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Case Report

Intrahepatic Biliary Dilatation Caused by a Small Simple Hepatic Cyst: Report of a Case

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Biliary obstruction caused by small simple cysts is very rare. We present a case of biliary dilatation caused by a simple cyst with a 4-cm diameter. The patient was a 75-year-old woman referred to our hospital for evaluation of a cystic tumor associated with peripheral biliary duct dilatation in the left segment of the liver. Computed tomography and magnetic resonance imaging showed that the cyst probably communicated with the intrahepatic bile duct. Malignant tumors, including intrahepatic papillary neoplasms of the bile duct, could not be ruled out; therefore, we performed surgery with the patient's consent. Histopathologic examination of the resected liver showed that the cystic lesion was a simple cyst. The finding that even small simple cysts can obstruct the biliary tract is important for the management of cystic lesions of the liver.

Key words: Hepatic cyst – Biliary obstruction – Biliary dilatation

Simple cysts of the liver are usually asymptomatic. However, if the cyst is large, patients may experience abdominal bloating or a dull pain in the upper quadrant of the abdomen. Biliary obstruction caused by a simple cyst is very rare,[1–4] and dilatation of the intrahepatic bile duct in association with tumor lesions usually indicates malignancy.[4–6] With the development of computed tomography and magnetic resonance imaging, it is now possible to accurately differentiate between benign cysts and malignant neoplasms, including intraductal papillary neoplasms of the bile duct. However, it remains difficult to correctly establish the diagnosis preoperatively for all patients with cystic lesions of the

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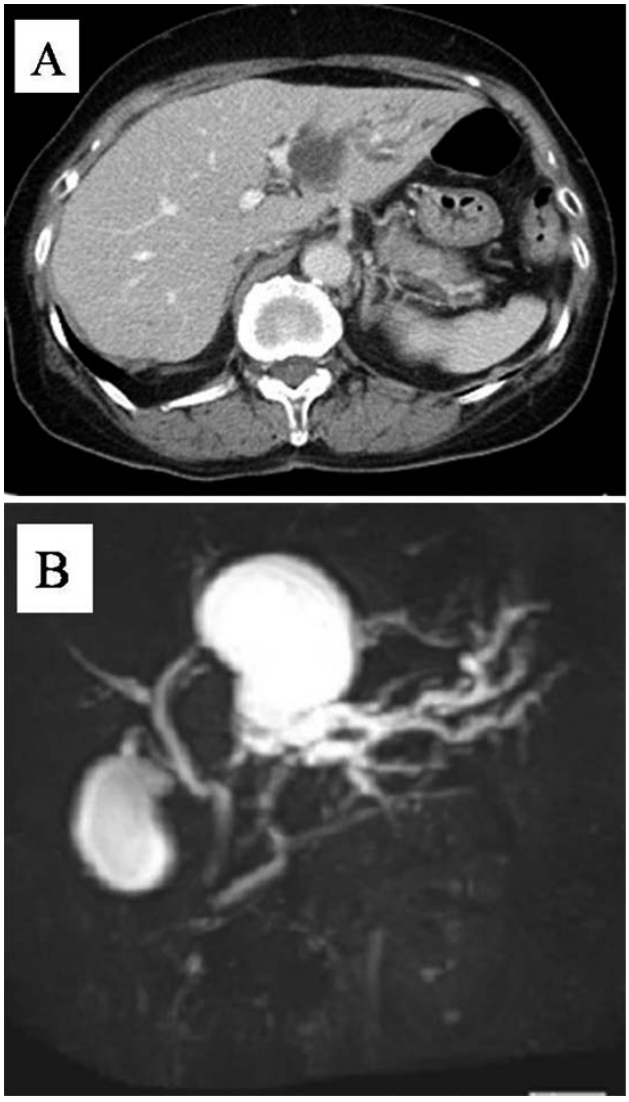

Fig. 1 Images from (A) CT and (B) magnetic resonance cholangiography reveal a cystic lesion in the lateral segment associated with peripheral dilatation of the bile duct.

liver. In this report, we present a case of intrahepatic bile duct dilatation caused by a small simple cyst and discuss the management of cystic lesions of the liver.

Case Report

A 75-year-old woman was referred to our hospital for the evaluation of a cystic tumor of the liver with bile duct dilatation, which had been incidentally detected on ultrasonography. An examination performed in the preceding year had demonstrated the presence of the cystic lesion, but bile duct dilatation had not been identified. Computed tomography (CT) scanning and magnetic resonance imaging (MRI) showed a cystic mass with a 4-cm diameter located in the lateral segment of the liver accompanied by peripheral bile duct dilatation. CT and MRI findings also suggested a communication between the cystic mass and the dilated bile duct (Fig. 1). Endoscopic retrograde cholangiopancreatography revealed that the bile duct was interrupted at the left hepatic duct, but it did not show either a cystic mass or dilatation of the distal portion of the bile duct (Fig. 2). Cytologic examination of bile samples aspirated from the occluded biliary duct did not show any atypical cells. Furthermore, elevation of serum tumor markers was not detected. Given that malignant tumors, including IPNB, could not be ruled out, a left hepatectomy was performed with the patient's consent. Examination of the resected specimen showed that the cyst had compressed the bile duct and that dilatation of the peripheral bile duct began at the point of compression. Moreover, there was no communication between the cyst and the biliary tract (Fig. 3A). Microscopic examination showed that the cyst wall was lined with a single layer of flat epithelial cells and that no signs of malignancy were present (Fig. 3B); the cyst was diagnosed as a simple cyst.

Fig. 2 An image from endoscopic retrograde cholangiopancreatography reveals that the bile duct was interrupted at the left hepatic duct, but there was no evidence of a cystic mass or dilatation of the distal portion of the bile duct.

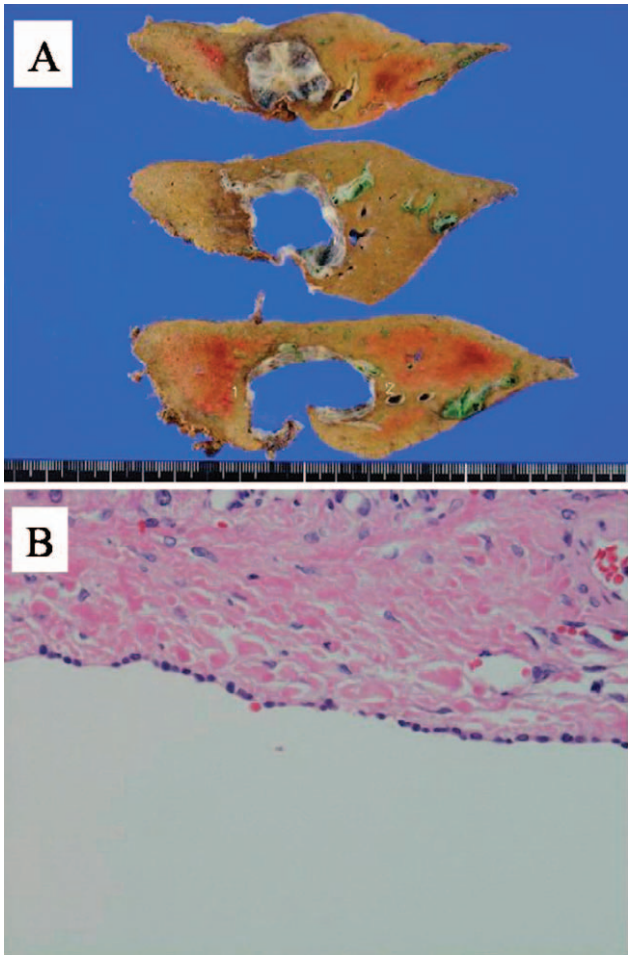

Fig. 3 (A) The resected specimen did not show any evidence of communication between the cyst and the biliary tract. (B) Microscopic examination shows that the cyst wall was lined with a single layer of flat epithelial cells.

Discussion

Biliary tract obstruction is a very rare complication associated with simple liver cysts, and it is generally caused by cysts that have diameters greater than 10 cm.[1] Indeed, a literature search revealed only 4 other cases of cysts with diameters less than 5 cm that had caused intrahepatic biliary dilatation.[2–4] However, one of these case reports indicated that the obstruction was probably caused by a coexisting cavernous hemangioma,[1] and in another case, the peripheral biliary duct dilatation occurred after cholecystectomy, possibly because of postoperative changes.[2] Thus, only 2 of the reports specifically discuss biliary dilatation caused by small simple cysts.

Ito *et al* reviewed 19 cases of simple cysts with biliary obstruction and reported that 18 of these cases had cysts located in the central portion of the liver.[1] One case was similar to the present case with the cyst located in the lateral segment. Lapeyre *et al* discuss whether a relationship exists between the anatomic structure of segment IV, defined according to Couinaud's classification system, and the compression of the biliary duct by small benign lesions.[3] The present case shows that cysts in the lateral segment can also cause peripheral dilatation of the bile duct.

In 2010, the World Health Organization categorized cystic neoplasms of the liver into 2 classes. This classification removed cystadenomas and cystadenocarcinomas and introduced IPNBs and mucinous cystic tumors. IPNB, which is characterized by cystic dilatation and communication with the bile duct,[7,8] is potentially malignant. In the present case, we could not rule out the possibility of the cyst being an IPNB because of the upstream bile duct dilatation and the possibility of communication between the cyst and bile duct demonstrated by CT and MRI. Although all reported cases of biliary dilatation with small simple cysts were treated surgically, cases of simple cysts without severe complications do not usually require surgery. As complications associated with small simple cysts are extremely rare, no guidelines or consensus statements have been developed for their treatment.[9,10] It is now recognized that even small simple cysts can obstruct the biliary tract; therefore, it is important to determine best practices for the management of cystic lesions in the liver. However, when diagnostic uncertainty exists, surgical procedures, including liver resection, should be considered the standard treatment for cystic liver lesions.

References

1. Ito K, Taira K, Arii S. Intrahepatic bile duct dilatation with a liver cyst and hemangioma: report of a case. *Surg Today* 2009; **39**(3):256–260
2. Inaba T, Nagashima I, Ogawa F, Tomioka M, Okinaga K. Diffuse intrahepatic bile duct dilation caused by a very small hepatic cyst. *J Hepatobiliary Pancreat Surg* 2003;**10**(1):106–108
3. Lapeyre M, Mathieu D, Tailboux L, Rahmouni A, Kobeiter H. Dilatation of the intrahepatic bile ducts associated with benign liver lesions: an unusual finding. *Eur Radiol* 2002;**12**(1):71–73
4. Lee K, Hong T. A small solitary non-parasitic hepatic cyst causing an intra-hepatic bile duct stricture: a case report. *J Med Case Rep* 2010;**4**(7):254

5. Del Poggio P, Buonocore M. Cystic tumors of the liver: a practical approach. *World J Gastroenterol* 2008;**14**(23):3616–3620
6. Kim JY, Kim SH, Eun HW, Lee MW, Lee JY, Han JK *et al*. Differentiation between biliary cystic neoplasms and simple cysts of the liver: accuracy of CT. *AJR Am J Roentgenol* 2010; **195**(5):1142–1148
7. Chen TC, Nakanuma Y, Zen Y, Chen MF, Jan YY, Yeh TS *et al*. Intraductal papillary neoplasia of the liver associated with hepatolithiasis. *Hepatology* 2001;**34**(4, pt 1):651–658
8. Zen Y, Fujii T, Itatsu K, Nakamura K, Konishi F, Masuda S *et al*. Biliary cystic tumors with bile duct communication: a cystic variant of intraductal papillary neoplasm of the bile duct. *Mod Pathol* 2006;**19**(9):1243–1254
9. Macutkiewicz C, Plastow R, Chrispijn M, Filobbos R, Ammori BA, Sherlock DJ *et al*. Complications arising in simple and polycystic liver cysts. *World J Hepatol* 2012;**4**(12):406–411
10. Nakanuma Y, Sato Y, Harada K, Sasaki M, Xu J, Ikeda H. Pathological classification of intrahepatic cholangiocarcinoma based on a new concept. *World J Hepatol* 2010;**2**(12):419–427