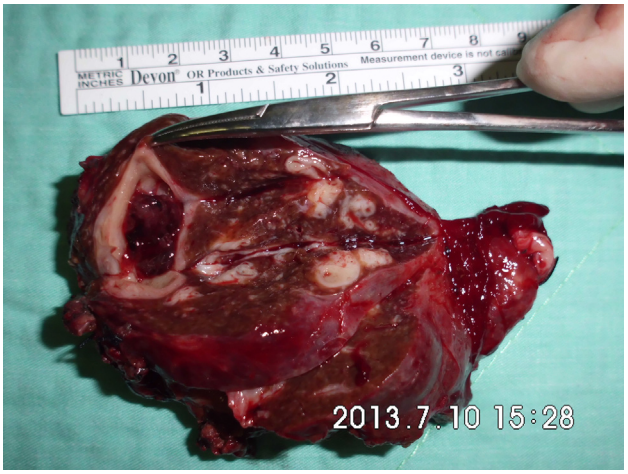

Figure 3 Left lateral liver segment. Dilatation of the intrahepatic bile ducts with a papillary tumor.