World Journal of Gastroenterology

Online Submissions: http://www.wjgnet.com/1007-9327office
wjg@wjgnet.com
doi:10.3748/wjg.v16.i41.5263

World J Gastroenterol 2010 November 7; 16(41): 5263-5266
ISSN 1007-9327 (print) ISSN 2219-2840 (online)

CASE REPORT

Primary malignant liver mesenchymal tumor: A case report

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Received: August 14, 2010 Revised: September 13, 2010
Accepted: September 20, 2010
Published online: November 7, 2010

Abstract

Primary malignant liver mesenchymal tumor is a rare condition defined as a tumor with vascular, fibrous, adipose, and other mesenchymal tissue differentiation. We report a case of primary malignant liver mesenchymal tumor in a 51-year-old male with anemia, weight loss and hepatomegaly. Finally unconventional liver biopsy and histological manifestation led to the definitive diagnosis.

Key words: Hepatic mesenchymal tumor; Liver biopsy; Hepatic leiomyosarcoma; Hepatic schwannoma

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Chen J, Du YJ, Song JT, E LN, Liu BR. Primary malignant liver mesenchymal tumor: A case report. *World J Gastroenterol* 2010; 16(41): 5263-5266 Available from: URL: http://www.wjgnet.com/1007-9327/full/v16/i41/5263.htm DOI: http://dx.doi.org/10.3748/wjg.v16.i41.5263

INTRODUCTION

Primary malignant liver mesenchymal tumor is a very rare tumor, accounting for less than 1% of all hepatic malignancies[1]. Hepatic angiosarcoma, leiomyosarcoma, embryonal sarcoma, schwannoma and lymphoma are the more common mesenchymal tumors. The diagnosis is dependent on histological imaging. However, some cases of multiple nodular lesions can only be diagnosed by percutaneous liver biopsy (PLB) without surgery. We present a case of malignant liver mesenchymal tumor in an adult and its diagnosis and treatment were discussed.

CASE REPORT

A 51-year-old Chinese male was referred to our hospital with a 2-mo history of mild abdominal pain, an abdominal mass in the upper quadrant, fatigue and progressive weight loss. His symptoms gradually worsened with fatigue 1 wk after onset of the disease. He had no fever, cough or skin lesions in the past 2 mo and no family history of liver and autoimmune diseases. Physical examination revealed mild pale conjunctiva and skin, and enlargement of the liver without percussion pain.

His complete blood hemoglobin was 58 g/L. Stool occult blood test was negative. Liver functional test showed that his alanine aminotransferase, aspartate aminotransferase, alkaline phosphatase, γ glutamyl tranzpehtidaze, total bilirubin, direct bilirubin, and albumin were 81 U/L, 86 U/L, 224 U/L, 73 U/L, 23.55 μmol/L, 11.74 μmol/L, 30.5 g/L, respectively. Series hepatitis markers (hepatitis A virus, hepatitis B virus, hepatitis C virus, hepatitis D virus, hepatitis E virus, Epstein-Barr virus, and cucumber mosaic virus) were negative. α-fetoprotein, carcinoembryonic antigen and CA19-9 concentrations were also normal. Human immunodeficiency virus and syphilis antibodies were negative. Coagulation profile was normal.

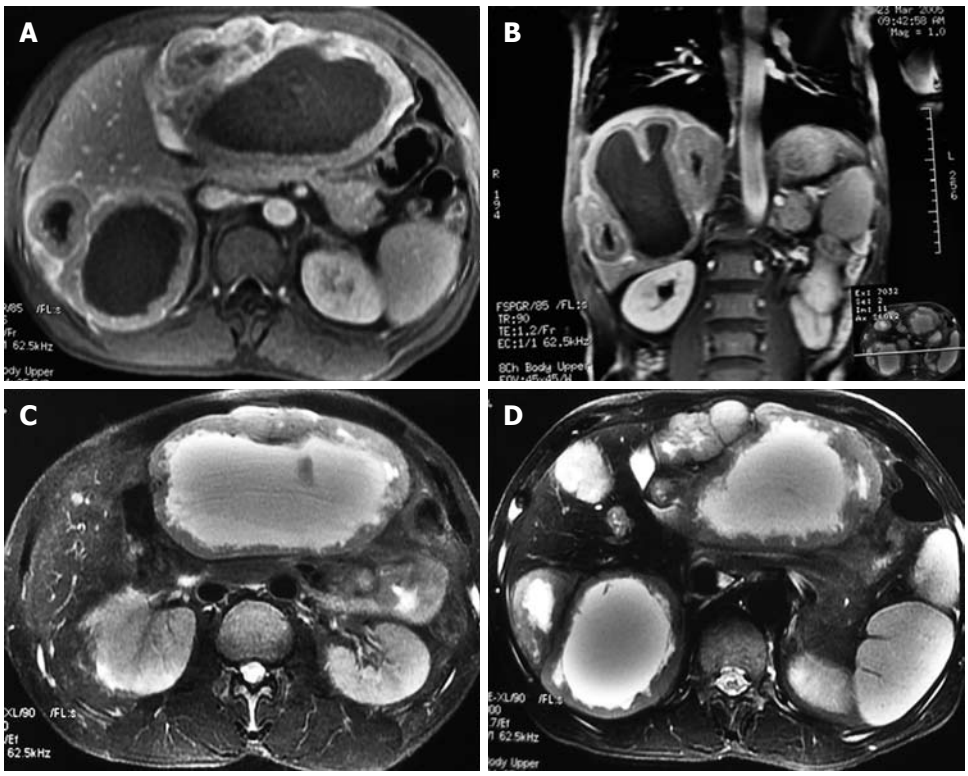

Figure 1 Axial fat-suppressed T2-weighted turbo spin echo magnetic resonance imaging showing two large lesions (A) and several small cyst-like lesions (B) in liver, and coronal and transverse T1-weighted imaging showing thickened wall of lesions with a ragged appearance (C) and enhancement (D) in liver.

Ultrasound and magnetic resonance imaging (MRI) were used as the initial diagnostic tools. Abdominal ultrasound revealed a solid mass in the liver with a low echo, a sharp border, a rich blood supply and liquefaction necrosis. The maximum diameter of the mass was 13.9 cm. Abdominal MRI showed multiple diffuse abnormal signals suggestive of a possible malignant tumor. Axial fat-suppressed T2-weighted turbo spin echo imaging showed two larger and several smaller well-margined cyst-like lesions in the liver (Figure 1A and B). Coronal and transverse T1-weighted imaging demonstrated thickened wall of lesions with a ragged appearance and enhancement (Figure 1C and D). Endoscopy showed duodenal ulcer (S2 stage), normal colon and rectum.

PLB was performed under ultrasonography (US) guidance using a Bard biopsy gun (18-gauge cut needle) to make a final diagnosis of the lesions in liver. During biopsy, bloody liquid was observed in the lesion. Microscopic examination confirmed that the bloody liquid contained a large number of erythrocytes. However, pathology of the liver only showed a spindle cell tumor. The possible reasons are as follows. First, the liver specimen was too small because the lesion contained only fluid and was surrounded by a very thin wall. Second, the location of tumor and its hardness limited the adjustable puncture angle.

To confirm the diagnosis of this patient, another unconventional liver biopsy was performed with gastroscopic biopsy forceps as follows.

At beginning of the procedure, a local anesthetic agent (5% lidocaine) was injected subcutaneously through a 25-gauge needle. Next, a 15 cm long 18 gauge puncture needle was passed into the cyst under US guidance. After the local skin and subcutaneous tissue were dilated, a PTC exchange stainless steel guide wire with a flexible tip was introduced into the cyst cavity through the needle, and then the needle was withdrawn. A 5-French catheter was plated into the cyst using the Seldinger technique. Finally, gastroscopic biopsy forceps was passed into the cyst through the catheter to get the liver tissue sample. The histological results indicated that spindle-shaped tumor cells were well-oriented, arranged in bundles and clustered in some part of the regional tumor. Nuclei with rare mitosis were elongated in a rod-like shape with different sizes and blunt ends. The final diagnosis was established as a low grade malignant liver mesenchymal tumor (Figure 2).

The patient was suggested to receive hepatic arterial chemoembolization. Selective digital subtraction angiography in the early and later phases showed lesions surrounded by abnormal tortuous tumor arteries (Figure 3A) and patchy enhancement (Figure 3B), respectively, in different areas. Then, 5-Fu (750 mg), mitomycin (5 mg), pyrazine imidacloprid Star (30 mg), and super-liquid iodized oil (5 mL) were infused into the tumor arteries. After treatment, liver function of the patient was improved and his hemoglobin level increased (Table 1). The patient died 2 years after treatment.

DISCUSSION

In this paper, we presented a case of multiple nodular cys-

Table 1 Changes in serum biochemical index before and after treatment

	WBC ($\times 10^9$/L)	HGB (g/L)	PLT ($\times 10^{12}$/L)	ALT (U/L)	AST (U/L)	TBIL (μmol/L)	ALB (g/L)
Diagnosis	10.44	58	294	81	86	23.55	30.5
Treatment	6.24	86	241	17	24	20.74	34.1

WBC: White blood cell count; HGB: Hemoglobin; PLT: Platelet count; ALT: Alanine aminotransferase; AST: Aspartate aminotransferase; TBIL: Total bilirubin; ALB: Albumin.

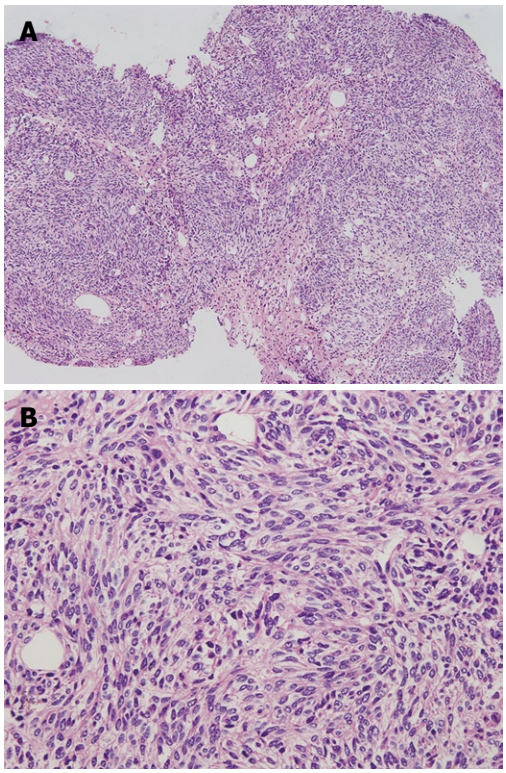

Figure 2 Hematoxylin and eosin staining showing a low grade malignant liver mesenchymal tumor under the magnification × 10 (A) and × 40 (B).

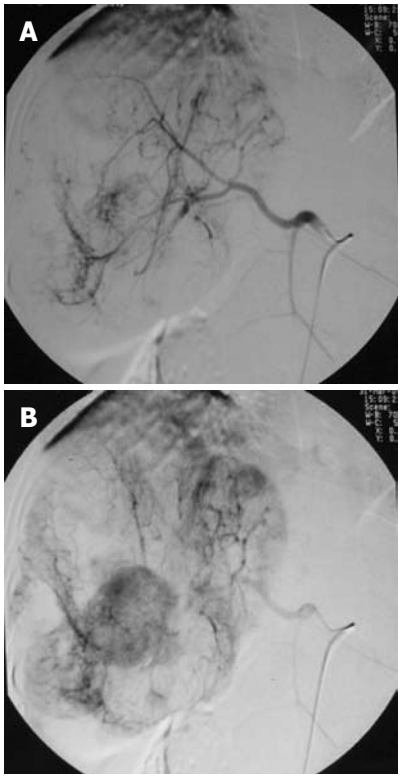

Figure 3 Selective digital subtraction angiography showing abnormal tortuous arteries in the early phase (A) and patchy enhancement in later phase (B) in different areas.

tic lesions in liver. The final diagnosis was established as a low grade malignant liver mesenchymal tumor. However, HE staining could not show the source of mesenchymal cells. Further immunohistochemistry staining with Vimentin, Desmin and α-SMA was performed, which could not sill show the source of mesenchymal cells.

Primary malignant liver leiomyosarcoma and schwannoma are both rare tumors in the liver. Primary liver leiomyosarcoma is a rare malignancy involving the liver, occurring as a primary liver sarcoma in patients without any underlying disorder, and its incidence increases as a primary tumor in immunodeficiency patients[2]. Its usual clinical presentation is painful hepatomegaly or epigastric mass[3]. Malignant liver schwannoma is the most common soft tissue sarcoma in adults, but primary liver schwannoma is extremely rare. Only 12, 8, and 1 cases of benign, malignant, and semimalignant liver schwannoma are available from the literature worldwide[4-12]. Hepatic schwannoma is usually associated with neurofibromatosis. However, two cases of malignant liver schwannoma without neurofibromatosis have been reported[9,11].

Unfortunately, it is difficult to make the diagnosis of malignant liver mesenchymal tumor because both its clinical presentation and imagine are nonspecific.

The common symptoms and signs of patients with malignant liver mesenchymal tumor include abdominal pain, weight loss, weakness, loss of appetite, vomiting, enlargement of the liver, ascites, and jaundice, which lack of specificity in differential diagnosis between benign and malignant mesenchymal tumors.

Furthermore, imaging studies, such as MRI scan, computed tomography (CT) scan and angio photography are the commonly used methods to identify the characteristics of liver tumor. However, imaging findings of liver mesenchymal tumors, including leiomyosarcoma and schwannoma, are nonspecific and infrequently reported[13-19]. It is quite difficult for MRI and CT scan to differentiate primary liver mesenchymal neoplasms from other liver malignancies, although they should be included in differential diagnosis when MRI or CT scan demonstrates hepatic lesions without characteristics of hepatocellular carcinoma,

especially in patients with no extrahepatic primary malignancies.

The diagnosis of malignant liver mesenchymal tumor depends on histological change in either needle or open biopsy, while metastatic status is facilitated by the presence of extrahepatic primary tumor.

Liver biopsy is an important diagnostic tool and helps make therapeutic decision for liver tumor. Open biopsy is a major surgical procedure for liver tumor. PLB under ultrasound or CT guidance is a safe and almost painless procedure for lesions with a soft tissue component or located close to vital structures[20]. Most reported cases of cystic liver mesenchymal tumor were diagnosed by open liver biopsy. In this case, uncommon PLB method was used instead of the routine PLB to make the final diagnosis, avoiding injury and complications of open biopsy. Minimally invasive treatment devices, such as gastroscopic biopsy forceps and catheters used in liver cancer intervention, make the new PLB method possible, which is safe and reliable and can thus be used in diagnosis of cystic liver lesions.

In conclusion, mesenchymal liver tumor is rare in adults and cross-sectional findings are varied, which can be diagnosed with the uncommon PLB method.

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S- Editor Sun H **L- Editor** Wang XL **E- Editor** Zheng XM