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CASE REPORT

Carcinoma *in situ* arising in a tubulovillous adenoma of the distal common bile duct: A case report

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Abstract

Tubulovillous adenomas are common in the colon and rectum, but are rare in the common bile duct. Biliary adenomas may produce obstructive jaundice, which can be easily confused with a malignant neoplasm or stone. We report a case of a carcinoma *in situ* arising in a tubulovillous adenoma of the distal common bile duct causing obstructive jaundice. A 55-year-old male presented with a 10-d history of pruritus and progressive jaundice. Abdominal sonography and computed tomography showed a mass in the distal common bile duct. Endoscopic retrograde cholangiopancreatography showed luminal narrowing of the bile duct due to a polypoid mass. Positron emission tomography demonstrated no abnormal uptake. It was thought that this mass was a malignant tumor, thus a pylorus-preserving panceaticoduodenectomy was performed. The final pathology showed a tubulovillous adenoma with carcinoma *in situ* of the distal common bile duct. At follow-up 8 mo later, endoscopy showed multiple polyps in the rectum, colon and stomach. The polyps were removed by endoscopic mucosal resection and shown to be tubular adenomas with high grade dysplasia. Biliary adenomas require careful follow-up for early detection of recurrence and malignant transformation.

Key words: Common bile duct; Adenoma; Carcinoma *in situ*

INTRODUCTION

Biliary adenomas are very rare tumors, which may pose diagnostic dilemmas preoperatively. Due to recent advances in diagnostic techniques and early diagnosis, biliary adenomas are often detected as high grade dysplasias or carcinomas *in situ*. The optimal therapeutic strategy for adenomas of the distal common bile duct (CBD) or ampullary region has not been established. We report herein a case of a bile duct tubulovillous adenoma with carcinoma *in situ* presenting painless jaundice in a man who was treated with a pylorus-preserving pancreaticoduodenectomy (PPPD) and presented 8 mo later with a tubular adenoma in the gastrointestinal tract with high grade dysplasia. Our patient did not exhibit any clinical signs of familial adenomatous polyposis[1] or Gardner's syndrome[2]. We also reviewed the literature about the common bile duct adenoma found in the English literature[3-22] (Table 1).

CASE REPORT

A 55-year-old man was admitted to our hospital with a 10-d history of painless obstructive jaundice and pruritus. He had diabetes mellitus which had been controlled by an oral hypoglycemic agent. He had no family history of colorectal cancer or polyposis. On physical examination, he had icteric sclerae. Abdominal examination revealed no palpable mass or tenderness. Laboratory tests showed elevations of total bilirubin (8.2 mg/dL), direct bilirubin (6.3 mg/dL), alkaline phosphatase (302 IU/L), alanine transaminase (27 IU/L) and gamma glutamyl transpep-

Table 1 Reported cases of common bile duct adenomas

Author	Sex	Age(yr)	Presentation	Treatment	Histology
Hulten, 1970[3]	M (n = 2)	61, 80	Bilary colic (n = 1) and jaundice (n = 2)	Local excision (n =2)	Papilloma (n = 2) with moderate atypia
Styne, 1986[4]	F (n = 1)	59	Recurrent cholangitis	Local excision	Papilloma
Saxe, 1988[5]	M (n = 1)	64	Painful jaundice and pruritus	Whipple	Villous adenoma
Harshfield, 1990[6]	M (n = 1)	78	Chronic right upper quadrant pain	Local excision	Villous adenoma
Sturgis, 1992[7]	F (n = 1)	81	Right upper quadrant pain	Endoscopic excision	Tubulovillous adenoma
Hanafy, 1993[8]	M (n = 1)	76	Mild jaundice and abdominal mass	Local excision	Villous adenoma
Buckley, 1993[9]	M (n = 1)	34	Chronic jaundice and abdominal pain	Whipple	Villous adenoma with malignant foci
Blot, 1996[10]	M (n = 1)	84	Febrile jaundice	Local excision	Villous adenoma
Kawakatsu, 1997[11]	F (n = 3) M (n = 2)	60.6 (mean)	Febrile jaundice	Whipple (n = 2), local excision (n = 3)	Villous adenoma with mild dysplasia (n = 1), malignant foci (n = 4)
Chae, 1999[12]	M (n = 1)	77	Painless jaundice and pruritus	Local excision	Villous adenoma with malignant foci
Inagaki, 1999[13]	M (n = 1)	73	Epigastric pain and jaundice	Whipple	Papilloma
Chang, 2001[14]	M (n = 1)	51	Febrile jaundice, abdominal mass	Operation refused	Papilloma with focal dysplasia
Oshikiri, 2002[15]	F (n = 1)	69	Jaundice	Whipple	Papilloma
Ariche, 2002[16]	F (n = 1)	77	Abdominal pain	Local excision	Villous adenoma with adenocarcinoma
Aggarwal, 2003[17]	M (n = 1)	55	Abdominal pain	Whipple	Adenoma with moderate dysplasia
Jao, 2003[18]	M (n = 1)	60	Abdominal screening ultrasound	Endoscopic excision	Tubulovillous adenoma
Lou, 2003[19]	M (n = 1)	47	Fever, abdominal pain	Local excision	Tubular adenoma with moderate dysplasia
Fletcher, 2004[20]	M (n = 1)	74	Painless jaundice and pruritus	Whipple	Papilloma
Katsinelos, 2006[21]	M (n = 1)	58	Painful jaundice	Whipple	Villous adenoma with atypia
Xu, 2008[22]	F (n = 1)	27	Painless jaundice and pruritus	Whipple	Villous adenoma with mild dyplasia
Present case	M (n = 1)	55	Painless jaundice and pruritus	PPPD	Tubulovillous adenoma with carcinoma *in situ*

PPPD: Pylorus-preserving pancreaticoduodenectomy.

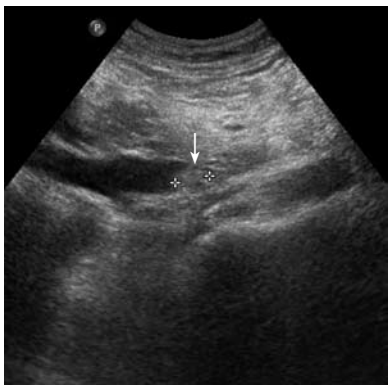

Figure 1 Ultrasound examination reveals a 2 cm sized non-shadowing mass and a dilated common bile duct (arrow).

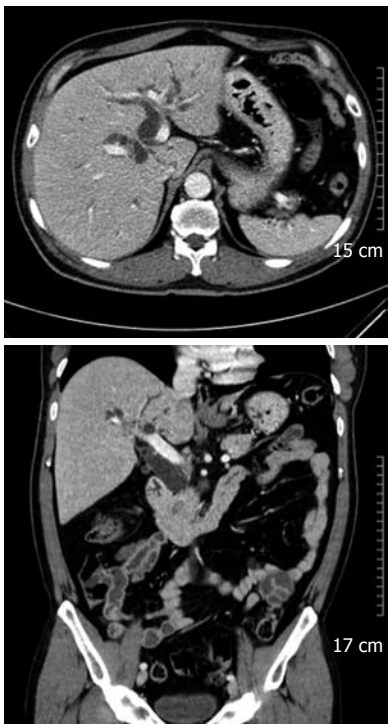

Figure 2 Computed tomography shows a diffuse dilatation of the common bile duct with an intraluminal mass in the distal common bile duct.

tidase (321 IU/L). The serum amylase was within the normal range. The carbohydrate antigen 19-9 level was 131.6 U/mL (normal range, ≤ 27 U/mL) and the carcinoembryonic antigen level was 1.5 ng/mL (normal range, 5 ng/mL). The hepatitis serologic markers were all negative. On abdominal ultrasonography, the CBD was dilated with a distal non-shadowing polypoid mass, however, there was no pancreatic duct dilatation (Figure 1). The CT findings were similar to the ultrasonographic findings and therefore a distal CBD tumor was suspected (Figure 2). Endoscopic retrograde cholangiopancreatography showed a 2cm polypoid mass and stricture in the distal CBD (Figure 3). Bile cytology revealed no malignancy. Positron emission tomography (PET) showed no hypermetabolic lesions. Based on the preoperative diagnosis of a distal tumor, a PPPD was performed.

The resected specimen revealed a 2 cm × 1.5 cm polypoid mass in the distal CBD (Figure 4). The final pathology showed a tubulovillous adenoma with carcinoma *in situ* in the distal CBD (Figure 5). There was no lymph node metastasis. The patient recovered uneventfully. Eight months later, he developed multiple polyps (two in rectum, three in the colon and two in the stomach). An endoscopic mucosal resection was performed, which revealed a tubular adenoma with high grade dysplasia.

DISCUSSION

Tubulovillous adenomas are usually encountered in the gastrointestinal tract, but as a primary site, the CBD is

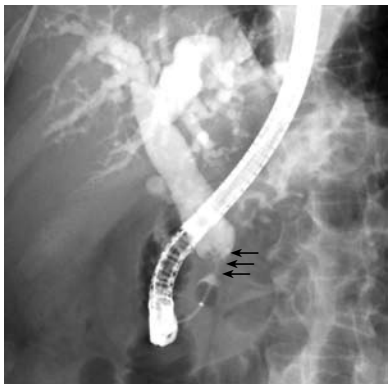

Figure 3 Endoscopic retrograde cholangiopancreatography shows a 2 cm round lobulated filling defect in the distal common bile duct (arrows).

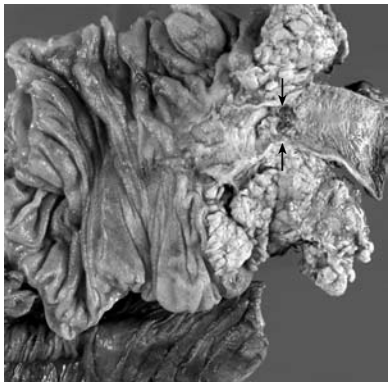

Figure 4 A photograph of the pylorus-preserving pancreaticoduodenectomy specimen shows a polypoid mass in the distal common bile duct, 2 cm x 1.5 cm x 1.5 cm size (arrows).

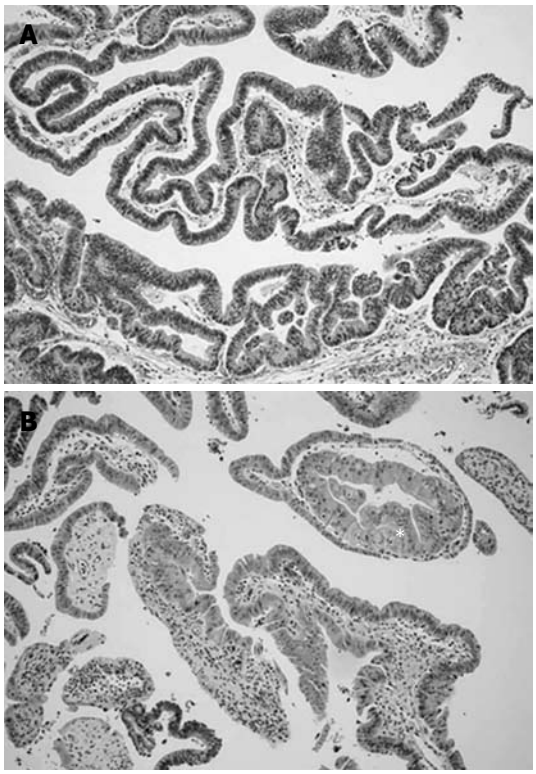

Figure 5 Microscopic features of the tubulovillous adenoma. Hematoxylin-eosin stain. **A**: The elongated and pseoudostratification of nuclei are stained (x 40); **B**: Higher magnification on this slide demonstrates carcinoma *in situ* (asterisk; x 100).

rare. Adenomas arising from the CBD are summarized in Table 1. Saxe *et al*[5] was first to report a case of a villous adenoma in the CBD. The clinical manifestations of a biliary adenoma include jaundice, right upper quadrant abdominal pain, dyspepsia, nausea and vomiting in a fashion similar to ampullary tumors. Villous adenomas are benign tumors, but are considered to be premalignant. It is possible that the adenoma-to-carcinoma sequence occurs in biliary tumors[23-26]. Considering the similarity of the histologic and biologic characteristics of adenomas in the other segments of the GI tract, such an adenoma-to-carcinoma carcinogenic process involving the rectum, ampulla, gallbladder, and biliary duct occurs within the biliary tract[27,28]. Therefore, complete resection of the lesion makes it possible to avoid development of carcinoma[29]. It is difficult to differentiate biliary adenomas from other malignant lesions with radiologic imaging[23]. Predicting the presence of malignant foci preoperatively is difficult. However, suspicion of malignancy could be made by an experienced biliary endoscopist.

Appropriate management of these lesions in the distal CBD has not been clearly defined. In 1992, Sturgis *et al*[7] first reported that high-risk patients with tubulovillous adenomas of the CBD were best treated by endoscopic resection but the risk of recurrence is high. Other treatment options, such as local resection, are performed in high-risk patients thought preoperatively to have benign tumors[10]. Ariche *et al*[16] proposed that resection with free margins of the CBD with lymph node dissection of the hepatoduodenal ligament for tumors in the mid part of the CBD is an appropriate treatment option. If the remaining duct length is inadequate, local resection is impossible and PD should be considered mandatory in cases involving cancer of the distal CBD. If malignancy is suspected or the size is larger than approximately 2 cm, radical resection is needed. We considered the other treatment option, duodenum preserving pancreatic head resection(DPPHR) which was first introduced by Beger *et al*[27] for chronic pancreatitis and has been increasingly used in neoplastic lesions, cystadenoma, borderline lesions, and carcinoma *in situ*[28-30]. However, we suspected that this tumor would be malignant and performed PPPD considering the size, site and clinical findings. Compared to Whipple-type resection, duodenum preserving pancreatic head resection has benefits in regard to postoperative morbidity and mortality, maintenance of glucose metabolism, absence of delay of gastric emptying, shorter hospital stay offers better quality of life of patients. However, duodenum preserving pancreatic resection has two major problems, incomplete lymph node dissection and ischemia of duodenum and has not been used yet as a surgical option of adenoma of common bile duct. Maeda *et al*[31] reported that duodenum preserving pancreatic resection in the treatment of pancreatic metastasis from renal cell carcinoma should be considered as radical lymph node dissection is not necessary. And then, if the nature and extent of common bile duct adenoma is suggestive of benign tumor and lymph node enlargement is absent, preoperatively, DPPHR should be considered in the treatment of common bile duct adenoma.

In this case, colonoscopy has not been performed and gastroscopy revealed no tumors in the stomach and

duodenum preoperatively. At the time of follow-up 8 mo postoperatively, colonoscopy and gastroscopy revealed multiple polyps in the rectum, sigmoid colon, and stomach. They were removed by endoscopic mucosal resection and confirmed as tubular adenomas with high grade dysplasia. In view of the risk of recurrence of adenomas, careful follow-up is in order.

Järvinen *et al*[32] have reported biliary involvement in familial adenomatous coli patients. The present report is the first case of a tubulovillous adenoma with carcinoma *in situ* of the distal CBD and several tubular adenomas with high grade dysplasia of the GI tract confirmed 8 mo apart. We think that although adenoma of the biliary tract and GI tract did not exist concurrently, tubulovillous adenoma of the distal CBD may have developed by a similar mechanism to that of the GI tract. In conclusion, this case suggests that adenomas arising from the distal CBD can transform into carcinoma and support the existence of an adenoma-to-carcinoma sequence given that carcinoma *in situ* with an adenomatous lesion of the distal CBD and tubular adenoma of the GI tract adenoma ultimately developed.

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