

Case Report

Adenomyoma of the Common Bile Duct

— Report of a Case —

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**Abstract:** Adenomyomas in the extrahepatic bile duct are extremely rare. In a 75-year-old male with acute cholangitis due to adenomyoma forming a protruding lesion in the terminal bile duct, pancreatoduodenectomy was carried out, resulting complete cure.

**Key words:** Adenomyoma of the common bile duct, Early bile duct cancer, Obstructive jaundice

INTRODUCTION

Adenomyoma in the biliary ductal system is most frequently found in the gallbladder. The gallbladder wall is more abundant in muscle fibers than is the wall of the bile duct. Adenomyoma in the gallbladder is known to be closely related to the formation of gallstones. In other parts of the biliary ductal system, however, adenomyoma is very rare<sup>1),2)</sup>, although a few cases of a tumor arising from the papilla of Vater<sup>3)</sup> have been reported.

We encountered a patient with severe acute cholangitis with a 10 mm diameter shadow defect on the wall of the terminal bile duct demonstrated by percutaneous transhepatic cholangiography (PTC) and endoscopic retrograde cholangiopancreatography (ERCP). As early cancer of the bile duct was suspected from the radiologic findings, pancreatoduodenectomy was per-

formed, and from the histologic examination of the surgical specimen, adenomyoma of the common bile duct was confirmed.

Although benign neoplasma of the bile duct system are uncommon, the tumors are clinically very important because they can cause obstructive jaundice<sup>4)</sup> and require differentiation from early cancer of the bile duct.

We report here the surgical results and pathologic findings in a case of adenomyoma of the terminal bile duct.

CASE

On December 10, 1983, a 75-year-old man had a sudden onset of severe right hypochondralgia, high fever of 40°C and chills after supper. Medication was provided and these symptoms subsided within two days, and subsequently, he was admitted to Hospital of Yamanashi Medical College. He was 146 cm tall, weighed 47 kg and had no past history of abdominal pain or jaundice.

*Laboratory findings:* Laboratory findings

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on admission to our hospital were as follows: T. Bili. was 0.6 mg/dl, D. Bili. 0.2 mg/dl, Alp 228 mIU, LAP 54 mIU, r-GTP 157 mIU, GOT 29 mIU, GPT 29 mIU, LDH 301 mIU, s-Amylase 150 U (Somogyi), RBC  $431 \times 10$ . Slight increase in Alp and r-GTP levels were observed.

*Cholangiography:* Drip infusion cholangiography (DIC) disclosed a slightly dilated extrahepatic bile duct and a shadow defect in the terminal region (Fig. 1). ERCP showed no evidence of tumor, ulcer or



Fig. 1. DIC. A shadow defect is shown in the terminal bile duct.

bleeding in the papilla of Vater, but revealed a circular protruding lesion in the terminal bile duct. No change was observed in the main pancreatic duct (Fig. 2). PTC showed no dilatation in the intrahepatic bile duct, but a moderately dilated extrahepatic bile duct was demonstrated. The location of this shadow defect was not altered by the changes in the body position, suggesting the presence of a lesion in the biliary duct wall (Fig. 3). In computed tomography (CT), the left lobe of the liver

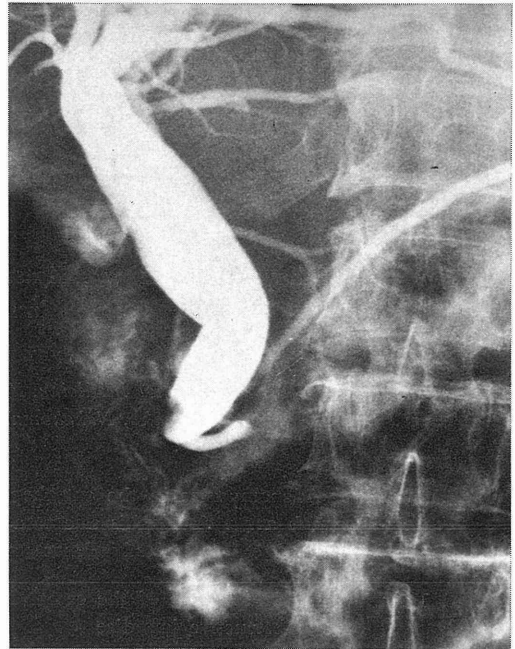


Fig. 2. A 10 mm diameter circular elevated lesion is demonstrated in the terminal bile duct.

was larger than the right lobe, but no abnormal density area was confirmed. Slightly prolonged excretion of the radionuclide into the liver, the intrahepatic bile ducts, the common bile duct, and the duodenum was demonstrated using a  $^{99m}\text{Tc}$ -HIDA hepatobiliary scintigram.

*Operation:* December 4, 1984 laparotomy was performed. Neither ascites nor adhesions were shown in the abdominal cavity. Both the left and the right lobes of the liver were normal in color, surface texture and hardness, with the left lobe larger than the right lobe, as visualized by CT. There were no inflammatory changes in the gallbladder or in the wall of the extrahepatic bile duct. The common bile duct was incised, and an endoscopic examination of the biliary ductal system was performed, revealing a flat semicircular protruding lesion on the wall of the terminal common duct. The surface of the



Fig. 3. PTC. Moderately dilated extrahepatic bile duct and shadow defect in the terminal bile duct are revealed and the location of the lesion did not vary with changes in body position.

lesion was red and granular, but no bleeding was apparent.

Although biopsy failed to show malignancy of the tumor in the terminal bile duct, pancreatoduodenectomy was performed, as we could not exclude early cancer of the bile duct. The postoperative course was uneventful, and he was discharged from the hospital in the fourth postoperative week and has been healthy during the past four years.

*Pathologic findings of the specimen:* The wall of the terminal common bile duct was thickened to 2 mm, and a protruding lesion measuring 10×5×5 mm was present on the wall of the common bile duct 2 mm from the orifice of the ampulla of Vater. Histologically, the elevated lesion of biliary ductal wall had adenomatous features, presenting lobular arrangement of the cana-

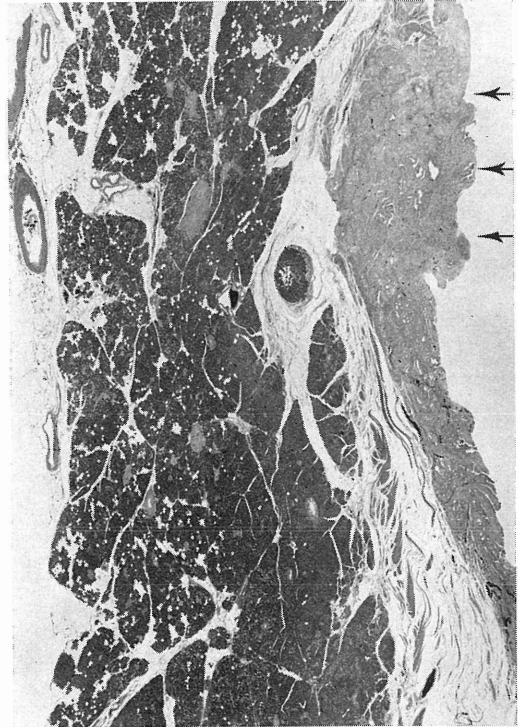


Fig. 4. Marked thickening (arrow) of the mucosa of the terminal bile duct. (H.E. stain, ×20)

liculi. In the interstitium of adenomatous lesion, hyperplastic smooth muscles were sporadically observed around the canaliculi. From these findings, the lesion was diagnosed as adenomyoma or adenomyomatosis (Fig. 4 and 5). Microscopic examination did not show adenomyoma in the ampulla of Vater or in the gallbladder.

#### DISCUSSION

In the literatures, there were 33 cases of epithelial tumors including papillomas, polyps, adenomas and cystadenomas; four cases of lipoma, three cases of xanthomas, one case of myxoma, and five cases of mixed tumors (adenofibroma, adenomyofibroma, adenomyoma and myoadenoma)<sup>1), 2), 5), 6)</sup>. In recent years, owing to the widespread use of ERCP, it has been possible to make a diagnosis before surgery in an increasing

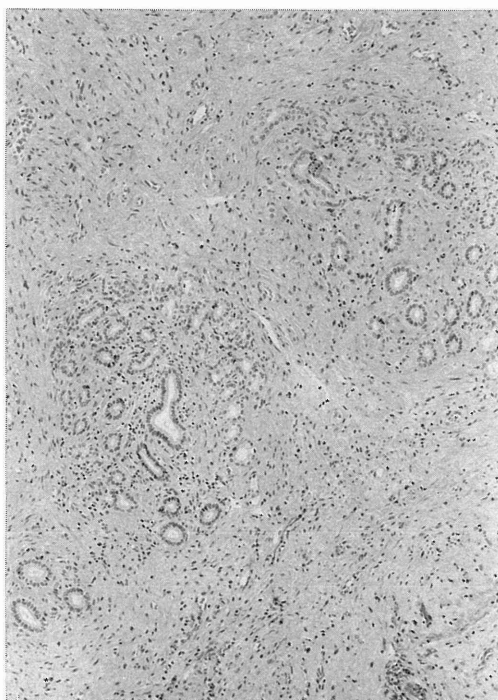


Fig. 5. High power view of Fig. 4. The mucosal thickening consisting of glandular hyperplasia in combination with either muscle or fibrous connective tissue. (H, E, stain,  $\times 100$ )

number of cases<sup>7)</sup>. However, very few cases of adenomyoma arising in the extrahepatic bile duct except for the gallbladder and the papilla of Vater have been reported. According to Dowdy's total<sup>2)</sup>, only one out of 16 cases of adenoma, was an adenomyoma in the left hepatic duct. Subsequently classified adenomas of the biliary duct system into three types<sup>1)</sup>: the solid type, cystic type and mixed type. Adenomyomas were grouped in the mixed type, i.e., "the mixed type includes tumors of glandular elements in combination with either muscle or fibrous connective tissue, and appears as a variously sized, firm and well circumscribed tumor within the wall of the bile duct. Histologically, the tumor consists of abundant muscle or fibrous connective tissue elements in which are

disposed scattered epithelial glandular element, similar in appearance to those found in the solid type."

In cases of adenomyoma, the tumor may occasionally appear as a diffuse thickening of the duct wall. However, the tumor mass which grows gradually in the lumen of the bile duct usually causes abdominal pain and obstructive jaundice, and, as seen in this case, the lesion arising in the portion near the terminal bile duct may cause bacterial infection, presenting attacks of acute cholangitis.

In cases with appearances of these symptoms, the elevated lesion on the wall of the bile duct can be easily detected by direct cholangiography such as ERCP and PTC, but it will be extremely difficult to differentiate the lesion from early bile duct cancer. In particular, it is not easy to confirm before or during surgery that there are no malignant changes in the elevated lesion in the intrapancreatic portion of the bile duct. For these reasons, we believe that when a protruding lesion in the terminal bile duct has been detected, pancreatoduodenectomy must be carried out for successful removal of the tumor.

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