Case Report

Concomitant Hepatolithiasis with Primary Hepatocellular Carcinoma: An innovative problem

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ABSTRACT
Concomitant primary hepatocellular carcinoma with hepatolithiasis is a rare form. We present a case of left hepatolithiasis accompany with primary hepatocellular carcinoma of right lobe in a 62-year-old man, who underwent left lateral hepatectomy, cholecystectomy, common bile duct exploration and T-tube drainage 20 years ago for hepatolithiasis and gallstones. The tumor was detected during the follow-up for his residual hepatolithiasis. A right hemihepatectomy and common bile duct exploration and T-tube drainage was performed and left hepatolithiasis was remained for next operation to prevent hepatic failure. A histological examination of the specimen revealed typical features of hepatocellular carcinoma. This report describes staged management for patient with both benign and malignant hepato-biliary disease.

KEY WORDS: Hepatocellular carcinoma, Hepatolithiasis, Liver.

INTRODUCTION
Hepatolithiasis is an endemic disease commonly encountered in Southeast Asia and is especially prevalent in China¹², which is defined as the occurrence of stones in any intrahepatic bile duct proximal to the confluence of right and left hepatic ducts. The most serious complication of this disease is the development of intrahepatic cholangiocarcinoma (CC) and less frequently of hepatocellular carcinoma (HCC).³ Concomitant hepatolithiasis with primary HCC is a rare form of hepatic disease that contains both benign (hepatolithiasis) and malignant (HCC) lesions, and such sort of case has not been reported previously. We herein report a case of hepatolithiasis accompany with HCC in a Chinese man. The management of this rare disease is discussed.

CASE REPORT
A 62-year-old male patient was admitted to our department with right upper quadrant abdominal pain. His medical history was notable for nonanatomical resection in the left lateral lobe because of hepatolithiasis in a local hospital 20 years ago. However, aggressive treatment was not performed for the residual calculi as there were no clinical symptoms. And there was no tumor in the right lobe, he was just asked for follow-up every year. On his last follow-up, a physical examination revealed mild pain in the right upper quadrant of the abdomen. Colour Doppler Ultrasonography detected a huge hypoechoic lesion in right lobe and regional hepatolithiasis located in left lobe.

Further evaluation with magnetic resonance imaging (MRI) of the abdomen demonstrated a huge tumor of 14.7×11.6×15.3cm in right lobe, multiple...
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intrahepatic calculi located within the left biliary tree, and atrophy of the left lobe with compensatory hyperplasia of caudate lobe (Fig.1). The results of laboratory tests showed that the serum levels of γ-glutamyltransferase (124 U/l; normal 0–38 U/l), aspartate aminotransferase (48 U/l; normal 0–31 U/l), alkaline phosphatase (271 U/l; normal 30–120 U/l), and alanine aminotransferase (82 U/l; normal 0–35 U/l) were elevated. Tumor markers including carcinoembryonic antigen and carbohydrate antigen 12-5 were within normal limits, except α-fetoprotein (>1200ng/ml; normal 0-8ng/ml), carbohydrate antigen 19-9 (64.41 U/ml; normal 0-22U/ml). Serologic tests for hepatitis A, B, and C were all negative. The indocyanine green clearance rate at 15 min was 14.2% (normal range<15%).

The patient underwent right hepatectomy to remove the tumor. During the operation, the left lobe was found to be atrophic and fibrotic. The surgeon did not resect left lobe for the reason of prevention of hepatic failure. After tumor resection, common bile duct exploration, cholangioscopic lithotomy and T-tube drainage was performed. The surgical specimen with pseudocapsule was measured 14×11×15cm (Fig.2). A histological examination of the specimen revealed typical features of hepatocellular carcinoma. The patient was discharged on the 14th day with T-tube occlusion on the 12th day postoperatively; cholangiography was performed one month later with no stone detected in common bile duct, and T-tube was completely withdrawn due to

Fig.1: Abdominal MRI shows a huge tumor of 14.7×11.6×15.3cm in right lobe, multiple intrahepatic calculi located within the left biliary tree, and atrophy of the left lobe with compensatory hyperplasia of caudate lobe.

Fig.2: The surgical specimen with pseudocapsule was measured 14×11×15cm

Fig.3: The abdominal CT shows the remnant liver in the fourth month after operation, and intrahepatic calculi still located within the left biliary tree.
fail to extract hepatolithiasis, which still located in left lobe. He was asked to have a second surgical procedure of left hepatectomy for removing regional hepatolithiasis according to the regeneration of liver. Follow-up has been continued for five months without any particular findings (Fig.3).

**DISCUSSION**

Hepatolithiasis, as a common disease of the biliary system, is a significant cause for deaths due to benign liver disease in China. The treatment is mainly dependent on surgical operation. Especially for regional hepatolithiasis, anatomical liver resection should be performed. When accompanied with HCC, although a rare case, it is very difficult to deal with both the malignant and benign lesions at the same time, because preservation of enough liver parenchyma is critical for the success of the operation. Considering the huge HCC in right lobe and the hepatic functional reserve, we used the method of staged resection, first to resect HCC and second to hepatolithiasis. However, this case addresses few questions which still need to be resolved.

**Firstly, why not deal with both the lesions at the same time?** The regeneration of a normal liver is strong, and the acceptable residual functioning volume should range from 30 to 60% in patients with hepatitis, and from 40 to 70% in cases of cirrhosis. In spite of improvements in increasing surgical confidence and expertise, the parameters have remained largely unchanged. This patient suffered left lateral hepatectomy 20 years ago, and the indocyanine green clearance rate at 15 min was 14.2%, together with atrophy and fibrosis of left lobe. Resection of the lesions at the same time was impossible. The consequence of excessive liver resection is the development of progressive hepatic failure. No evidence is currently available according to our best knowledge to address this issue. Although there are options for such case like liver transplantation, considering lack of donors, high expenses for transplantation, postoperative immune rejections and tumor recurrence, there seemed to have no best method. As a result, staged resection may be adapted.

**Secondly, face both the malignant and benign lesions, which should be resolved firstly?** To prevent immediate and late sequelae of hepatolithiasis, such as suppurative cholangitis, septicemia, secondary common bile duct stones and cholangiocarcinoma, aggressive treatment is needed. On the other hand, as a deadly disease, HCC also needs treatment immediately. A big dilemma arises for surgeons, with which should we deal first? Theoretically, we should deal with the more dangerous and deadly lesion. Of this patient, hepatolithiasis was in its silent period and with no urgent complications, which should be kept for next surgical procedure.

**Thirdly, when should we deal with the hepatolithiasis and what is the best time?** This issue is also debatable. Clinical studies suggest that regeneration is evident within two weeks following resection and is complete at three months. However, for the diseased liver, regeneration should experience a longer time. So the best time for next operation is unclear, which may be dependent on the routine test of follow-up, the complication of hepatolithiasis, as well as the possible recurrence of HCC.

**CONCLUSION**

In conclusion, since science and technology are in era of its advancement, the management of this type of disease with both malignant and benign lesions is complex and difficult. This is still a big challenge for surgeons for the lack of cases and principles of evidence based medicine.

**REFERENCES**