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A Case of Adenomyomatous Hyperplasia of the Extrahepatic Bile Duct

Masakatsu Numata^a Soichiro Morinaga^a
Takuo Watanabe^a Hiroshi Tamagawa^a Naoto Yamamoto^a
Manabu Shiozawa^a Yoichi Kameda^b Shinichi Ohkawa^c
Yasushi Rino^d Makoto Akaike^a Munetaka Masuda^d

Departments of ^aHepatobiliary and Pancreatic Surgery, ^bPathology and
^cHepatobiliary and Pancreatic Medicine, Kanagawa Cancer Center, Yokohama,
and ^dDepartment of Surgery, Yokohama City University, Yokohama, Japan

Key Words

Adenomyomatous hyperplasia · Extrahepatic bile duct · Benign tumor

Abstract

Adenomyomatous hyperplasia is rarely found in the extrahepatic bile duct. A 54-year-old man was referred to our center with a diagnosis of extrahepatic bile duct stenosis which had been detected by endoscopic retrograde cholangiopancreatography. Abdominal computed tomography revealed thickening of the wall of the middle extrahepatic bile duct, however no malignant cells were detected by cytology. Since bile duct carcinoma could not be ruled out, we performed resection of the extrahepatic duct accompanied by lymph node dissection. Histopathologically, the lesion was diagnosed as adenomyomatous hyperplasia of the extrahepatic bile duct. Present and previously reported cases showed the difficulty of making a diagnosis of adenomyomatous hyperplasia of the extrahepatic bile duct preoperatively or intraoperatively. Therefore, when adenomyomatous hyperplasia is suspected, a radical surgical procedure according to malignant disease may be necessary for definitive diagnosis.

Introduction

Adenomyomatous hyperplasia is considered to be a non-neoplastic, tumor-like, inflammatory lesion which commonly develops in the gallbladder. Benign tumors of the extrahepatic bile duct, including adenomyomatous hyperplasia, are rarely found in clinical practice. Therefore, the etiology of adenomyomatous hyperplasia of the

extrahepatic bile duct has not been clarified. Its clinical importance lies in its potential to cause symptomatic biliary tract obstruction or cholangitis and its propensity to be confused with malignant lesions, thereby leading to extensive surgical resection. There are no useful diagnostic methods except histologic examination that can be used to distinguish adenomyomatous hyperplasia from malignant tumor. We present a resected case of symptomatic adenomyomatous hyperplasia of the extrahepatic bile duct and review the published reports of adenomyomatous hyperplasia of the extrahepatic bile duct to discuss the diagnostic and clinical relevance of this case.

Case Report

The patient, a 54-year-old man, was admitted to our hospital with a chief complaint of jaundice. Nothing special was found in physical examination except for icteric change. Laboratory studies revealed aspartate aminotransferase 189 U/l, alanine aminotransferase 779 U/l, alkaline phosphatase 1,766 U/l, total bilirubin 7.9 μ mol/l, carcinoembryonic antigen 0.8 ng/ml, and carbohydrate antigen 19-9 306.2 U/ml. Endoscopic retrograde cholangiopancreatography (ERCP) showed stenosis of the middle bile duct and slight distention of the upper and intrahepatic bile duct above the stenosis ([fig. 1](#)). Abdominal ultrasonography revealed a dilatation of the upper common bile duct to 9 mm in diameter. Computed tomography detected thickening of the wall in the middle hepatic duct. Neither bile cytology nor brush cytology showed any malignant cells (class I and III). Since bile duct cancer could not be ruled out, extrahepatic bile duct resection accompanied by lymph node dissection was performed. Gross observation of the cut surface showed a white-colored, thickening lesion 20 $\times$ 15 mm in size ([fig. 2](#)). Pathological examination demonstrated that multiple hyperplastic glands without cellular atypia were present in the middle bile duct wall, along with proliferation of both epithelial and smooth muscle components and infiltration by inflammatory cells ([fig. 3](#)). The patient has been well without any evidence of recurrence for 3 years since his operation.

Discussion

Benign tumors of the extrahepatic bile duct are rare, and many are malignant [1]. Among benign tumors, adenomyomatous hyperplasia of the extrahepatic duct bile duct is extremely rare, while adenomyomatous hyperplasia is most commonly found in the stomach, gallbladder, duodenum, and jejunum [2]. Microscopically, adenomyomatous hyperplasia is characterized by proliferation of both epithelial and smooth muscle components, and involvement may extend to the serosa [3]. The surrounding glands usually are dilated cystically and filled with mucus, and there is minimal infiltration by chronic inflammatory cells in the stroma in most cases [4]. The microscopic appearance is similar to that of a lesion of the gallbladder known as adenomyomatous hyperplasia [1].

[Table 1](#) shows the reported cases of adenomyomatous hyperplasia of the extrahepatic bile duct. In total, 13 cases have been reported previously [1–3, 5–14]. Of these, 2 were men and 11 were women. Mean age was 59.9 years (range 31–82 years). The location of the lesion was upper bile duct in 3 cases, middle in 5 cases, and lower in 5 cases. The most frequent chief complaint was abdominal pain (46.1%), followed by no complaint (30.7%).

It is difficult to diagnose adenomyomatous hyperplasia of the extrahepatic bile duct preoperatively or intraoperatively. In fact, the precise preoperative or intraoperative diagnosis could not be given in all previously reported cases, except one that was diagnosed as adenoma by ERCP biopsy. Adenomyomatous hyperplasia of the extrahepatic bile duct is usually diagnosed by histopathologic examination after surgery,

and previous reports indicate that there is no useful imaging technique to distinguish this lesion from cancer [14]. Consequently, this leads patients to undergo radical resection and not conservative resection. Of the reported cases, 5 were treated by pancreatoduodenectomy and 8 by resection of the extrahepatic bile duct. In our case, we performed resection of the extrahepatic bile duct accompanied by radical lymph node resection, according to operative procedure for bile duct cancer.

Adenomyomatous hyperplasia is thought to be a benign lesion, however previous reports revealed that there is a possibility of malignant transformation of adenomyomatous hyperplasia of various organ, or of recurrence in insufficiently resected cases [1, 6]. Iwaki et al. pointed out the risk of making a diagnosis of adenomyomatous hyperplasia based on findings of preoperative biopsy alone and suggested the necessity of performing radical surgery to make a definitive diagnosis [1].

In conclusion, adenomyomatous hyperplasia of the extrahepatic bile duct has to be taken into consideration when treating patients with bile duct stenosis. Preoperative endoscopic and radiological evaluations and intraoperative section biopsies are insufficient for differentiating adenomyomatous hyperplasia from other malignant tumors. Thus, radical surgical procedure and histological confirmation by surgical specimen is needed for a definitive diagnosis and local control when adenomyomatous hyperplasia is suspected.

Table 1. Reported cases of adenomyomatous hyperplasia of the extrahepatic bile duct

First author, year	Sex	Age	Location in bile duct	Chief complaint	Pathologic diagnosis before surgery	Treatment
Dowdy (1962)	F	38	middle	jaundice	not performed	resection of EHBD
Burhans (1971)	F	62	upper	weight loss	not performed	resection of EHBD
Cook (1988)	F	82	middle	abdominal pain	not performed	resection of EHBD
Ikei (1989)	M	52	lower	abdominal pain	not performed	PD
Legakis (1990)	F	55	middle	abdominal pain	not performed	resection of EHBD
Matsumoto (1992)	M	75	lower	not described	not described	PD
Imai (1995)	F	54	upper	none	not performed	resection of EHBD with LND
Lauffer (1998)	F	69	middle	none	adenoma by ERCP biopsy	resection of EHBD
Tsukamoto (1999)	F	31	middle	abdominal pain	not performed	resection of EHBD
Ojima (2000)	F	64	lower	abdominal pain	not performed	PD
Sato (2000)	F	64	upper	none	no malignancy by cytology	resection of EHBD with LND
Aoun (2005)	F	71	lower	abdominal pain	no malignancy by cytology	PD
Iwaki (2008)	F	62	lower	none	no malignancy by cytology	PPPD
Present case	M	54	middle	jaundice	no malignancy by cytology	resection of EHBD with LND

EHBD = Extrahepatic bile duct; ERCP = endoscopic retrograde cholangiopancreatography; LND = lymph node dissection; PD = pancreatoduodenectomy; PPPD = pylorus-preserving pancreatoduodenectomy.

Fig. 1. ERCP showed a 15-mm-long stenosis of the middle bile duct (arrow) and a slight dilatation of the common hepatic bile duct above the stenosis.

Fig. 2. Gross examination showing a locally hypertrophic lesion in the middle bile duct. The lesion is 15 × 20 mm in-size, whitish, and solid (arrows).

Fig. 3. Histological examination of the resected specimen demonstrated fibrous thickening of the wall in the middle bile duct along with multiple hyperplastic glands with no atypia, proliferation of smooth muscle components, and local infiltration by inflammatory cells (H&E, × 40).

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