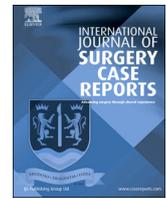




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## Long-term observation and treatment of a widespread intraductal papillary neoplasm of the bile duct extending from the intrapancreatic bile duct to the bilateral intrahepatic bile duct: A case report

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## ABSTRACT

**INTRODUCTION:** Few studies have reported the long-term outcomes of surgical resected intraductal papillary neoplasm of the bile duct (IPNB). Here, we describe the long-term observation and treatment of a case of widespread IPNB.

**PRESENTATION OF CASE:** A 57-year-old male was referred to our hospital due to jaundice and dilation of the intrahepatic bile duct. Computed tomography showed dilation and irregularities of the right intrahepatic and extrahepatic bile ducts together with a 3 cm nodule in the common hepatic duct. Peroral cholangioscopy revealed mucinous discharge from the ampulla of Vater, which resulted in a diagnosis of IPNB. A biopsy of the nodule and the bile duct revealed papillary adenoma in all of them. Right hepatectomy, caudate lobectomy, extrahepatic bile duct resection, and left hepaticojejunostomy were performed. The nodule was histologically diagnosed as papillary carcinoma in situ, and R0 resection was performed. However, mucus production from the papillary adenoma in the B3 and B4 was observed. We carefully managed the patient's biliary tract by inserting a biliary drainage tube into the segment 2, and he has survived for more than 7 years since the initial treatment.

**DISCUSSION:** Mucus might be produced after the surgical resection of IPNB even if a surgical margin was benign. Five-year survival rate of benign IPNB was reported from 85% to 100%. That might be caused by difference of the postoperative management of the biliary tract.

**CONCLUSIONS:** Careful management of the biliary tract should be performed after surgical resection of IPNB.

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### 1. Introduction

Intraductal papillary neoplasm of the bile duct (IPNB) is a rare tumor that was only recently classified as a distinct pathological entity [1]. It can produce multifocal lesions, develop within any part of the biliary tree, and typically displays an exophytic growth pattern [2]. One-third of IPNB cases are associated with macroscopic mucin hypersecretion, and dilation of the bile duct is often observed [3]. IPNB is considered to be the biliary equivalent of intraductal papillary mucinous neoplasm of the pancreas (IPMN-P), and identical histological subclassifications have been described for these types of tumor [4]. However, the mucus produced by IPNB is more a serious problem compared with that produced by IPMN-P because

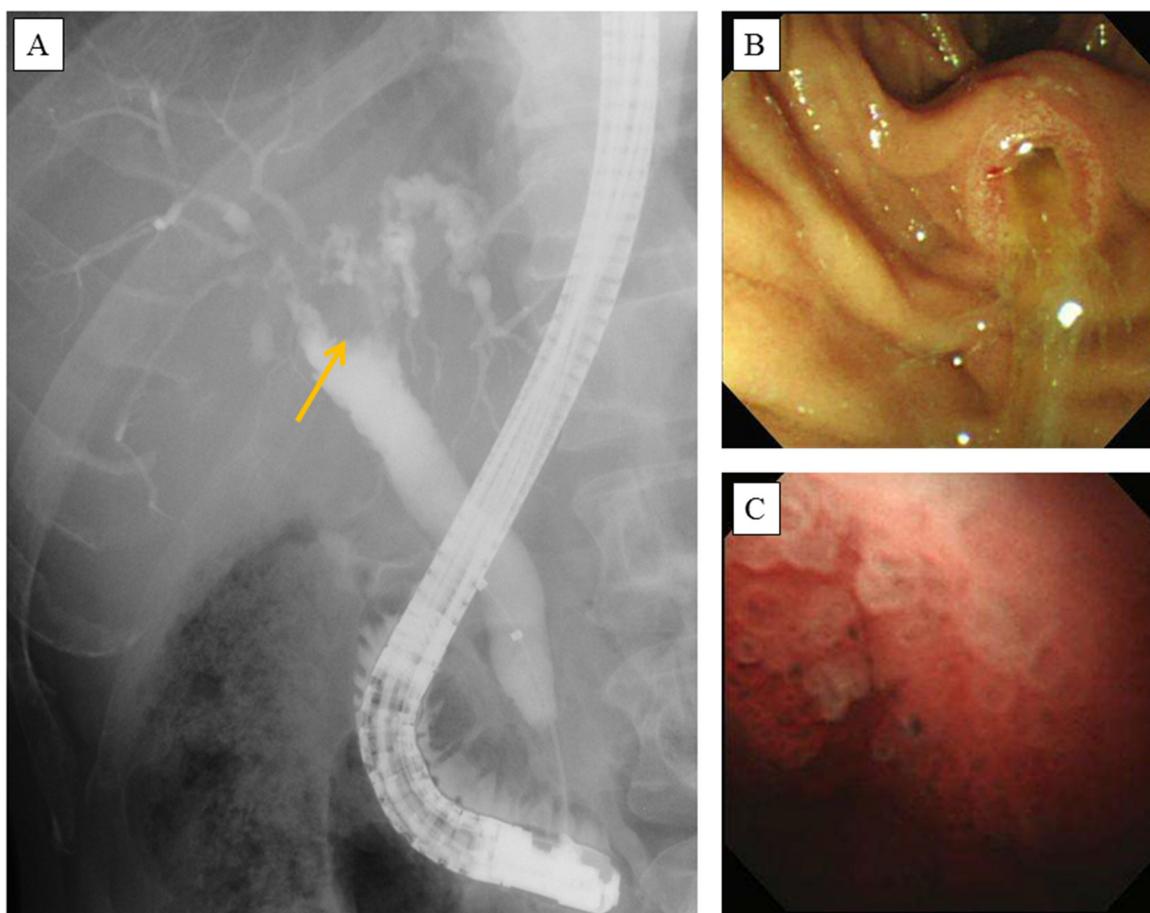
the obstruction caused by the mucus within the biliary tree can induce cholangitis or obstructive jaundice.

There have been few studies of the long-term outcomes and recurrence pattern of surgically resected IPNB [3,5–7]. IPNB includes adenoma, carcinoma-in-situ, and invasive carcinoma. The surgical margins of IPNB are usually evaluated histologically. If adenoma is detected in the surgical margins, the surgical resection is classified as R0. However, mucus production is even observed in cases of adenoma [1]. Even if the malignant region of an IPNB is completely resected, the mucus produced by the benign component can cause liver dysfunction or cholangitis. In such cases, the remnant IPNB could be considered clinically malignant, even if it is histologically benign.

Here, we report the long-term observation and treatment of a case of IPNB, in which the patient was finally diagnosed with widespread IPNB extending from the lower bile duct to the bilateral intrahepatic bile ducts. The mucus production in the remnant liver was observed even after R0 resection, and frequent cholangi-

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**Fig. 1.** A The findings of endoscopic retrograde cholangiography performed at the initial diagnosis are shown. Bile duct irregularities were observed in the region from the bilateral hepatic ducts to the peripheral bile duct, and a nodule measuring 1.5 cm in diameter was seen in the common hepatic duct (arrow). **B:** An endoscopic examination showed mucinous discharge from the ampulla of Vater. **C:** Peroral cholangioscopy showed papillary tumors (salmon roe-like lesions) located in the common hepatic duct.

tis were occurred. The patient has survived for over 7 years after surgical resection due to careful management of his biliary tract.

This work has been reported in line with the SCARE criteria [8,9].

## 2. Case reports

A 57-year-old male was referred to our hospital due to symptomless dilation of the intrahepatic bile duct in January 2010. Mild liver dysfunction was detected during a laboratory examination, while jaundice was not observed [total bilirubin (T-bil), 0.9 mg/dL; alkaline phosphatase (ALP), 599 U/L; aspartate aminotransferase (AST), 44 U/L]. Computed tomography (CT) and magnetic resonance cholangiopancreatography (MRCP) showed dilation of the bilateral intrahepatic and extrahepatic bile ducts and a nodule measuring 1.5 cm in the common hepatic duct. Endoscopic retrograde cholangiography (ERC) revealed bile duct irregularities extending from the bilateral hepatic ducts to the peripheral bile duct. In addition, the common bile duct was dilated (to 1.5 cm in diameter) and contained a nodule measuring 1 cm in diameter (Fig. 1A). Peroral cholangioscopy (POCS) revealed mucinous discharge from the ampulla of Vater, and the bile duct was full of mucus. In addition, many round tumors (salmon roe-like lesions) were observed in the region from the middle common bile duct to the common hepatic duct (Fig. 1B/C). The nodule in the common hepatic duct was biopsied, and a pathological examination revealed intraductal papillary adenoma. The lower bile duct and the left hepatic duct were also biopsied, which led to the lesions within them being diagnosed as intraductal papillary adenoma. A cytological examination of the

mucus did not reveal any evidence of malignancy. Based on these findings, the patient was diagnosed with IPNB, which was mainly located in the region from the middle common bile duct to the intrahepatic bile duct of the right and left hepatic lobes.

A watchful waiting strategy was employed because total resection of the irregular bile duct was impossible, and none of the histological examinations had detected findings that were suggestive of malignancy. However, 6 months later he was admitted to our hospital with jaundice and liver dysfunction [T-bil, 6.1 mg/dL; ALP, 1456 U/L; AST, 827 U/L]. MRCP and CT revealed that the nodule in the common hepatic duct had increased in size from 1.5 cm to 3 cm and that the hilar bile duct was obstructed (Fig. 2A/B/C/D/E). On the other hand, CT demonstrated that the irregularities extending from the left hepatic duct to the umbilical portion had disappeared (Fig. 2F/G). At this time, we determined that the IPNB, including the nodule and the irregular bile duct, could be resected via right hepatectomy and extrahepatic bile duct resection. An endoscopic nasobiliary drainage tube was inserted into the left hepatic duct, and the patient's jaundice was ameliorated. At the same time, the nodule was biopsied again, and a pathological examination revealed intraductal papillary adenoma. It was estimated that the remnant liver would be 27.7% of its original size after right hepatectomy combined with resection of the caudate lobe, and percutaneous transhepatic portal vein embolization was performed. Three weeks later, the estimated size of the remnant liver had increased to 36.6%. Right hepatectomy combined with resection of the caudate lobe and extra-hepatic bile duct followed by recon-

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