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Villous Adenoma of the Bile Ducts: A Case Report and a Review of the Reported Cases in Korea

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Abstract

Villous adenomas are benign epithelial lesions with malignant potential which can occur at any site in the gastrointestinal tract. They are usually encountered in the rectum and colon, less frequently in the small bowel and very rarely in the biliary trees. Nine cases of bile duct villous adenomas have been reported in the literature. However, 4 cases of bile duct villous adenomas have been reported in the Korean literature. Recently, we experienced a case of villous adenoma in the common hepatic duct in a 77-year-old man presenting with obstructive jaundice in which preoperative histologic diagnosis of villous adenoma played a critical role in managing this patient. Herein, we present a case report of bile duct villous adenoma and a review of the reported cases in Korea to help define and manage this rare disease entity in the bile ducts. In addition, confusing nomenclature of bile duct adenomas is discussed.

Key Words: Villous adenoma, bile duct, obstructive jaundice

INTRODUCTION

Villous adenomas are benign epithelial lesions with malignant potential which can occur at any site in the gastrointestinal tract. They are usually encountered in the rectum and colon, less frequently in the small bowel and very rarely in the biliary trees.[1] Since Saxe et al. first reported a case of villous adenoma of the common bile duct in 1988,[2] a total of 9 cases have been reported so far.[1-9] In Korea, however, 4 cases of bile duct villous adenoma have been reported since 1995.[10-12] Recently, we experienced a case of villous adenoma in the common hepatic duct (CHD) in a 77-year-old man in which preoperative histologic diagnosis of villous adenoma played a critical role in managing this patient. We believe that the addition of these 5 Korean cases to the previous 9 cases may help to define and manage this rare disease entity in the bile ducts.

CASE REPORT

A 77-year-old man was admitted to the hospital because of a 40-day history of itching sensation on April 22, 1998. He had visited the department of dermatology 20 days prior to admission. At that time, the blood chemistries were checked and revealed an obstructive pattern with a total serum bilirubin of 4.8 mg/dl, alkaline phosphatase of 372 U/L, AST of 81 IU/L, and ALT of 147 IU/L. However, he did not return and took some herb medicine which he said partially helped. Persistent itching led him to seek medical care again. The patient had undergone a hemorrhoidectomy in 1987 and had been admitted to hospital because of duodenal ulcer bleeding in 1992, 1994 and 1995. He had stopped drinking alcohol and smoking 10 years previously.

On admission, he complained of general weakness and dyspepsia, but denied fever, nausea, vomiting, epigastralgia, and weight loss. Physical examination revealed a temperature of 36.3°C, blood pressure of 130/70 mmHg, and a pulse of 84/min. The skin and sclerae were slightly icteric. The abdomen was soft but revealed a movable firm non-tender mass in the right upper quadrant, which was felt to be a distended gall bladder. Rectal examination was normal.

Laboratory findings included a hemoglobin of 11.1 g/dl, platelet count of 312,000/mm^3, and leukocyte count of 5,770/mm^3. The serum electrolyte, blood urea nitrogen, creatinine, amylase, total cholesterol, total protein, and albumin were normal. The total serum bilirubin was 1.8 mg/dl, the alkaline phosphatase 223 U/L, the AST 34 IU/L, and the ALT 43 IU/L. Coagulation studies were normal. Urinalysis was

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normal. Tumor markers including CA 19-9 and CEA were within normal limits. The stool was negative for occult blood.

An abdominal ultrasonography (US) and a subsequent computerized tomography (CT) scan showed a soft tissue mass from the confluence of the main right and left bile ducts to the CHD (Fig. 1). The CHD and common bile duct (CBD) were dilated, and the gall bladder was distended. However, no regional lymphadenopathy was found. A duodenoscopy showed normal ampulla of Vater. During the endoscopic retrograde cholangiopancreatography (ERCP) the distal CBD was so dilated with a filling defect that it was very difficult to push the contrast dye up the proximal biliary tree (Fig. 2A). An ERCP demonstrated an irregular-shaped filling defect in the CHD with dilatation of both intrahepatic ducts (IHD) (Fig. 2B). The pancreatic duct was normal. A sphincterotomy was performed, and then mucinous bile gushed out. A nasobiliary tube was placed and a cholangiogram via the nasobiliary tube revealed that the tumor appeared to extend to both IHDs (Fig. 3). A follow-up cholangiogram through the nasobilary tube done 6 days later revealed the same result, so this mass was considered to be unresectable. Palliative endoscopic stenting was planned. A cholangiogram showed several small filling defects in the CBD in addition to the main mass in the CHD. To rule out the presence of stones, a basket was inserted, but nothing was retrieved. A subsequent balloon extraction took a fragmented tissue out (Fig. 4). This tissue was retrieved by a biopsy forcep and later proved to be villous adenoma with microscopic foci of adenocarcinoma. A 10 French, 10 cm Amsterdam type plastic stent was placed. Because the result of histologic examination revealed that the main mass was a villous adenoma, we thought that the mass might be resectable with safety margins.

Surgical exploration was performed on 21st hospital

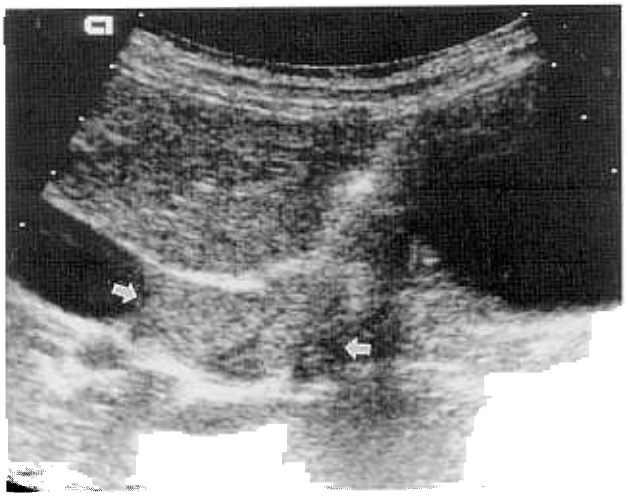

Fig. 1. Abdominal ultrasonography showing a soft tissue mass (arrows) from the confluence of the main right and left bile ducts to the common hepatic duct.

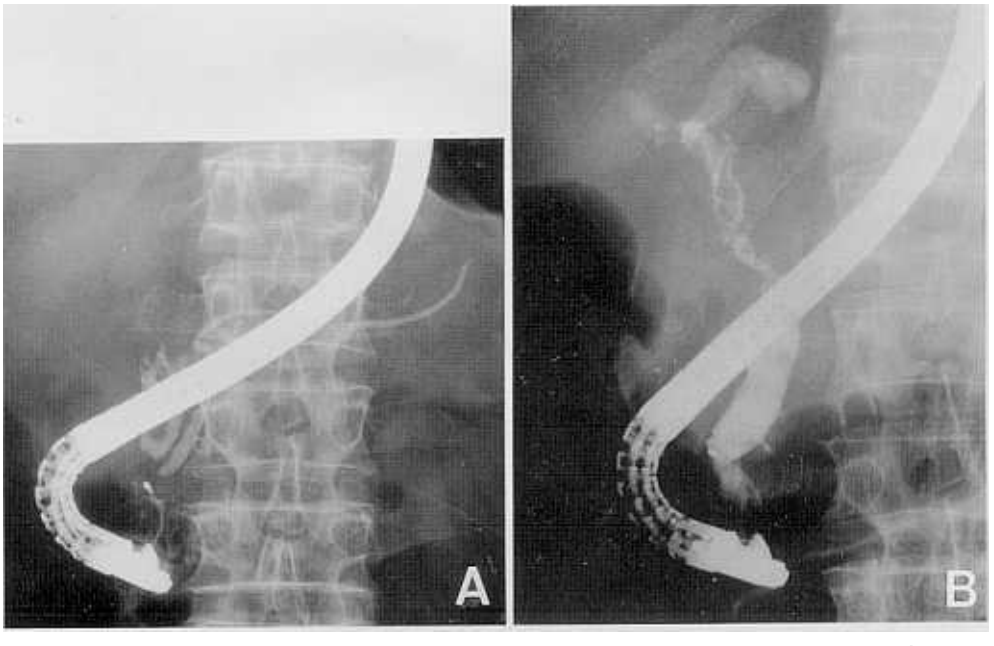

Fig. 2. Endoscopic retrograde cholangiopancreatography revealed a filling defect in the dilated distal common bile duct that initially made it difficult to push the contrast dye up the proximal biliary tree (A). An irregular-shaped filling defect is demonstrated in the common hepatic duct with dilatation of both intrahepatic ducts (B).

day and revealed a distended and thickened gall bladder. The mass was not palpable even after dissection of the porta hepatis. Segmental resection of the CHD and CBD and cholecystectomy were performed. However, despite several extensions of resection, the distal resection margin which was the proximal portion of the intrapancreatic CBD and the proximal resection margin of the left IHD were positive for dysplasia. In consideration of the patient's age and general condition, such extensive surgeries as left lobectomy and pancreaticoduodenectomy were abandoned. The resected specimen of the CHD and CBD showed a sessile and villous mass, measuring 2.8×1.0 cm. On microscopic examination, the tumor was composed of stratified tall columnar cells with capillary fronds extending into the lumen and supported by connective tissue from the lamina propria. Foci of well-differentiated adenocarcinoma were found in the villous adenoma and pushing into the subepithelial connective tissue with lymphovascular invasion (Fig. 5, A and B). The mucosal areas other than this villous tumor were flat dysplasia, but intervened by denuded areas. The lymph nodes were negative for malignancy.

The patient recovered uneventfully and has been on an oral anti-cancer agent (Doxifluridine) and is doing well (6 months after discharge).

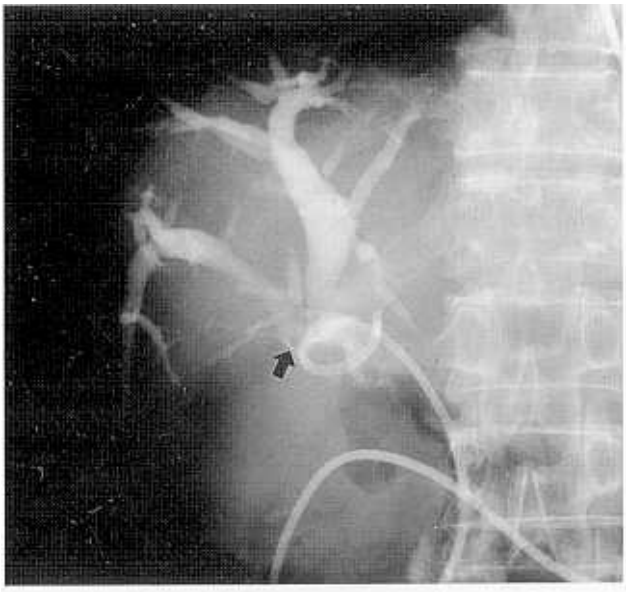

Fig. 3. Cholangiogram via the nasobiliary tube shows the tumor appearing to extend to right intrahepatic duct (arrow).

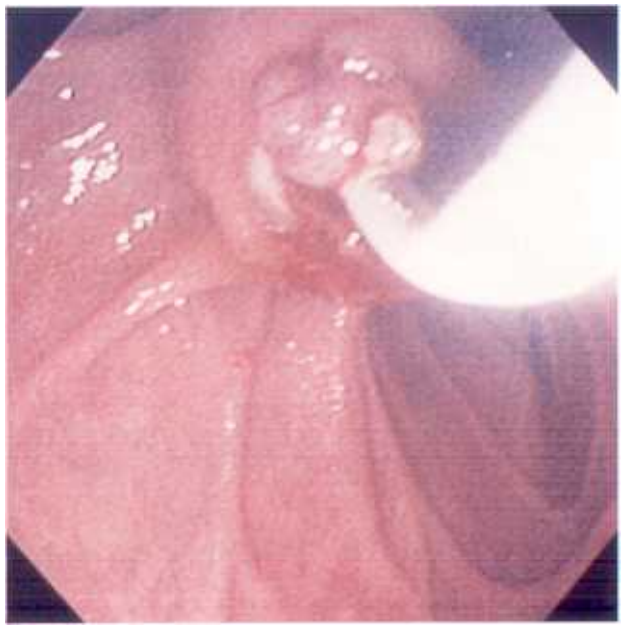

Fig. 4. An endoscopic view of a fragmented tissue on the ampulla of Vater taken out by balloon extraction.

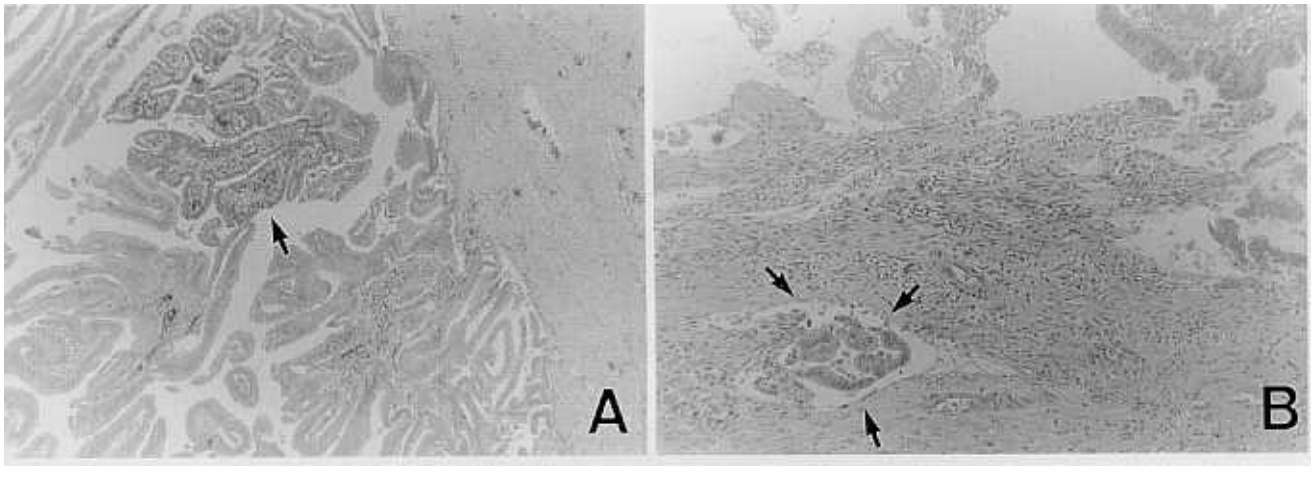

Fig. 5. (A) Microscopic findings of the operative specimen show villous adenoma with malignant foci (arrow) (H & E, ×40). (B) Foci of well differentiated adenocarcinoma with lymphovascular invasion (arrows) (H & E, ×100).

Table 1. Summary of the Cases of Bile Duct Villous Adenoma Reported in Korea

Case	Authors	Sex / Age	Symptoms	Duration	Total bilirubin (mg/dl)	Alkaline phophatase	Diagnostic procedures	Location	Size	Mucin hypersecretion	Diagnosis before definitive treatment	Treatment	Histology	Follow-up result	Comments
1	Roe et al.	M / 72	Fever, chill, RUQ abdominal pain jaundice	2 months	18.2	512	US, CT, ERCP	CHD	2.3×2.2 cm	Yes	No	Segmental resection	Benign villous adenoma	NM	
2	Lyou et al.	M / 59	Fever, chill, RUQ abdominal pain jaundice	2 months	6	746	US, CT, ERCP, PTBD	CBD & cystic duct orifice	3.5×1.8 cm	No	Yes	Roux-en-Y Hepaticojejunostomy	Benign villous adenoma	NM	Associated with C. sinensis infestation
3	Jeoung et al.	M / 72	RUQ abdominal pain, jaundice	7 days	11.2	675	US, CT, EUS, ERCP	CBD	1.5×1.3 cm	No	Yes	Transpapillary hot biopsy under PTCS and a metallic stent insertion	Benign villous adenoma	7 months, died due to sepsis	Associated with stenosis distal to the mass
4	Jeoung et al.	M / 76	Epigastric discomfort	3 months	0.6	66	US, EUS, CT, ERCP	CBD	1.4 cm	No	No	Transpapillary endoscopic resection under PTCS	Benign villous adenoma	6 months, alive	
5	Present case	M / 77	Pruritus	40 days	4.8	372	US, ERCP, CT	CHD with extension to both IHD's and CBD	2.8×1.0 cm	Yes	Yes	Segmental resection	Villous adenoma with foci of adenocarcinoma	6 months, alive	

NM, not mentioned; US, ultrasonography; CT, computerized tomography; ERCP, endoscopic retrograde cholangiopancreatography; PTBD, percutaneous transhepatic biliary drainage; EUS, endoscopic ultrasonography; CHD, common hepatic duct; CBD, common bile duct; IHD, intrahepatic duct; PTCS, percutaneous transhepatic choledochoscopy.

SUMMARY OF THE KOREAN PATIENTS

Five cases including the present case are summarized in Table 1. All were male and the mean age was 71.2 years old (Range, 59−77 years old). Four patients presented with symptoms related to biliary obstruction. In Case 4, presenting with a non-specific symptom, the tumor was discovered by a screening US. Elevation of serum total bilirubin and alkaline phophatase was noted in 4 patients presenting with symptoms related to biliary obstruction. US, CT, and ERCP were used in all patients and endoscopic ultrasonography (EUS) was performed in 2 patients who underwent endoscopic treatment. The main mass was located in the CBD in 3 patients and in the CHD in 2 patients. The mean maximal diameter of the tumor was 2.3 ± 0.89 cm. Mucin hypersecretion was noted in 2 cases. Before a definitive treatment, a diagnosis of villous adenoma was made in 3 patients. Treatment included a segmental resection in 2, endoscopic treatment in 2, and a bypass surgery in 1. The techniques of endoscopic treatment were a transpapillary hot biopsy of the tumor under percutaneous transhepatic choledochoscopy (PTCS) in one and a transpapillary polypectomy with a snare under PTCS in the other. Follow-up data were available in 3 cases, but the follow-up duration was too short to draw any conclusions. Final histologic diagnoses were benign villous adenomas in 4 and villous adenoma with foci of adenocarcinoma in 1 case.

DISCUSSION

In 1988, Saxe et al. first described a case of villous adenoma in the CBD. According to Doberauer et al. 9 cases of villous adenoma in the bile ducts (including 1 case of tubulovillous adenoma) have been reported in the literature, including their own.[9] It is noteworthy that 5 additional cases have clustered in Korea in a 3-year period. It may be partly attributed to the widespread use of ERCP in Korea. The reason that the incidence of bile duct villous adenoma is low in Western contries might be the result of confusion in terminology.

Adenomas of the bile ducts were divided into papillary adenomas, pedunculated adenomas, and sessile adenomas according to the gross configuration of the tumors.[13] This classification replaced an earlier classification of papillomas, polyps, and adenomas.[14] Villous adenomas are thus classified as frond-like sessile adenomas, and papillomas are classified as papillary adenomas.[3] Histologically, however, adenomas are classified into tubular adenoma, tubulovillous adenoma, and villous adenoma.[15] Thus, papillomas or papillary adenomas are equivalent or at least very close to villous adenomas histologically. In fact, Kawakatsu et al. used papillary adenomas and villous adenomas interchangeably.[16] Biliary papillomatosis (or papillary adenomatosis) is composed of villous tumors with slender fibrovascular cores and tends to secrete much mucin. Therefore, it may also be classified as villous adenoma. If these are taken into account, the incidence of bile duct villous adenoma in Western contries would be higher. Other bile duct adenomas in Korea, included 2 case of tubulovillous adenomas from Korea University[17] and Yonsei University (our own unpublished case), 1 case of papillary adenoma,[18] and 10 cases of biliary papillomatosis.[19,20]

We believe that confusion in terminology of bile duct adenomas should be settled. Histologic classification including tubular, tubulovillous, and villous adenomas would be a good option. Otherwise, a term that encompasses all clinical and histologic features of bile duct adenomas should be adopted. For example, intraductal papillary mucinous tumors (IPMT) of the pancreas encompass hyperplasia to invasive carcinoma and main duct type to branch duct type with or without mucin hypersecretion.[21,22] Bile duct adenomas have a close resemblance to IPMT of the pancreas in that they are composed of mucin secreting cells, may hypersecrete mucins, may be focal or diffuse, and most importantly they are pre-malignant lesions which need to be completely resected. Therefore, we suggest that IPMT's of the bile ducts be used to describe these rare conditions in the bile ducts. This problem of nomenclature should be ironed out by consensus meetings or authorities.

Of note in reviewing the reported cases of bile duct villous adenomas in Korea is that the preoperative diagnoses or diagnoses without operation were possible in 4 out of 5 cases reflecting the current sophisticated diagnostic and therapeutic techniques of the biliary tract and pancreas. In the present case, preoperative histologic diagnosis of villous adenoma played a critical role in managing this patient. If the lesion had been cholangiocarcinoma, it would have been unresectable because the lesion appeared to involve both IHD's cholangiographically. Although the patient had positive margins for dysplasia, the main mass and foci of adenocarcinoma were removed successfully and he has remained asymptomatic after operation. Cholangioscopic examination through a transpapillary or percutaneous route would help to plan the management of these kind of extensive cases.[19,23] Endoscopic removal of the tumors was per-

formed in 2 cases which reportedly refused to undergo surgery. They used a novel approach including a transpapillary endoscopic tumor resection using a hot biopsy or a snare under PTCS guidance. This technique would be inappropriate in treating villous adenomas of the bile ducts because villous adenomas by definition generally have broad bases. However, in selected patients who have small tumors but are poor surgical candidates or refuse operation, this technique would be a feasible therapeutic alternative.

In summary, we described a case of bile duct villous adenoma in which preoperative histologic diagnosis of villous adenoma played a critical role in managing a 77-year-old man and reviewed the reported cases in Korea to help define this rare entity in the bile ducts. Nomenclature of bile duct adenomas was discussed and it is suggested that IPMT's of the biliary tract be used so that they encompass all the clinical and histologic features of bile duct adenomas.

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