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 $15 \times 10^9/l$ 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.
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 is in 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.
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 post-operative anatomy and in locally advanced disease. Operative management should be individualized, and one can take reference to previous cases.
| 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 | Penduculated 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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