

## Resected cystadenoma of the common bile duct

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### Abstract

Biliary cystadenoma in the extrahepatic bile ducts is a very rare tumor. A 62-year-old woman with jaundice was admitted to our hospital. Imaging studies revealed a 4-cm cystic lesion around the hepatic hilum, compressing the common bile duct (CBD). When laparotomy was performed, a cystic tumor was detected in the hepatic hilum, filling the lumen of the CBD. Bile duct resection that included the tumor was performed, followed by biliary reconstruction. Microscopically, the cyst wall was lined by a single layer of cuboidal epithelial cells, covering an ovarian-like stroma. The degree of atypia was low and warranted the diagnosis of cystadenoma.

**Key words** Cystadenoma of the common bile duct · Biliary cystadenoma · Obstructive jaundice

### Introduction

Biliary cystadenomas are rare neoplasms, usually occurring in the liver, and less frequently in the extrahepatic bile ducts.<sup>1–7</sup> We present a patient with obstructive jaundice caused by cystadenoma of the common bile duct.

### Case report

A 62-year-old Japanese woman with jaundice was admitted to Kochi Municipal Central Hospital in October 2002. Laboratory data revealed elevated levels of serum aspartate aminotransferase (258 IU/l; normal range, 9–37), alanine aminotransferase (302 IU/l; 4–46), alkaline phosphatase (2783 IU/l; 95–305), gamma glutamyl transpeptidase (1129 IU/l; 8–45), lactate dehydrogenase (521 IU/l; 211–475), total bilirubin (17.8 mg/dl; 0.2–1.1), and direct bilirubin (12.2 mg/dl; 0.1–0.7). The serum

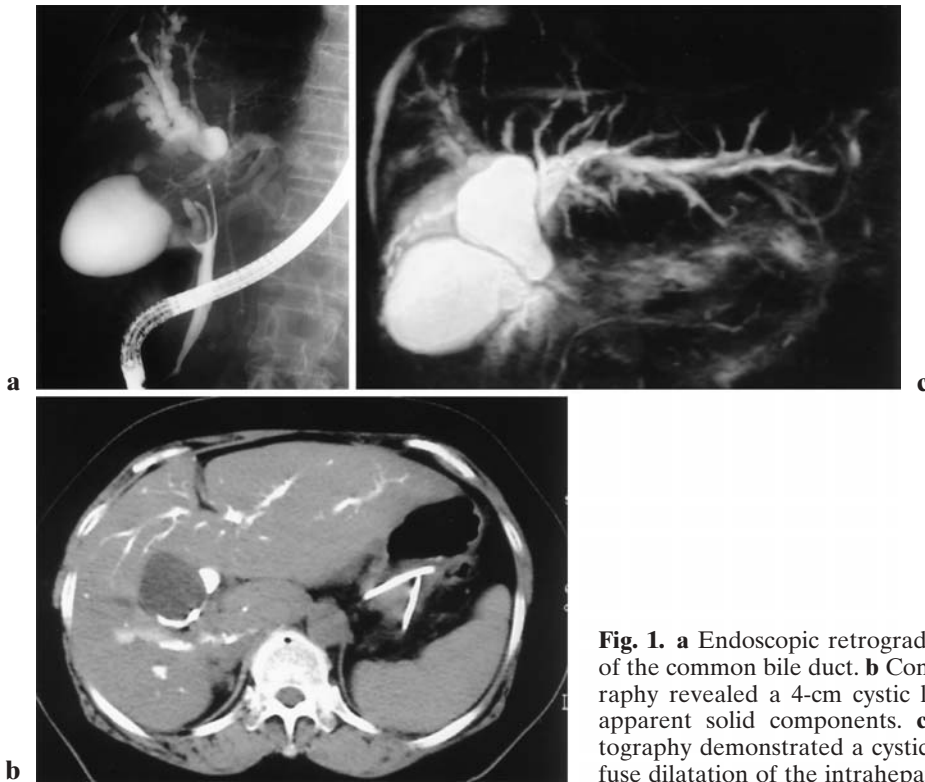
carbohydrate antigen (CA) 19-9 concentration was elevated (140.1 U/ml), but carcinoembryonic antigen (CEA) was within the normal range (1.9 ng/ml). Ultrasonography (US) showed diffuse dilatation of the intrahepatic bile ducts (IHBD) in both lobes of the liver. Endoscopic retrograde cholangiography (ERC) demonstrated stenosis of the common bile duct (CBD; Fig. 1a), and endoscopic nasal biliary drainage (ENBD) was then performed. Computed tomography (CT) scan after ERC revealed a 4-cm cystic lesion around the hepatic hilum, without apparent solid components (Fig. 1b). Magnetic resonance cholangiopancreatography (MRCP) demonstrated a cystic lesion around the hepatic hilum and diffuse dilatation of the IHBD (Fig. 1c). These findings suggested that the obstructive jaundice was caused by the cystic lesion compressing the hepatic hilum. ENBD was not effective for the jaundice, and percutaneous transhepatic cyst drainage was performed. Clear, yellow cystic fluid was aspirated. Results of cytology of the cystic fluid were negative. Despite the ENBD and cyst drainage, the serum total bilirubin concentration remained at about 10 mg/dl.

In November 2002, laparotomy was performed to resect the cystic lesion and reconstruct the bile duct. The cystic mass was detected in the hepatic hilum, filling the lumen of the CBD. The distal common bile duct was divided superior to the pancreatic head. Cholangioscopy was then performed, and there were no changes in the mucosa of the bile duct. The proximal bile duct was divided 1 cm superior to the cystic mass after histopathological confirmation of negative tumor margins. Biliary reconstruction was carried out by bilateral hepaticojejunostomy. The postoperative course was uneventful, and the jaundice gradually abated.

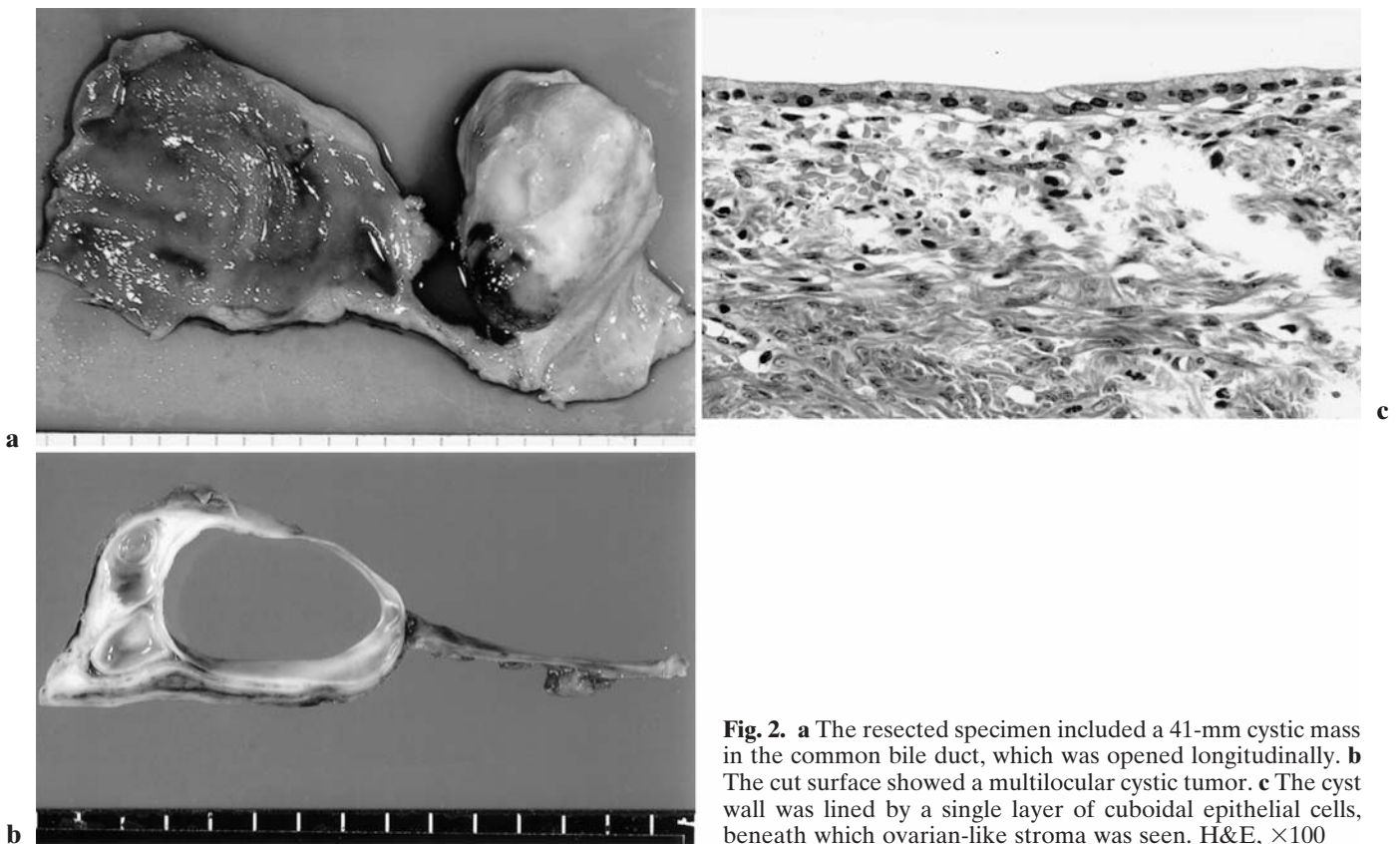
The resected specimen included a 41-mm multilocular cystic mass in the CBD (Fig. 2a,b). Microscopically, the cyst wall was lined by a single layer of cuboidal epithelial cells, beneath which an ovarian-like stroma was seen (Fig. 2c). Spindle cells in the ovarian-like

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**Fig. 1.** **a** Endoscopic retrograde cholangiography demonstrated stenosis of the common bile duct. **b** Computed tomography scan after cholangiography revealed a 4-cm cystic lesion around the hepatic hilum, without apparent solid components. **c** Magnetic resonance cholangiopancreatography demonstrated a cystic lesion around the hepatic hilum and diffuse dilatation of the intrahepatic bile ducts



**Fig. 2.** **a** The resected specimen included a 41-mm cystic mass in the common bile duct, which was opened longitudinally. **b** The cut surface showed a multilocular cystic tumor. **c** The cyst wall was lined by a single layer of cuboidal epithelial cells, beneath which ovarian-like stroma was seen. H&E,  $\times 100$

stroma were positive for progesterone receptors. The degree of atypia was low and warranted the diagnosis of cystadenoma. Fourteen months after resection, the patient was doing well without any symptoms.

## Discussion

Cystadenomas of the CBD are very rare tumors that are seen in middle-aged women.<sup>5,6</sup> In their review of 52 biliary cystadenomas, Devaney et al.<sup>5</sup> reported that 43 were located in the liver, while only 3 were in the CBD. The characteristic microscopic findings of the cystadenomas include a single-layer lining of columnar or cuboidal epithelial cells with a submucosal layer consisting of a cellular band comprising spindle cells reminiscent of ovarian stroma.<sup>6</sup> The spindle cells of the ovarian-like stroma are usually positive for progesterone and estrogen receptors.

Cystic lesions around the hepatic hilum can be confused with choledochal cyst, liver cyst, and retention cysts of periductal glands. Choledochal cysts are the most common congenital anomaly of the extrahepatic biliary tree, and ERC is a useful method for differential diagnosis. Retention cysts of periductal glands are usually multiple, asymptomatic, and smaller than cystadenomas.<sup>8</sup> Their walls lack the ovarian-like stroma and may contain inflammatory cells. It has been reported that liver cysts in the hepatic hilum have caused obstructive jaundice.<sup>9,10</sup> Although our patient was diagnosed with these cysts before laparotomy, liver cysts causing obstructive jaundice are usually large in size.

Biliary cystadenomas are considered to be premalignant lesions. Despite foci of dysplasia being present in 7 of the above reviewed 52 cystadenomas, malignant transformation has not been reported in tumors of the

CBD.<sup>5</sup> Cystadenomas of the bile ducts were reported to recur following incomplete resection, and should therefore be treated by wide surgical resection.

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