



Hepatocellular carcinoma in the native liver of a 38-year-old female patient with biliary atresia



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ABSTRACT

We report a rare case of hepatocellular carcinoma in native liver in a case of biliary atresia. The patient was a 38-year-old female with three children who had an aggressive tumor, resulting in her subsequent death. We also review 14 reports, published previously in the English language medical literature, concerning hepatocellular carcinoma originating from native liver in biliary atresia cases and discuss the possible etiology, and propose more careful follow up for the patients with biliary atresia who suffer from repetitive cholangitis and/or experience the child delivery.

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Hepatocellular carcinoma (HCC) is rarely seen in cirrhotic liver of biliary atresia, with only 20 cases having been reported in the English language medical literature [1–14]. Almost all hepatocellular carcinomas seen in biliary atresia are slow growing and not very aggressive tumors, with half of them incidentally found in extracted livers at autopsy or after liver transplantation.

Here we report on a 38-year-old female patient with biliary atresia, who had long suffered from repetitive cholangitis and finally died of a hepatocellular carcinoma in her native liver.

1. Case

A 38-year-old female patient was referred to our department because a hepatic tumor was suspected after a high serum alpha-fetoprotein (AFP) level, and as subsequently demonstrated by abdominal computed tomography (CT) scan. She had previously been admitted to our institute because of prolonged jaundice during her early infantile years. As a 71-days-old baby, after

several examinations, the patient was diagnosed with biliary atresia type III and underwent portoenterostomy with a bile drainage stoma. After the operation, her jaundice subsided and since then, she has been followed at our outpatient clinic. Periodic blood tests showed mild liver dysfunction and she underwent splenectomy at 24 years of age because of worsening hypersplenism caused by prolonged portal hypertension. The patient had borne three children, delivering them at 30, 35, and 38 years of age. She was followed at our outpatient clinic every three months and at 38 years of age abdominal ultrasound were taken and a hepatic mass, 3 cm in diameter, was found in the posterior segment of the right lobe (Fig. 1a) which was first thought to be focal nodular hyperplasia. But a blood test showed a high AFP level (2632.8 ng/mL). The mass grew rapidly to 6 cm by two weeks which was ascertained by abdominal CT scans, and hepatocellular carcinoma was strongly suspected (Fig. 1b and c). Serological tests were negative for hepatitis B antigen and hepatitis C antibody. A month later, a laparotomy revealed a bloody ascites in the abdominal cavity, which suggested a spontaneous rupture of the tumor. The posterior segment of the right lobe (Fig. 2a) was resected and the tumor was macroscopically removed. The pathological diagnosis was a moderately differentiated hepatocellular carcinoma (Fig. 2b and c); background liver tissue showed severe

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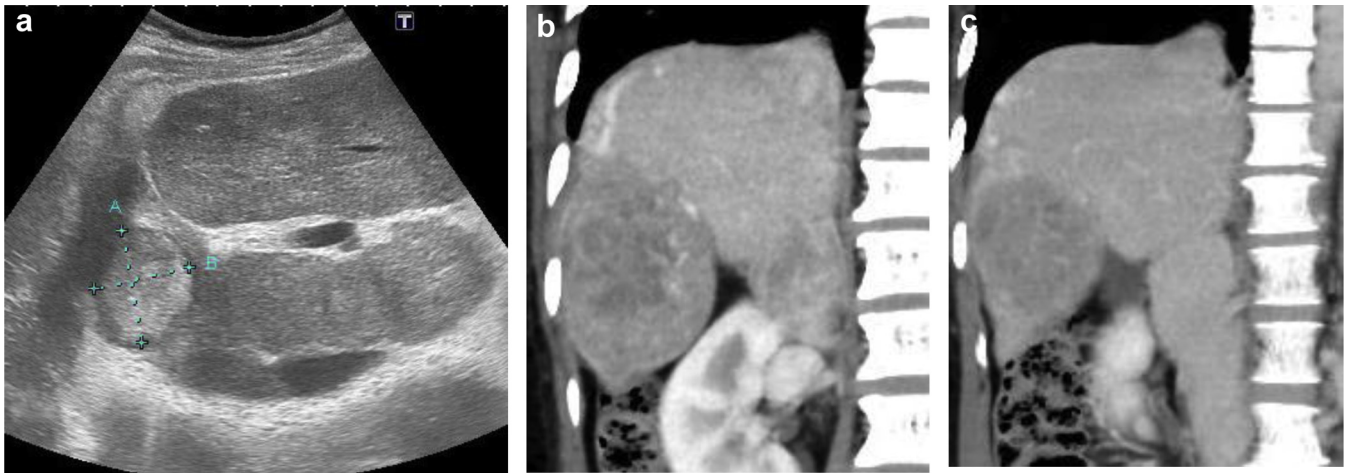


Fig. 1. (a) An abdominal ultrasound examination demonstrated a mass of 3.0 cm diameter in the right lobe of the liver. (b) Two weeks later, an abdominal CT scan showed the mass in the liver had grown rapidly to a diameter of 6.0 cm. Arterial phase imaging revealed the mass to have an enhanced margin. (c) An abdominal CT of the venous phase showed a relatively contrasting mass with low density. These results strongly suggested the mass was a hepatocellular carcinoma.

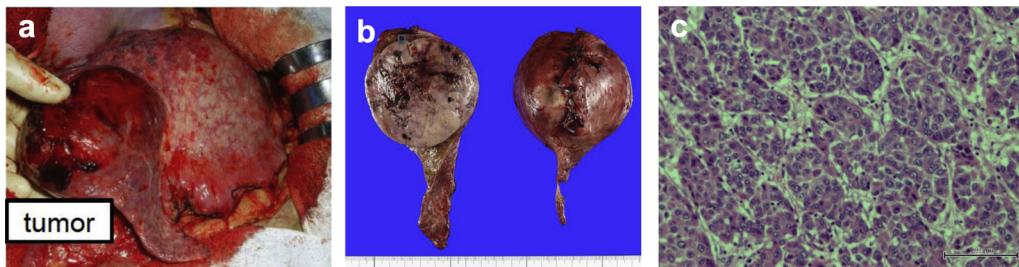


Fig. 2. (a) Intraoperative photo of the tumor. The tumor had ruptured and was sited in the posterior segment of the right liver. Segment 1 (caudate lobe) was very hypertrophic. (b) Macroscopic appearance of the tumor, which was well encapsulated, but partially necrotic and ruptured. (c) Microscopic findings revealed that the tumor was a moderately differentiated, hepatocellular carcinoma.

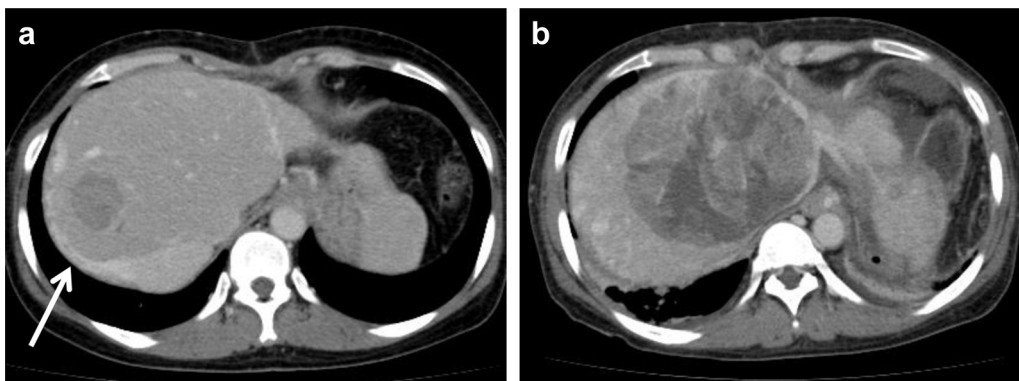


Fig. 3. (a) Tumor recurrence was detected in the caudate lobe of the liver, which was found to be extremely hypertrophic, three months after the first operation. The tumor was resected in a second operation. The arrow indicates the middle hepatic vein. (b) The tumor recurred after a second operation. Its growth could not be controlled by chemotherapy combined with Sorafenib.

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