ACUTE ABDOMINAL PAIN DUE TO BILIARY CYSTADENOMA

K. Van den Bergh, K. Op de beeck1

We present a case of a biliary cystadenoma, a rare benign cystic tumor arising in most cases of the intrahepatic bile ducts. A 30-year old woman presented with abdominal pain in the right upper quadrant for about 10 days with increasing severity. Radiological evaluation by means of abdominal ultrasound and CT-scan revealed a multilocular cystic lesion in the right liver lobe, not present on abdominal ultrasound performed 10 years before. The diagnosis of a biliary cystadenoma was proposed and complete surgical resection of the mass was performed.

Key-word: Liver neoplasms, diagnosis.

Biliary cystadenomas are rare, usually slow growing, multilocular cystic tumors that represent less than 5% of intrahepatic cystic masses of biliary origin (1). They are generally intrahepatic (85%) and occur less commonly in the extrahepatic biliary system and gallbladder. Cystadenomas occur predominantly in middle-aged women (2). The tumor is well defined by a fibrous capsule and lined by cuboidal to columnar epithelium. Two histological variants are recognized: the more common mucinous type and the rare serous type (3). The clinical presentation is variable depending on the size and location of the cyst. Abdominal pain, obstructive jaundice, palpable mass, nausea and vomiting are common signs and symptoms. Occasionally, the tumor is incidentally discovered (2).



 $\it Fig.~1.$ — Abdominal ultrasound reveals a well defined, septated and thin-walled cystic mass. The cyst fluid contains some low-level echoes.

Case report

A 30-year-old woman was admitted to the emergency department with complaints of abdominal pain in the epigastric area and the right upper quadrant for 10 days with increasing severity. Physical examination revealed a tender upper abdomen. Her medical history was unremarkable. Laboratory data on admission were within normal limits.

Abdominal ultrasound (US) showed a well defined 10-cm, septated and thin-walled cystic (anechoic) mass with enhanced through transmission, involving segment 4 of the liver (Fig. 1). The cyst fluid contained some low-level echoes. The gallbladder was normal. Contrast-enhanced CT-scan confirmed the presence of a multilocular cystic mass in segment

4 of the liver, with mean attenuation values of 10HU (Fig. 2 A). The internal septations and the thin regular wall showed slight enhancement (Fig. 2B). A fluid-fluid level was present within the lower portion of the cystic mass. No mural nodularity was identified. Intrahepatic bile ducts showed no dilatation. There was mass effect of the lesion on surrounding structures.

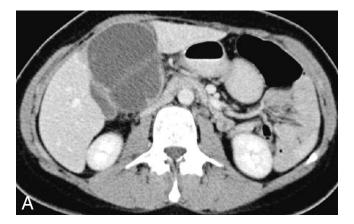
The diagnosis of a biliary cystadenoma was proposed. The differential diagnosis included hydatid disease, complicated hepatic cyst and abscess. Because of the unclear etiology and severe pain an enucleation of the lesion was set up. Finally complete surgical resection of the mass was performed. During surgery it was remarkable that the lesion was glued with the omentum and the stomach, due to an inflammatory reaction. Microscopic examination of the specimen confirmed the diagnosis of a biliary cystadenoma, there were no signs of malignancy.

Discussion

Biliary cystadenomas are benign cystic neoplasms lined by mucinsecreting epithelium. The fluid within the lesion can be proteinaceous, mucinous and occasionally gelatinous, purulent or hemorrhagic due to trauma (1).

Biliary cystadenomas are rare, usually slow growing, multilocular cystic tumors that represent less than 5% of intrahepatic cystic masses of biliary origin. Eighty-five percent arise from the intra-hepatic biliary ducts, the remaining 15% are extrahepatic. They are usually solitary and located in the right lobe of the liver and measure from 1.5 cm to 25 cm in diameter. They occur predominantly in middle-aged women

From: 1. Department of Radiology, University Hospitals Leuven, Leuven, Belgium. Address for correspondence: Dr K. Van den Bergh, Department of Radiology, KUL University Hospitals Leuven, Herestraat 49, B-3000 Leuven, Belgium. E-mail: katrien.vandenbergh@uzleuven.be



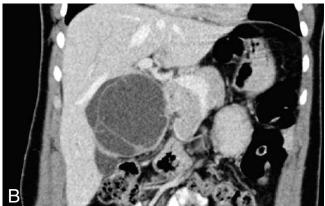


Fig. 2. — A. Contrast-enhanced CT-scan confirms the presence of a multilocular cystic mass in segment 4 of the liver, with mean attenuation values of 10HU. B. The internal septations and the thin regular wall show slight enhancement.

(mean age, 38 years). Although biliary cystadenomas are benign tumors, they may recur after excision and have potential to develop into biliary cystadenocarcinoma. Symptoms are usually related to the mass effect of the lesion and consist of intermittent pain or biliary obstruction (1, 2).

At US, a biliary cystadenoma appears as a unilocular or multilocular cyst with enhanced through transmission. Acoustic shadowing may be present from septal or wall calcification. The cyst fluid may contain low level echoes from blood products, mucin, or proteinaceous fluid. Serous and bilious fluid is generally anechoic. Echogenic mural nodules and papillary projections may be present (2, 4).

CT depicts a low-density, well defined cystic mass with internal septa and a thick fibrous capsule. The internal septa and capsule usually shows contrast enhancement. The CT attenuation of the fluid component varies depending on the fluid content. Higher attenuation may indicate recent hemorraghe. Tumor nodules and papillary excrescences can be present and appear as soft-tissue attenuation nodules that typically enhance with contrast material (2). These tumor nodules and papillary excrescences are seen more commonly in biliary cystadenocarcinoma than in cystadenoma, although they have also been reported in cystadenomas without frank malignancy.

Although the absence of mural nodularity is suggestive of cystadenoma (5). CT may also reveal septal and capsular calcifications (1, 4). Unfortunately there are no specific imaging features that permit reliable differentiation of biliary cystadenoma from cystadenocarcinoma (2).

The major advantage of MRI is that it can accurately define the internal architecture of the lesion and specify the fluid content because of the better soft tissue contrast resolution compared with CT. The MR imaging appearance of a cystadenoma varies depending on the protein content of the fluid and the presence of an intracystic soft-tissue component. In general, the (mucinous) fluid within a cystadenoma appears hypointense on T1-weighted images and markedly hyperintense on T2weighted images. On T1-weighted images, the signal intensity may change from hypointense to hyperintense as protein concentration increases. Also, on T2-weighted images the signal intensity may decrease from markedly hyperintense to hypointense as protein concentration and viscosity increases. Similar changes may be provoked by internal hemorraghe, however, fluidfluid levels are usually found in these cases (1, 3).

The differential diagnosis principally includes hepatic echinococcal cyst, hepatic abscess and a hemorrhagic bile duct cyst. In rare cases, other neoplasm such as mesenchymal hamartoma, undifferentiated embryonal sarcoma, cystic hepatocellular carcinoma and cystic metastasis may have similar appearance (4). Correlation of imaging findings with patient age and clinical data may be helpful in the differential diagnosis.

Because cystadenoma represents the premalignant form of biliary cystadenocarcinoma, resection is always recommended (5).

References

- Mortelé K.F., Ros P.R.: Cystic focal liver lesions in the adult: differential CT and MR imaging features. Radiographics, 2001, 21: 895-910.
- Levy A.D., Murakata L.A., Abbott R.M., Rohrmann C.A: Benign tumors and tumorlike lesions of the gallbladder and extrahepatic bile ducts: Radiologic-pathologic correlation. Radiographics, 2002, 22: 387-413.
- Frahm C., Zimmermann A., Heller M., Brossmann J.: Uncommon presentation of a giant biliary cystadenoma: correlation between MRI and pathologic findings. J Mag Res Imag, 2001, 14: 649-652.
- Buetow C.P., Buck J.L., Pantongrag-Brown L. et al.: Biliary cystadenoma and cystadenocarcinoma: Clinicalimaging-pathologic correlation with emphasis on the importance of ovarian stroma. *Radiology*, 1995, 196: 805-810.
- Anderson S. W., Kruskal J.B., Kane R. A.: Benign hepatic tumors and iatrogenic pseudotumors. Radiographics, 2009, 29: 211-229.