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# Inflammatory Polyp in the Common Bile Duct with Pancreaticobiliary Maljunction

Yuichiro Uchida Yusuke Ome Keita Shimata Yoshio Nagahisa  
Michio Okabe Kazuyuki Kawamoto Tae Bum Park Tadashi Itoh  
Keizo Ogasahara

Department of General Surgery, Kurashiki Central Hospital, Kurashiki, Japan

## Key Words

Inflammatory polyp · Bile duct polyp · Pancreaticobiliary maljunction

## Abstract

A 63-year-old woman was admitted because of epigastric pain and obstructive jaundice. Endoscopic retrograde cholangiopancreatography and intraductal ultrasonography revealed a 25-mm papillary nodule in the middle to inferior portion of the common bile duct (CBD). Pancreaticobiliary maljunction (PBM) was also identified. Contrast-enhanced computed tomography also showed an enhanced nodule in the CBD, and we suspected intraductal papillary neoplasm of the bile duct. We performed pylorus-preserving pancreatoduodenectomy. Postoperative pathological examination revealed an inflammatory polyp in the middle CBD. Inflammatory polyp in the bile duct is rare and there are no previous reports accompanied by PBM. PBM is a major risk factor for biliary tract cancer. Preoperative diagnosis of a benign disorder was difficult in this case.

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## Introduction

The preoperative diagnosis of biliary tumor can be difficult. Biopsy findings by endoscopic retrograde cholangiopancreatography (ERCP) sometimes conflict with the findings of diagnostic imaging. Pancreaticobiliary maljunction (PBM) is a risk factor for biliary tract cancer, the prevalence of biliary tract cancer being 21.6–42.4% among adult PBM patients [1]. Tumors in the biliary tract in patient with PBM are suggestive of malignancy. In 2010, the clinical entity intraductal papillary neoplasms of the bile duct (IPNB) was defined. IPNB

has an intraductal papillary growth pattern and includes intraductal papillary cholangiocarcinoma and its precursor lesions [2]. Most previous occurrences of common bile duct (CBD) polyps may now be categorized as IPNB. Surgical resection is recommended for IPNB without distant metastases because it has high malignant potential and often causes recurrent cholangitis and obstructive jaundice. Inflammatory polyps in the bile duct are extremely rare. This report presents the first case of inflammatory polyp in the CBD with PBM.

## Case Report

A 63-year-old woman was admitted to our hospital because of epigastric pain and obstructive jaundice. Contrast-enhanced computed tomography showed a 25-mm enhanced nodule in the middle to inferior portion of the CBD accompanied by proximal dilatation of the bile duct (fig. 1). Tumor markers such as DUPAN-2 and SPAN-1 were increased to 9,700 and 160 U/ml, respectively. Carcinoembryonic antigen and carbohydrate antigen 19-9 were within normal limits. ERCP, intraductal ultrasonography and magnetic resonance cholangiopancreatography (MRCP) revealed that the nodule was papillary, accompanied by PBM (fig. 2). In transpapillary histological biopsy, a papillary structure with edematous interstitial tissue was observed. Plasmacyte and lymphocyte migration was observed, but neoplastic lesions and epithelial dysplasia were not identified. No atypical cells were identified in cytological specimens of bile juice. Considering the findings of PBM, we suspected bile duct neoplasms, including bile duct cancer or IPNB. We performed pylorus-preserving pancreaticoduodenectomy (PPPD). Macroscopically, we found a 6-mm polyp in the middle CBD (fig. 3a). Gallstones were not found. Microscopically, we identified a normal ductal structure with multiple plasmacyte migration (fig. 3b). Immunostaining did not show proliferation of p53-positive or MIB-1-positive cells. The patient was finally diagnosed with an inflammatory polyp in the CBD accompanied by PBM.

## Discussion

In the biliary tract, gallbladder polyps are common, but CBD polyps are rare. Most CBD polyps have neoplastic changes. Burhans and Myers [3] reported that among 88 cases of extrahepatic bile duct polyps, papilloma was found in 44 cases and adenoma in 39. Those papillomas and adenomas may now be categorized as IPNB.

In the English and Japanese language literature, only five cases of inflammatory polyps in the CBD have been reported (table 1). It is suggested that mechanical stimuli are among the causes of inflammatory polyps in the CBD. T tube placement was done in three cases [4, 5]. A CBD stone was found in one case [6], and none of the previous cases had PBM.

PBM is a congenital anomaly in which the main pancreatic duct and CBD converge outside the duodenal wall. PBM is frequently reported in Asian countries and is one of the major causes of biliary tract cancer. The theory of the hyperplasia-dysplasia-carcinoma sequence seems to explain the carcinogenesis of PBM. Pancreatic juice refluxes to the biliary tract and mixes with bile, and the production of cytotoxic substances such as activated pancreatic enzymes occurs. Repeated damage and repair of the biliary mucosa contribute to various gene mutations. This in turn causes histological changes such as hyperplasia, metaplasia and dysplasia, and finally results in biliary carcinogenesis [7, 8]. Once PBM is diagnosed, surgery is recommended, although the type of procedure is controversial [1]. A relationship between

PBM and IPNB is unclear, but considering the hyperplasia-dysplasia-carcinoma sequence, there may be some correlation.

In our case, we estimated that reflux of pancreatic juice induced inflammation and polyp formation, but there are no reports of inflammatory CBD polyp accompanied by PBM to date. CBD stones or gallstones were not found. We performed PPPD because we suspected bile duct cancer or IPNB. The pathological diagnosis was an inflammatory polyp. Considering that PBM is a risk factor for biliary malignancy, preoperative denial of malignancy was difficult in this case, even though we obtained benign results in preoperative biopsy and cytological specimens.

### Disclosure Statement

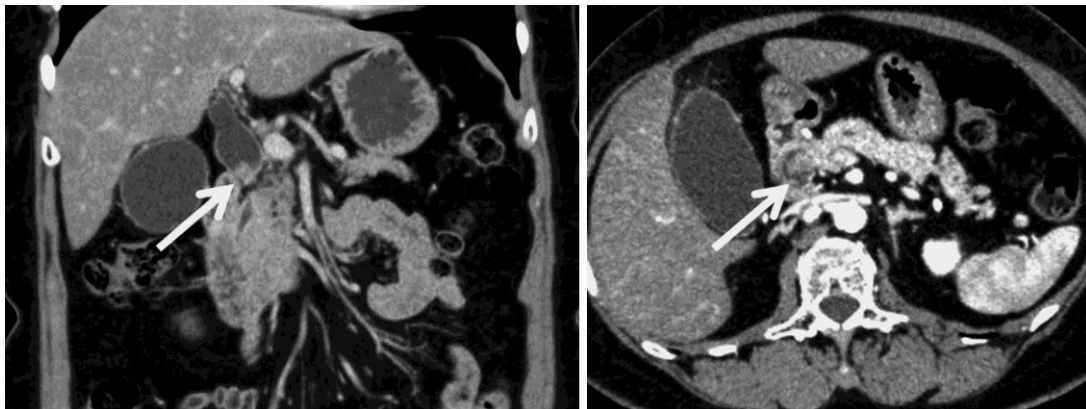
The authors have no potential conflicts of interest.

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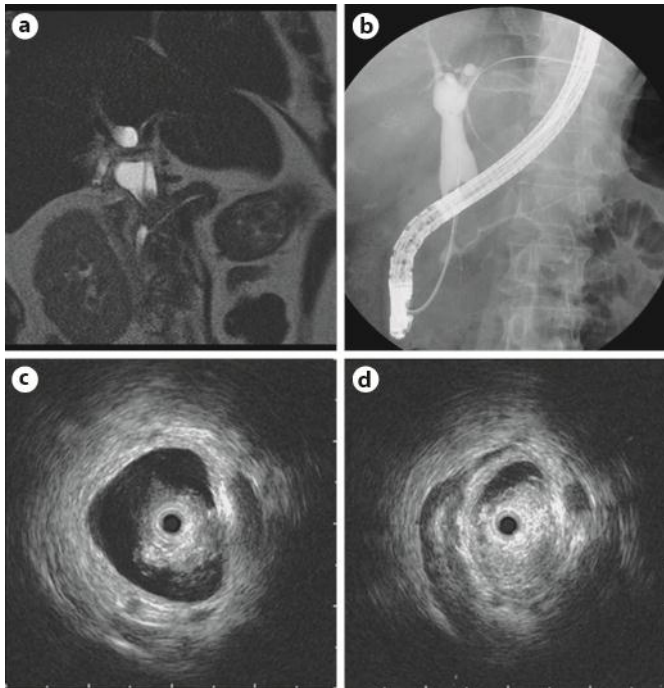
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**Table 1.** Reported cases of inflammatory polyp in the CBD

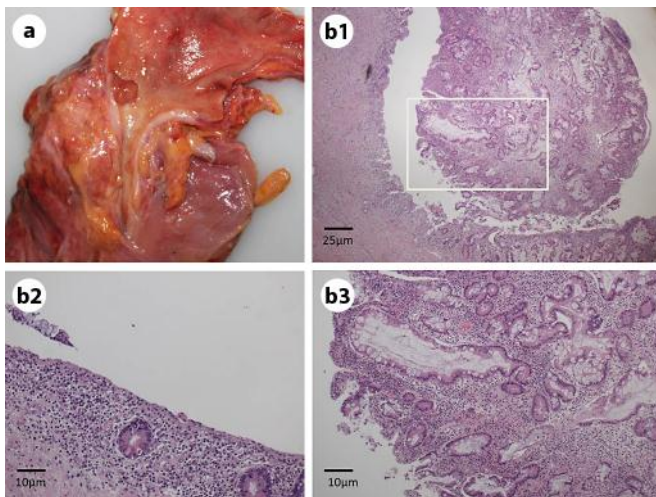
Reference	Age (years), sex	Symptom	Complication	Treatment
Ro [9]	55, m	jaundice	gallbladder stone	polyp resection
Shepherd et al. [6]	84, m	pancreatitis, jaundice	CBD stone	ERCP (sphincterotomy and stone removal)
Ishimura et al. [5]	80, m	none	CBD stone, T tube placement	endoscopic resection
Watanabe et al. [4]	27, m	jaundice	CBD stone, T tube placement	observation
	73, f	jaundice	CBD stone, T tube placement	observation
Present case	63, f	jaundice	PBM	PPPD



**Fig. 1.** A 25-mm enhanced nodule (arrow) in the middle to inferior portion of the CBD accompanied by proximal dilatation of the bile duct.



**Fig. 2.** a, b PBM was identified by MRCP (a) and ERCP (b). c, d Intraductal ultrasonography showed that the nodule was papillary.



**Fig. 3.** a A 6-mm polyp in the middle portion of the CBD. b1 Normal ductal structure and multiple plasmacyte migration were identified in the polyp (hematoxylin and eosin staining). b2 No atypical cells were found in the biliary mucosa around the polyp. b3 Enlargement of the inset in b1. b1–b3 Hematoxylin and eosin staining.