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Case Report

CASE REPORT

Remnant cystic duct adenocarcinoma presenting as gastric outlet obstruction

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Abstract

Only a few case reports of remnant cystic duct carcinoma exist. The presented case of remnant cystic duct carcinoma with invasion to pylorus and bulbus of duodenum leading to gastric outlet obstruction was the first of its kind. We reviewed all cases of remnant cystic duct carcinoma that we found in the literature and summarized its definition, presentation, extent of invasion and clinical outcome after operation. The diagnosis can be difficult due to the rarity of disease, locally advanced nature of disease and distorted postoperative anatomy. A high index of suspicion can increase the likelihood of a preoperative diagnosis.

CASE PRESENTATION

A 54-year-old Chinese gentleman presented with recurrent vomiting for 2–3 days. He had a medical history of laparoscopic cholecystectomy, performed 18 years ago for symptomatic gallstones. Pathology was negative for malignancy. History of presenting illness was negative for diarrhoea, abdominal pain, tarry stool, weight loss or dizziness. Since the cholecystectomy, he had no symptoms of post-cholecystectomy syndrome.

On examination, he was stable with a soft abdomen and no peritoneal signs. Succussion splash test was positive. There was no groin hernia detected, and per-rectal examination was unremarkable. Abdominal X-ray showed a distended gastric bubble and no dilated bowel. Initial blood tests showed leukocytosis up to 15×10^9/l but haemoglobin, platelets, liver and renal function were normal. He was diagnosed with gastric outlet obstruction, and nasogastric tube was inserted for gastric drainage.

After a prolonged period of fasting, oesophagogastroduodenoscopy was done and showed extrinsic compression at the pylorus. Biopsy taken from pylorus was negative for mucosal malignancy. Computed tomography showed eccentric mural thickening in the gastric pylorus close to cystic duct stump with loss of intervening fat plane, worrisome of neoplastic process (Fig. 1).

Laparotomy was arranged. Intraoperatively, a tubulo-cystic structure was found adhered to thickened pyloric and duodenal wall. The tubulo-cystic structure was initially thought to be the common bile duct. It was confirmed to be the dilated remnant of cystic duct after tracing and dissecting to porta hepatica. The common hepatic duct and common bile duct were uninvolved. The cystic duct stump was divided at the T-junction with the common bile duct, a distal gastrectomy with tumour excision done and a Roux-en-Y gastrojejunostomy anastomosis was performed.

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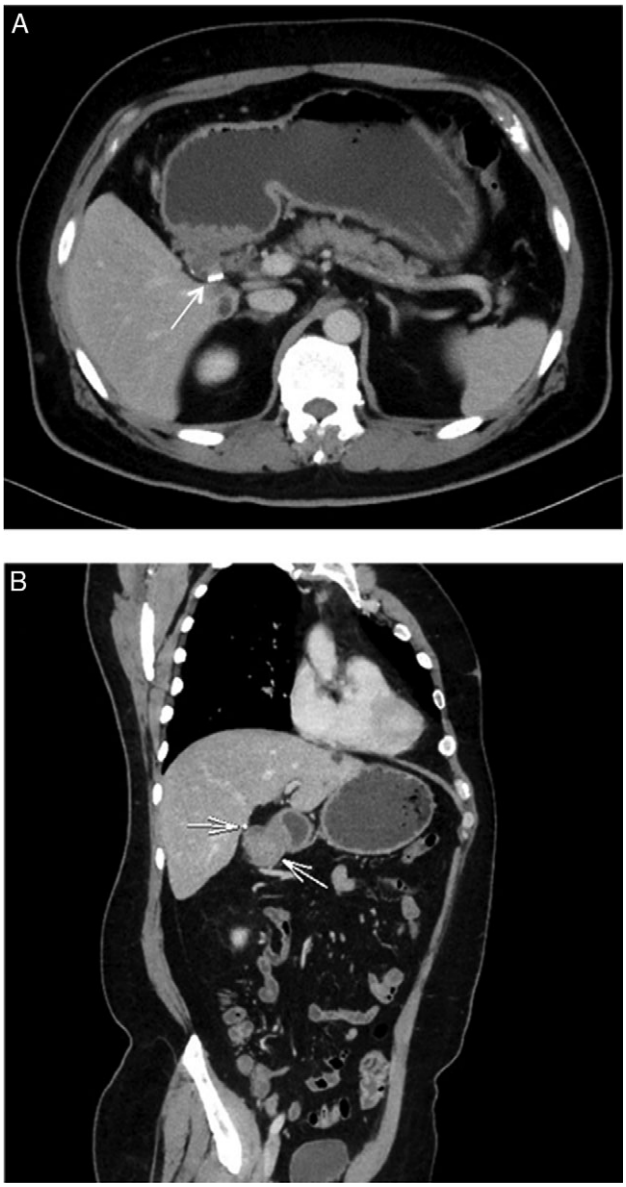

Figure 1: Preoperative contrast abdominal computed tomography scan showing hyperdense clips from previous cholecystectomy and an eccentric heterogenously hypoenhancing mural thickening at the gastric pylorus with loss of intervening fat plane with the adjacent cystic duct stump, suggestive of tumour arising from the remnant cystic duct invading into the pylorus and causing gastric outlet obstruction. The common bile duct and common hepatic duct were spared. (**A**) White arrow indicating cystic duct clip; (**B**) white arrows indicating neoplastic lesion.

His postoperative recovery was uneventful. He tolerated diet well and was fit for discharge on Day 9. His postoperative liver function was normal.

Pathological examination showed a moderately differentiated adenocarcinoma developed in cystic duct with invasion into mucosa of proximal duodenum and submucosa of pylorus, TNM pT3 pN0 (AJCC, 7th edition). The cystic duct was tortuous and dilated, measuring 25 mm long and 5–15 mm in diameter. It looked like a multicystic lesion adhered to gastroduodenal serosa. Only after histologic examination of the entire lesion, its cystic duct nature was proven (Fig. 2). Lymphovascular invasion was detected, but there was no nodal metastasis and cystic duct margin was clear. He was given chemotherapy with capecitabine and radiotherapy.

He remained asymptomatic post-operation but on follow-up, he was found to have rising tumour marker CA19-9. positron emission tomography–computed tomography scan 8 months post-operation revealed new hypodense lesions with mild activity in both lobes of the liver, suspicious of hepatic metastases. There was no obvious activity seen over the operative site. He continues to be followed up by surgical and oncology departments.

DISCUSSION

Primary cystic duct carcinoma is a rare disease. Rarer still are cases of carcinoma arising from a remnant cystic duct after cholecystectomy. It has been reported to occur up to 20 years post-cholecystectomy [1].

The presentation of primary cystic duct carcinoma can differ from remnant cystic duct carcinoma. Primary cystic duct carcinoma usually presents either with upper abdominal pain and a palpable mass in the right upper quadrant or with obstructive jaundice [2]. The abdominal pain and mass is due to gall bladder hydrops or cholecystitis secondary to cystic duct narrowing by the tumour [3]. Obstructive jaundice implies invasion to the biliary system.

Presentation of remnant cystic duct carcinoma can be asymptomatic (such as submucosal mass in duodenum or pylorus or incidental finding on computed tomography scan) or present as biliary obstruction or upper abdominal pain. This is the first report of remnant cystic duct carcinoma presenting with gastric outlet obstruction with invasion to the pylorus and proximal duodenum. The anatomical relations of the cystic duct make it easy to see how a remnant cystic duct carcinoma can invade to the biliary tract, stomach, duodenum [1, 4] and hepatic/transverse colon [5].

While ultrasound, computed tomography and cholangiography can delineate the disease extension and even sometimes diagnose cystic duct carcinoma, the definitive diagnosis is a histopathological one. A new working definition of cystic duct carcinoma was proposed by Ozden [6]: a gall bladder tumour, of which the geometric centre of the tumour being the cystic duct [6]. To rule out local recurrence of inadequately managed gall bladder carcinoma, Noji defined remnant cystic duct carcinoma as occurring more than 5 years post-cholecystectomy [5].

Even then, diagnosis of remnant cystic duct carcinoma can be difficult. Postoperative changes can render the anatomical location and histology of the remnant cystic duct to be altered, like in the presented case. In locally advanced cases, there is an issue of distinction of cancer arising from the remnant cystic duct, as opposed to arising from the common hepatic duct or the common bile duct.

The operative management of primary and remnant cystic duct carcinoma also differs. Primary cystic duct carcinoma can usually be managed with combined excision of the gall bladder, extrahepatic bile ducts and extended lymph node dissection, including the hepatoduodenal, parapancreatic and para-aortic lymph nodes [3]. There are no guidelines for operative management of remnant cystic duct carcinoma. The exact operation should be individualized to each patient. Previous cases of remnant cystic duct carcinoma have been managed with excision

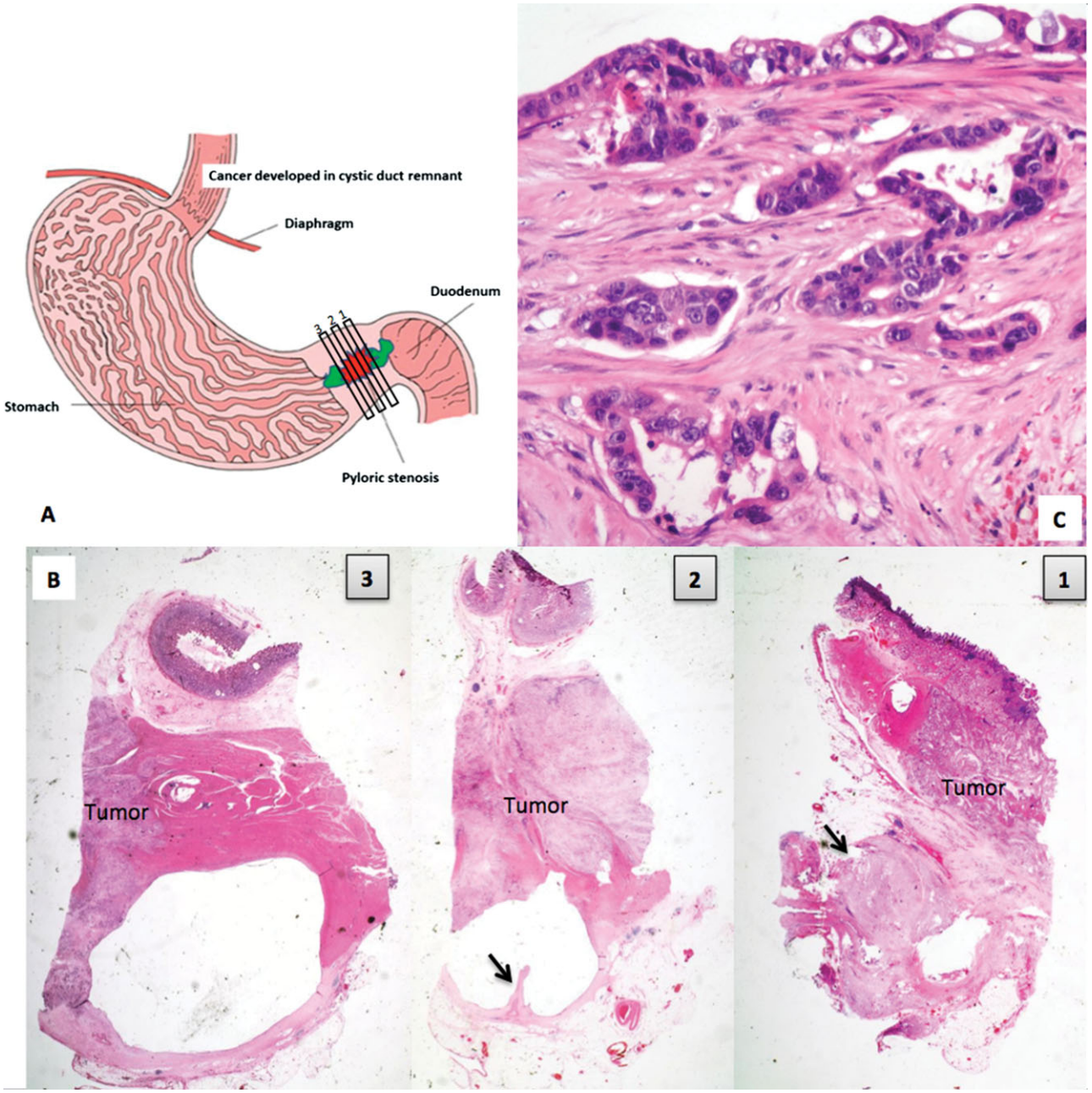

Figure 2: (**A**) Schematic diagram depicting dilated cystic duct stuck on posterior wall of pylorus and D1. Corresponding tissue blocks of 1, 2 and 3 were illustrated in (**B**). Tissue Block 1 showed cystic duct stump where traumatic neuroma (arrow) was seen. Tissue Blocks 2 and 3 revealed the cystic duct was dilated and incorporated into gastric wall with loss of intervening tissue plane. Spiral valve of Heister was discernible in Tissue Block 2 (arrow). (**C**) Dysplasia and invasive malignancy developed on epithelium of dilated cystic duct.

of the remnant cystic duct +/- extrahepatic bile ducts, resection of tumour from the organ of invasion e.g. liver/stomach/transverse colon +/- lymphadenectomy +/- reconstruction. Whipple operation accompanied by resection of extrahepatic bile duct has been performed with success in cases with duodenal involvement (Table 1).

In this case, there was only invasion to the pylorus without evidence of involvement of the common bile duct. Intraoperatively, the decision was to divide the cystic duct at the T-junction and perform distal gastrectomy. In retrospect, a distal gastrectomy, extrahepatic bile duct excision and pancreaticoduodenectomy with extended lymphadenectomy may have been an oncologically safer option.

In conclusion, remnant cystic duct carcinoma is a rare disease, diagnosed when the geometric centre of the tumour is the cystic duct and occurs more than 5 years post-cholecystectomy. The diagnosis can be difficult due to its rarity, distorted postoperative anatomy and in locally advanced disease. Operative management should be individualized, and one can take reference to previous cases.

Table 1 Case reports of remnant cystic duct carcinomas in the literature

Age/sex	Reason for cholecystectomy	Duration after cholecystectomy	Presentation	Author/year/country	Operation	Histopathology	Outcome	Notes
45/M	Cholelithiasis	20 years	Incidental finding of duodenal submucosal tumour on OGD	Eum *et al.* 2008 [1] (Korea)	Total excision of extrahepatic bile duct and remnant cystic duct	Poorly differentiated adenocarcinoma extending to CBD and duodenum; no lymph node or vascular invasion	Well at 6 months	
54/M	Symptomatic gallstones	18 years	Gastric outlet obstruction	Present case	Distal gastrectomy with tumour excision, cystic duct divided at CBD T-junction, Roux-en-Y gastrojejunostomy	Moderately differentiated adenocarcinoma with invasion into submucosa of gastric pylorus and mucosa of D1	Recurrence with bilobar liver metastases after 8 months	
46/F	Papillary carcinoma of gall bladder limited to mucosa with clear cystic duct margin	17 years	Epigastric pain; raised CA19-9	Kurata *et al.* 2009 [11] (Japan)	Left hepatic lobectomy and pylorus preserving pancreaticoduodenectomy	Pedunculated polypoid tumour in cystic duct with widespread dysplasia in bile duct and synchronous intrahepatic cholangiocarcinoma and common bile duct carcinoma	Recurrent metachronous intrahepatic cholangiocarcinoma 7 years later	
62/F	Cholelithiasis	15 years	Upper abdominal pain, hepatic dysfunction and obstructive jaundice	Noji *et al.* 2003 [5] (Japan)	Extended lymphadenectomy, transverse colectomy, en bloc resection of right hepatic lobe, caudal lobe and bile duct	Moderate to poorly differentiated tubular adenocarcinoma with invasion into transverse colon	Well at 6 months	
74/F	Acute cholecystitis with gallstones	10 years	Right upper quadrant pain, nausea, vomiting	Do *et al.* 2014 [3] (Korea)	Complete excision of the remnant cystic duct, wedge segment IVb and V and lymphadenectomy	2 × 1 cm thickened remanant cystic duct wall containing adenocarcinoma	Well at 1 year	Residual/recurrent stones in remnant cystic duct 2 years after cholecystectomy
69/F	Cholelithiasis	7 years	Incidental finding of duodenal submucosal tumour on OGD	Yasuda and Kanamiya 2010 [4] (Japan)	Bile duct excision, distal gastrectomy, duodenectomy and Roux-en-Y reconstruction	11.8 cm tumour of remnant cystic duct with invasion into D1	Recurrence 1 year later	
55/F	Symptomatic cholelithiasis	5 years	Abdominal wall mass at site of cholecystectomy scar	Bhuiya *et al.* 1997 [7] (Japan)	Right hepatic lobectomy with en bloc resection of the caudate lobe, extrahepatic bile duct and right portal vein and abdominal wall tumour excision	Remnant cystic duct carcinoma with invasion into common hepatic duct and abdominal wall with lymph node metastasis	Died 16 months after the operation due to distant metastasis to chest wall and lung	
75/F	Incidental pT1 gall bladder carcinoma with clear cystic duct margin	2 years	Recurrence detected by computed tomography	Horiguchi *et al.* 2012 [8] (Japan)	Subtotal gastrectomy, pancreaticoduodenectomy and hepatoduodenal ligament lymphadenectomy	Papillary adenocarcinoma with invasion to common hepatic duct and CBD	Not mentioned	
57/F	Mild chronic cholecystitis with gall bladder swelling and no stone	15 months	Deranged liver function and obstructive jaundice	Fujii *et al.* 2000 [9] (Japan)	Total resection of extrahepatic bile ducts, pancreaticoduodenectomy	Remnant cystic duct well-differentiated adenocarcinoma adhered to D1	Well at 15 months	Possibility of recurrent of gall bladder cancer cannot be excluded
70/M	Cholelithiasis	6 months	Abdominal pain, fever, jaundice	Gabata *et al.* 2003 [10] (Japan)	Total resection of extrahepatic bile ducts, pancreaticoduodenectomy	Remnant cystic duct carcinoma with widespread invasion along CBD	Not mentioned	

Remnant cystic duct carcinoma is defined as arising more than 5 years post-cholecystectomy, which would exclude three cases by Fujii, Horiguchi and Gabata.
D1, first part of duodenum; CBD, common bile duct.

CONFLICT OF INTEREST STATEMENT

None declared.

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