Spontaneous rupture of a recurrent hepatic cystadenoma

Hakim Elfadili, Anass Majbar, Fouad Zouaidia, Naoufal Elamrani, Farid Sabbah, Mohamed Raiss, Najat Mahassini, Abdelmalek Hrora, Mohamed Ahallat

INTRODUCTION
Biliary cystadenomas are rare benign tumors, although prone to malignant degeneration. Clinical and imaging features are non specific. Consequently, they are preoperatively misdiagnosed in 50% to 70% of cases, commonly resulting in delayed and inaccurate treatment[1].

We report a case of spontaneous rupture of a recurrent biliary cystadenoma.

CASE REPORT
A 32-year-old woman with a symptomatic hepatic cyst was referred to our department. Five months earlier, she had been operated on in another hospital for suspicion of hydatid cyst of the liver in the fourth segment. A conservative treatment was carried out (unroofing). Histological examination of the cystic wall revealed a biliary cystadenoma with mesenchymal stroma. 4 mo later, she presented right hypochondrium pain. Abdominal US revealed a heterogeneous mixed-echoic tumor in the fourth hepatic segment. An abdominal CT scan revealed a 10 cm × 20 cm cyst with septations in liver segments 3, 4 and 5 (Figure 1). Surgical treatment was decided for recurrent hepatic cystadenoma. 2 d before surgery, the patient presented with severe abdominal pain. Abdominal ultrasound showed diffuse peritoneal fluid. The patient had an emergency laparotomy for cyst rupture. After bilateral sub-costal incision, we found 2 liters of serous fluid in the abdomen and exploration showed a huge perforated cyst in the third, fourth and fifth hepatic seg-
Elfadili H et al. Spontaneous rupture of a recurrent hepatic cystadenoma

DISCUSSION

Cystadenoma is a benign tumor, supposedly originating in intrahepatic (and more rarely extrahepatic) embryonic tissue precursors of biliary epithelium. With its malignant counterpart (cystadenocarcinoma), it accounts for less than 5% of all cystic lesions of the liver. These tumors usually present in middle aged women with a mean age of 50 years and have a great variability in size, ranging from 1.5 cm to 30 cm.

The majority of patients are asymptomatic, but in the case of large tumors, they may present with a palpable mass and cause symptoms such as upper abdomen pain, dyspepsia, anorexia, nausea and fever.

The most widely used diagnosis methods are abdominal ultrasound and computed tomography. Ultrasound will reveal an anechoic mass with sharp demarcations and often with fine internal septations. Computed tomography usually shows a well-defined mass with low-density and internal septa. Its fibrous capsule and internal septations are often visible and help distinguish the lesion from a simple cyst. The presence of mural nodules or wall thickening should trigger the suspicion of cystadenocarcinoma. Unilocular lesions have been reported and are often incorrectly diagnosed, resulting in inadequate therapy. Hydatid cyst is one of the disorders most likely confused with hepatic cystadenoma specially in endemic hydatid disease regions. In our case, the patient had conservative surgical treatment for suspicion of hydatid cyst. Differential diagnosis may sometimes be difficult especially when serology is negative and for type II (cystic lesion with internal septa) and type IV lesions according to OMS Classification. These lesions have thick walls and contain intra cystic material.

Establishment of the correct diagnosis is crucial because application of inadequate treatment, including partial excision or fenestration, results in an unacceptably high rate of recurrence. Surgical resection is the treatment of choice for hepatobiliary cystadenoma because it avoids the risks of recurrence and malignant transformation.

To our knowledge, this is only the third case of spontaneous rupture of hepatobiliary cystadenoma reported in the English literature. This complication is potentially life-threatening and requires an emergency laparotomy.

In conclusion, hepatic cystadenomas are rare and should be expected when radiological imaging studies suggest a multilocular cystic hepatic lesion, especially in women. It can rarely present in an acute setting and rupture is an exceptional complication.

REFERENCES

1. Votanopoulos KI, Goss JA, Swann RP, O'Mahony CA, Jaffe BM, Bellows CF. Massive abdominal distension resulting from a giant hepatobiliary cystadenoma. Am Surg 2009; 75: 438-439

2. Hernandez Bartolome MA, Fuerte Ruiz S, Manzanedo Romero I, Ramos Lojo B, Rodriguez Prieto I, Gimenez Alvira L, Granados Carreño R, Limones Esteban M. Biliary cystadenoma. World J Gastroenterol 2009; 15: 3573-3575
Elfadili H et al. Spontaneous rupture of a recurrent hepatic cystadenoma

3 Del Poggio P, Buonocore M. Cystic tumors of the liver: a practical approach. World J Gastroenterol 2008; 14: 3616-3620
4 Kim HG. [Biliary cystic neoplasm: biliary cystadenoma and biliary cystadenocarcinoma]. Korean J Gastroenterol 2006; 47: 5-14
5 Yu FC, Chen JH, Yang KC, Wu CC, Chou YY. Hepatobiliary cystadenoma: a report of two cases. J Gastrointestin Liver Dis 2008; 17: 203-206
6 Kinoshita H, Tanimura H, Onishi H, Kasano Y, Uchiyama K, Yamaue H. Clinical features and imaging diagnosis of biliary cystadenocarcinoma of the liver. Hepatogastroenterology 2001; 48: 250-252
7 Qu ZW, He Q, Lang R, Pan F, Jin ZK, Sheng QS, Zhang D, Zhang XS, Chen DZ. Giant hepatobiliary cystadenoma in a male with obvious convex papillate. World J Gastroenterol 2009; 15: 1906-1909
8 Lewin M, Mourra N, Honigman I, Fléjou JF, Parc R, Arrivé L, Tubiana JM. Assessment of MRI and MRCP in diagnosis of biliary cystadenoma and cystadenocarcinoma. Eur Radiol 2006; 16: 407-413
9 Ramacciato G, Nigri GR, D’Angelo F, Aurello P, Bellagamba R, Colarossi C, Pilozzi E, Del Gaudio M. Emergency laparotomy for misdiagnosed biliary cystadenoma originating from caudate lobe. World J Surg Oncol 2006; 4: 76
10 International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. Acta Trop 2003; 85: 253-261
11 Jean-François Cadranel JPB. Cystadénome du foie. Hepatogastrology. 1998; 5: 109-114
12 Regev A, Reddy KR, Berho M, Sleeman D, Levi JU, Livingston AS, Levi D, Ali U, Molina EG; Schiff ER. Large cystic lesions of the liver in adults: a 15-year experience in a tertiary center. J Am Coll Surg 2001; 193: 36-45
13 Thomas KT, Welch D, Trueblood A, Sulur P, Wise P, Gorden DL, Chari RS, Wright JK Jr, Washington K, Pinson CW. Effective treatment of biliary cystadenoma. Ann Surg 2005; 241: 769-773; discussion 773-775
14 Chang MS, Chen MC, Chou FF, Sheen-Chen SM, Chen WJ. Spontaneous rupture of hepatobiliary cystadenoma: a case report. Chunggeng Yi Xue Za Zhi 1995; 18: 392-397

S- Editor Zhang HN  L- Editor Hughes D  E- Editor Liu N