Hepatic Intracystic Organizing Hematoma Mimicking Biliary Cystadenocarcinoma in a Patient with Polycystic Liver Disease

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Abstract

Hepatic intracystic hemorrhage is a rare complication of polycystic liver diseases, such as autosomal dominant polycystic kidney disease (ADPKD). A hepatic cyst with mural nodules and septation may suggest the presence of a cystic malignancy, such as biliary cystadenocarcinoma. We herein report a case of hepatic intracystic hematoma with a mural nodule mimicking biliary cystadenocarcinoma in a patient with ADPKD. Hepatic intracystic hemorrhage with a mural nodule is a very rare occurrence. A fat-saturated T1-weighted magnetic resonance image may be useful for making an accurate diagnosis of intracystic hematoma.

Key words: intracystic hematoma, mural nodule, organizing hematoma, PLD, polycystic liver disease, ADPKD

(DOI: 10.2169/internalmedicine.54.4218)

Introduction

Multiple liver cysts in patients with polycystic liver disease (PLD) may be complicated by intracystic hemorrhage and infection. We have encountered cases of hepatic intracystic hemorrhage that are difficult to differentiate from tumors. We herein report the case of a patient with autosomal dominant polycystic kidney disease (ADPKD) and hepatic intracystic organizing hematoma which appeared as a mural nodule in the cyst wall on various images of ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI).

Case Report

A 65-year-old Japanese woman with a history of ADPKD was referred to our hospital to be evaluated for a mural nodule in a hepatic cyst. She had no clinical symptoms, such as either abdominal pain or distension. Physical examination showed the liver to be enlarged. Laboratory examinations did not show either liver dysfunction, coagulopathy, or infectious disease, but the findings did reveal an elevated serum creatinine level (6.05 mg/dL) requiring hemodialysis. The patient’s serum carcinoembryonic antigen (CEA) (5.68 ng/mL) and serum carbohydrate antigen 19-9 (CA19-9) (229.6 U/mL) levels were elevated. One year prior to this presentation, percutaneous transhepatic sclerotherapy was performed to reduce the cyst volume using minocycline hydrochloride. The treatment was performed under US guidance, but the precise cyst location was unclear.

Both US and CT images demonstrated numerous cysts within enlarged kidneys and a liver consistent with ADPKD. An intracystic mural nodule was also observed in the liver that had increased in size since first being identified 6 months previously. Color Doppler US showed the absence of any Doppler signal, while unenhanced CT revealed a slightly hyperdense lesion with 40 Hounsfield Units. On contrast-enhanced CT, the nodule was partially and slightly enhanced, but the majority was not enhanced (Fig. 1, 2). Increased wall thickness of the liver cyst was clearly visualized on contrast-enhanced CT. The intracystic mural nodule

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Received for publication October 3, 2014; Accepted for publication January 13, 2015
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was also seen on MRI. That image revealed hyperintensity with a lower signal intensity center on fat-saturated T1-weighted images and hypointensity with a slightly hyperintense center on T2-weighted images (Fig. 3).

Based on these imaging features, intracystic hematoma of the liver was suspected. However, as the mural nodule was increasing in size, the possibility of a malignant cystic tumor first had to be ruled out. Surgical biopsy was not planned due to a high risk of dialysis related postprocedural comorbidities. As a result, percutaneous needle aspiration and a cutting biopsy was performed under CT guidance using a 20-gauge cutting needle. No bacteria, neoplastic cells, or parasites were detected in the cystic fluid. The CEA and CA19-9 levels in the cystic fluid were both elevated (64.6 ng/mL and 81,351.0 U/mL, respectively). A histopathological examination revealed the mural nodule to be an intracystic organizing hematoma of the liver that was partially organized and included various phases of bleeding (Fig. 4). There was no evidence of any neoplasm. Thereafter, the patient was carefully followed up with US and CT. One year later, the mural nodule was found to have further enlarged and also changed in shape (Fig. 5). Repeated percutaneous
biopsies showed the same histopathological diagnosis. The patient was therefore only followed closely without any particular therapy.

**Discussion**

Intracystic hemorrhage of the liver is a rare complication of PLD with an unknown etiology. Several hypotheses have so far been reported as follows: vessel rupture in the cyst wall caused by a rapid enlargement of the lesion (1), hemangioma, vascular malformation near the cyst wall and direct trauma to the cyst wall (2). We assume that the hemorrhage may repeatedly occur and organize and gradually contribute to the enlargement of the mural nodules (3). In our case, percutaneous aspiration and the injection of a sclerosing agent for liver cysts may have been the cause of these liver lesions. In addition, multiple phases of bleeding noted in the pathological examination indicated that repeated bleeding had occurred in the mural nodule, which had gradually increased in size. This is similar to the phenomena
observed in so-called chronic expanding hematoma. Chronic expanding hematoma, a rare benign condition caused by repeated hemorrhage into hematoma that was first described in 1980 by Reid et al. (4), it can be seen throughout the body and is relatively common in the thoracic cavity. One of its diagnostic criteria is a lack of coagulopathy. In this case, the patient received hemodialysis, but had no coagulopathy.

Intracystic hemorrhage is usually recognized radiographically by a change in the cyst density, the layering of blood within the cyst, or the presence of diffuse irregular hyperechogenicity. Since radiologic findings of intracystic hemorrhage vary greatly according to the hemorrhage phase, it is sometimes difficult to differentiate from biliary cystic neoplasms (5). In such cases, hepatectomy is often performed. US and CT may show abnormal findings, such as a thickened cyst wall, septation, a cystic mass, and mural nodules, similar to those of biliary cystic neoplasms (1, 5-7). The occurrence of intracystic hemorrhage with a mural nodule is very rare. In our review of previous reports, 3 such cases were reported (2, 5, 6). On MRI, a high signal intensity detected by T1-weighted image is very useful for differentiating intracystic hemorrhage from a cystic liver tumor that exhibits a low to homogenous signal intensity (2, 8, 9). The signal intensity of the hemorrhage decreases when blood clots are liquefied (10). In this case, the nodule partially showed a high intensity on the T1-weighted images; thus, an intracystic hemorrhage was suspected.

Intracystic mural nodules may be mildly enhanced on contrast enhanced CT and MRI (2, 6). Contrast-enhanced US imaging may play an important role in making a correct diagnosis of a hemorrhagic cyst since it demonstrates the avascularity of the visualized intracystic structures (11) and microbubbles oozing from the cyst wall into the cystic cavity (12). It should be noted that intracystic bleeding can occur in cystic liver tumors (12-14). In our case, we believe that the mural nodule was visible on US and CT because the blood clot had been organized. In this case, some partial contrast enhancement was seen in the mural nodule, perhaps due to the neovascularization of the organized blood clot that was histopathologically confirmed.

Tumor markers such as CA19-9 and CEA in the serum and cystic fluid have been used to distinguish benign from malignant lesions, but their relevance remains unclear. Horsmans et al. (15) reported the CA19-9 levels in the cystic fluid to be much higher than the upper normal limit for serum, not only in a cystadenoma or cystadenocarcinoma, but also in a benign hepatic cyst, which is not surprising because the epithelial cells of these different cysts all expressed CA 19-9 immunoreactivity. In this report, the serum CA 19-9 levels were elevated in two patients with cystadenoma or cystadenocarcinoma, but the levels remained normal in two other patients with a hemorrhagic simple cyst. Therefore, those findings suggest that the serum CA19-9 levels may be useful in the diagnosis of biliary cystic neo-
plasms. Conversely, Park et al. (16) reported that an elevated serum CA 19-9 level may not be helpful in the preoperative diagnosis of biliary cystic neoplasm. In other reports, elevated serum CA19-9 levels were observed in one patient with an infected simple hepatic cyst complicated by an intracystic hemorrhage (17) and also in another patient with a ruptured hepatic cyst (18). The serum and cystic fluid CEA levels were similar in both entities (15). In the present patient, although the usefulness of the CA19-9 levels in the serum and cystic fluid remains controversial, we could not conclude preoperatively that the hepatic lesion was truly benign due to the elevated serum CA19-9 levels.

Our patient’s PLD was complicated by an intracystic organizing hematoma that was mimicking a cystic neoplasm. The organizing hematoma was ultimately correctly diagnosed using biopsy specimens. Among the various images obtained in this case, a fat-saturated T1 weighted image may be useful for making a diagnosis of intracystic hematoma.

The authors state that they have no Conflict of Interest (COI).

References