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CASE REPORT

Adenomas of the common bile duct in familial adenomatous polyposis

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Abstract

Familial adenomatous polyposis (FAP) or Gardner's syndrome is often accompanied by adenomas of the stomach and duodenum. We experienced a case of adenomas of the common bile duct in a 40-year-old woman with FAP presenting with acute cholangitis. Only 8 cases of adenomas or adenocarcinoma of the common bile duct have been reported in the literature in patients with FAP or Gardner's syndrome. Those patients presented with acute cholangitis or pancreatitis. Local excision or Whipple procedure may be the reasonable surgical option.

Key words: Acute cholangitis; Gardner's syndrome; Familial adenomatous polyposis; Common bile duct

Core tip: Adenomas or adenocarcinoma in familial adenomatous polyposis (FAP) and Gardner's syndrome are less frequently in the common bile duct (CBD). We report a case of FAP associated with CBD adenomas presenting with symptoms of acute cholangitis and cured after a radical Whipple procedure.

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INTRODUCTION

Adenomas or adenocarcinomas in familial adenomatous polyposis (FAP) and Gardner's syndrome (GS) are often found frequently in the colon and rectum, and less frequently in the ampulla of Vater[1-3], stomach[4-6] and duodenum[7-10]. There are a few reports[11-15] of patients with FAP or GS who have developed pancreatitis or acute cholangitis caused by adenomas of the common bile duct (CBD). Here, we report a case of acute cholangitis due to adenomas of the CBD in a patient with FAP, accompanied by adenomatous changes in the stomach, duodenum and the ampulla

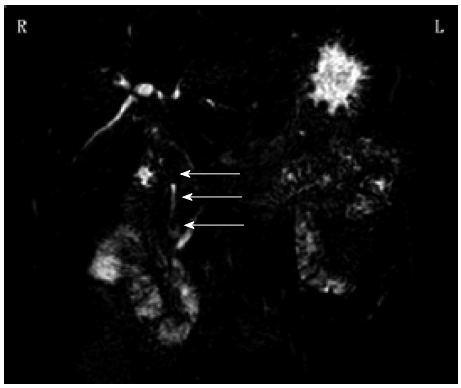

Figure 1 Magnetic resonance cholangiopancreatography shows numerous masses in the common bile duct.

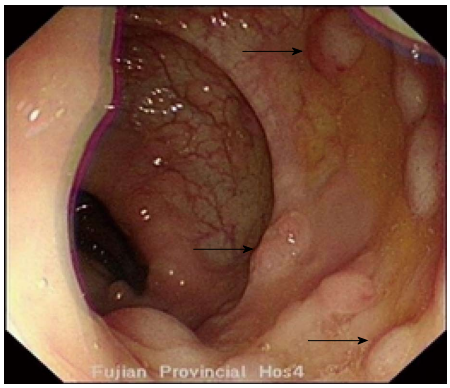

Figure 2 Colonoscopy shows multiple polyps in the colon.

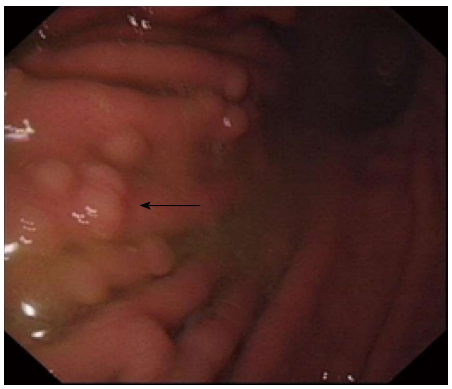

Figure 3 Gastroscopy shows multiple polyps in the duodenum and the distal stomach.

of Vater.

CASE REPORT

A 40-year-old woman was admitted to our hospital in July 2013 for fever and abdominal pain. Eleven months ago, she underwent laparoscopic cholecystectomy, CBD exploration and mass resection with T-tube drainage at the local hospital for acute cholangitis. Multiple polypoid lesions were seen in the distal CBD. Pathological analysis showed adenomas with mild to moderate dysplasia. She underwent adenoma resection through the T-tube in the CBD 8 mo ago, and pathology showed villous adenomas with moderate to severe dysplasia. Three days ago, she felt painful in the right upper quadrant with temperature up to 39.5 ℃ and symptomatic treatment was given at the local hospital.

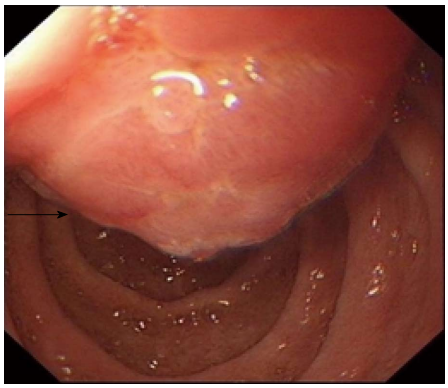

Figure 4 Ampulla of Vater is apparently protuberant and the surface is rough.

On admission, her liver function tests showed that total bilirubin was 1.0 mg/dL, serum amylase 145 IU/L, ALT 168 U/L, AST 88 U/L, GGT 858 U/L, and alkaline phosphatase 520 U/L. Magnetic resonance cholangiopancreatography revealed numerous masses in the CBD and the greatest diameter of the CBD was up to 1.7 cm (Figure 1). Her mother, younger brother and one of three sisters had FAP. Colonoscopy revealed villous polyps in the colon with the diameter from 0.3 to 2.5 cm (Figure 2). Duodenoscopy showed multiple polyps in the duodenum and the distal stomach (Figure 3). The ampulla of Vater was apparently protuberant and the surface was rough (Figure 4). The polyps were adenomas with low-grade intraepithelial neoplasia. Capsule endoscopy found no abnormality in the small intestine.

On July 29, a radical Whipple procedure was performed. After the operation, we found a 5 cm × 4 cm mass in the ampulla of Vater, and multiple polyps in the descendant duodenum and distal stomach with the largest diameter from 0.1 to 0.7 cm. Numerous polyps filled the middle and distal CBD, and the greatest diameter of polyps was about 1.2 cm (Figure 5). Microscopic examination of the ampullary and distal CBD polypoid lesions showed adenomas with high-grade intraepithelial neoplasia (Figure 6). She had an uneventful postoperative recovery. Seven months later, she underwent subtotal colectomy with ileosigmoid anastomosis. There were hundreds of polyps in the colon which turned out to be tubular adenomas pathologically.

DISCUSSION

There are a few reports of patients with FAP or

Table 1 Reported cases of familial adenomatous polyposis associated with adenomas or adenocarcinoma in the common bile duct

Ref.	Sex	Age	Presentation	Site of obstruction	Treatment	Pathology
Lees *et al*[11]	M	22	Jaundice	Distal CBD	T-tube drainage	Adenocarcinoma
	F	40	Jaundice, pruritus, bifurcation	Hepatic duct, radiation	T-tube drainage	Adenocarcinoma
Järvinen *et al*[12]	M	58	Jaundice with pain	CBD, left hepatic duct	T-tube drainage	Adenocarcinoma
	F	27	Choledochoscopy	Distal CBD	Whipple	Adenoma
Komorowski *et al*[13]	F	56	Abdominal pain	Distal CBD	Whipple	Carcinoma in situ
Spigelman *et al*[14]	M	78	Weight loss, abdominal pain	Extrahepatic duct	Not described	Adenocarcinoma
Futami *et al*[15]	F	40	Acute pancreatitis	Inferior CBD	Sphincterotomy	Adenoma
Present case	F	40	Acute cholangitis	Distal CBD	Whipple	Adenoma

CBD: Common bile duct.

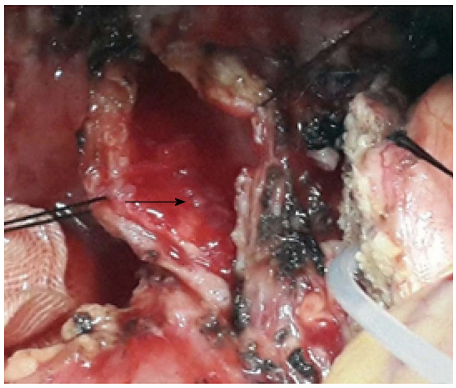

Figure 5 Numerous polyps fill the middle and distal common bile duct.

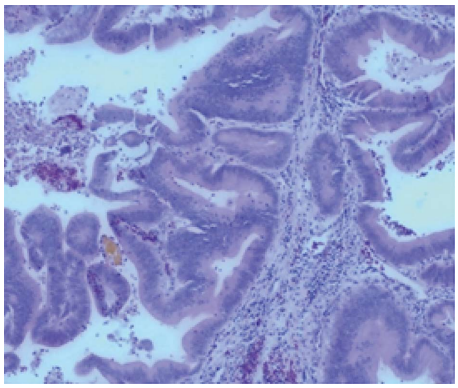

Figure 6 Common bile duct masses are tubular adenomas (HE, × 10).

GS who presented with abdominal pain, acute cholangitis or pancreatitis because of adenomas[12,15] or carcinoma[11,13,14] of the CBD. Previous cases[11-15] in the literature with FAP associated with adenomas in the CBD are summarized in Table 1.

Treatments for adenomas of FAP in the CBD include local excision and radical resection. Local excision is recommended for benign lesions of the inferior CBD[16], but its major problem is high risk of recurrence[3]. Therefore, complete resection of adenomas will not only improve the symptomatology but also prevent the development of carcinoma. Surgical resection is necessary when adenomas have malignant potential or when there are complications[2,7,17]. The present patient had many adenomas in the stomach, duodenum and CBD, and the Whipple procedure is the best choice for her. The Whipple procedure for advanced duodenal and ampullary adenomatosis in FAP is a safe and efficient therapeutic option[2,7]. For the patients in whom complete resection is possible, the long-term prognosis can be good.

It is worth mentioning that our patient firstly presented with biliary obstruction rather than those adenomas in the colon and rectum. We recommend that the patients with FAP or GS undergo endoscopic surveillance of the upper gastrointestinal tract and ultrasonography of the hepatobiliary system, especially those patients with elevated liver enzyme.

COMMENTS

Case characteristics

A case of familial adenomatous polyposis (FAP) with adenomas of the common bile duct (CBD) presented with acute cholangitis.

Clinical diagnosis

The patient was initially admitted for acute cholangitis.

Laboratory diagnosis

Liver function tests showed that the total bilirubin was 1.0 mg/dL, serum amylase 145 IU/L, ALT 168 U/L, AST 88 U/L, GGT 858 U/L, and alkaline phosphatase 520 U/L.

Imaging diagnosis

Preoperative magnetic resonance cholangiopancreatography indicated numerous masses in the CBD and the greatest diameter of the CBD was up to 1.7 cm.

Pathological diagnosis

Microscopic examination of the ampullary and distal CBD polypoid lesions indicated adenomas with high-grade intraepithelial neoplasia.

Treatment

A radical Whipple procedure was performed.

Related reports

Only 2 cases of adenomas of the common bile duct have been reported in the literature in patients with FAP or Gardner's syndrome.

Experiences and lessons

The patient firstly presented with biliary obstruction rather than those adenomas in the colon and rectum. The authors recommend that the patients with FAP or Gardner's syndrome undergo endoscopic surveillance of the upper gastrointestinal tract and ultrasonography of the hepatobiliary system, especially those patients with elevated liver enzyme.

Peer-review

The contents are interesting, but there is the part that does not make the appearance of the article. There is the need to correct.

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