

A Liver Inflammatory Pseudotumor Misdiagnosed as Liver Cancer Using Noninvasive Diagnostic Methods

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Abstract

Background: Inflammatory pseudotumor (IPT) is a rare liver tumor, and is not easy to distinguish from liver cancer in imaging examinations. We retrospectively analyzed the clinical data of a case of IPT misdiagnosed as liver cancer.

Case Report: A 39-year-old man was hospitalized in February 2014 because of right upper quadrant pain for 7 months. Enhanced multi-detector computed tomography and Gd-EOB-DTPA-enhanced magnetic resonance imaging findings were consistent with malignant liver tumors. The patient had a history of chronic hepatitis B. Therefore, he was diagnosed with liver cancer. However, his physical condition was not suitable for surgery, and he had a history of 2 abdominal surgeries and tested negative for alpha-fetoprotein (AFP). Thus, we performed a liver biopsy and the histological diagnosis was IPT. The patient avoided interventional therapy, which is the preferred treatment for patients with inoperable primary liver cancer.

Conclusion: IPT should be considered in the differential diagnoses when a mass lesion in the liver is encountered, especially for those patients with a history of infection in the abdomen or abdominal surgery who are AFP-negative. Pathologic examination may be necessary.

Keywords: Liver inflammatory pseudotumor; Liver cancer; Gd-EOB-DTPA

Abbreviations: ALT: Alanine Aminotransferase; AST: Aspartate Aminotransferase; A/G: Albumin to Globulin ratio; BUN: Blood Urea Nitrogen; LDH: Lactate Dehydrogenase; MDCT: Multi-Detector Computed Tomography; MRI: Magnetic Resonance Imaging; IPT: Inflammatory Pseudotumor; WI: Weighted Image

Introduction

Inflammatory pseudotumor (IPT) is a rare benign tumor, and most often occurs in the lung [1,2], followed in frequency by the liver [3]. Liver IPT is a tumor-like lesion, formed by the infiltration of inflammatory cells under the influence of various pro-inflammatory cytokines in liver tissue and hyperplasia of fibrous tissue. IPT is often difficult to differentiate from malignant liver tumors because of the lack of specific IPT symptoms and imaging findings [1,2]. Liver IPT has become detectable because of ongoing advances in imaging technology and new progress in the diagnosis of liver tumors [3]. A feature of liver cancer is that it allows for diagnosis with non-invasive examination without a liver biopsy [4]. The sensitivity and specificity of non-invasive diagnosis for liver cancer with nodules >2 cm and cirrhosis are more than 95% [5,6]. We report a case that was misdiagnosed as liver cancer using ultrasound, multi-detector computed tomography (MDCT), and Gd-EOB-DTPA-enhanced magnetic resonance imaging (MRI), and confirmed as liver IPT by biopsy.

Case Report

A 39-year-old man was hospitalized in February 2014 because of right upper quadrant pain for 7 months. During the nearly 7 months, he had paroxysmal pain with no obvious cause, and had no discomfort related to eating, breathing, or pulse rate. Moreover, he had remission after he received anti-inflammatory, antispasmodic, and other symptomatic treatments. The liver appeared uniform in B-mode ultrasonography, but a lesion in the right lobe prompted CT examination of the upper abdomen at People's Hospital of Fu Chuan County. He was diagnosed

with malignant liver tumors. For further diagnosis and treatment, he was transferred to the Affiliated Hospital of Guilin Medical University. He had a prior surgical history, with a "pancreatic subtotal resection" because of acute pancreatitis in 1999, and an "anastomosis of pancreatic pseudocyst to jejunum", and transfused 200 ml of blood in 2000. He had type 2 diabetes, but no hypertension, coronary heart disease, or tuberculosis. He had no family history of liver disease and habit of drinking and smoking.

On admission, his temperature was 36.3°C and body weight was 53 kg, with no obvious weight loss during the previous 6 months. His skin and sclerae were not yellow, and liver palms and spider nevi were not seen. Systemic superficial lymph nodes were not enlarged, the abdomen was soft and flat, the right upper quadrant and subxiphoid area showed mild tenderness, without rebound. The liver and spleen were not palpable or tender. No abnormality was present on heart and lung auscultation. Patient's mental outlook was well.

Ancillary examination results

The results of blood tests on admission were as follows: hemoglobin, 88 g/L (normal, 120 g/L to 160 g/L); white blood cell count, 15.280 × 10⁹/L (normal, 4 × 10⁹/L to 10 × 10⁹/L); platelet count, 318 × 10⁹/L (normal, 100 × 10⁹/L to 300 × 10⁹ L; neutrophils, 0.806; mononuclear

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cells, 1.240). The absolute value of total bilirubin was 5.99 $\mu\text{mol/L}$, albumin was 22.52 g/L, globulin was 52.46 g/L, albumin to globulin ratio (A/G) was 0.43, alkaline phosphatase was 258.30 U/L, cholinesterase was 3,157 U/L, γ -glutamyltranspeptidase was 184 U/L, blood urea nitrogen (BUN) was 8.06 mmol/L, alanine aminotransferase (ALT) was 8.59 U/L, aspartate aminotransferase (AST) was 14.2 U/L, lactate dehydrogenase (LDH) was 119.56 U/L, fibrinogen was 6.47 g/L, HBsAg was 18.13 ng/ml, HBeAg was 0.33 ng/ml, HBcAb was 3.56 ng/ml, CEA was 0.617 ng/ml, CA 19-9 was 14.090 U/mL, CA 125 was 49.140 U/ml, AFP was 1.970 ng/ml, CRP was 46.89 mg/L. The chest X-ray and electrocardiogram were normal.

On admission, the B-ultrasound showed hepatic parenchymal lesions and intrahepatic bile duct widening. MDCT showed a non-enlarged liver (Figure 1). An area of patchy, slightly lower density was seen in the left hepatic lobe, which had an unclear boundary. Focal liver lesions were present, with inhomogeneous enhancement in the arterial phase of an enhanced scan, and disappearance in the venous phase. The focus measured about 10.5 cm \times 6.5 cm, with remnant liver parenchyma, abnormal density, and abnormal enhancement foci, as well as slight dilation of the intrahepatic bile duct, and local scatter of a translucent gas shadow.

In (Figure 2), Gd-EOB-DTPA-enhanced MRI suggested that the liver volume was slightly increased. A cluster of irregular and abnormal signals was seen in the left lobe and right anterior lobe. The boundary of the lesions was not clear, and the area measured about 11 cm \times 7.2 cm. T1 weighted image (WI) showed a slightly low signal, and T2WI with T2 fat suppression showed a slightly high signal; diffusion-weighted imaging (DWI) showed a heterogeneous high signal, and the corresponding local diffusion coefficient (ADC) map showed a low signal. After intravenous injection of liver-specific contrast agent for enhancement, the lesions showed inhomogeneous medium reinforcement in arterial and venous phases. Lesion enhancement then gradually diminished in the delayed phase at 5 min, 15 min, 20 min, and 35 min, and the signal was lower than in normal liver tissue. Another lesion measuring 6 mm in diameter with long T1 and long T2 signal was present in the right liver lobe; the lesion showed ring-enhancement in the arterial phase, and the reinforcement signal filled the image from the periphery to the center in the delayed phase. There was good hepatic vein and portal vein filling. The left and right intrahepatic bile ducts were dilated and the extrahepatic bile duct was present. Resection of most of the pancreas could be seen, along with the residual pancreatic head. Abnormal clumps of signals were present around the pancreatic head, with no clear boundary between the lesions and the pancreas. DWI showed a high signal, ADC showed a low signal, and the enhancement scan showed uneven enhancement. The gallbladder image was not clear. The bilateral renal parenchyma and the spleen morphology, size, and signal were normal. The distal stomach wall was thickened and showed marked enhancement, and the boundary appeared distinct from the surrounding tissue. The greater omentum near the stomach had irregular nodular thickening, and there was reinforcement of signal on enhanced scanning. A small amount of effusion was present in the abdominal cavity. Multiple enlarged lymph nodes were observed near the abdominal aorta.

Based on the imaging examinations and a history of hepatitis B, the liver lesion was considered to be malignant. Because of insufficient normal liver volume and abdominal lymph node metastasis, he had no surgical indication and was recommended for liver biopsy in order to clarify the nature of the tumor. He underwent liver biopsy on hospital day 6. The histological evaluation (Figure 3) revealed multiple abscesses formation large clusters of bacteria in the abscesses and inflammatory cell infiltration and neutrophilic infiltrates in the

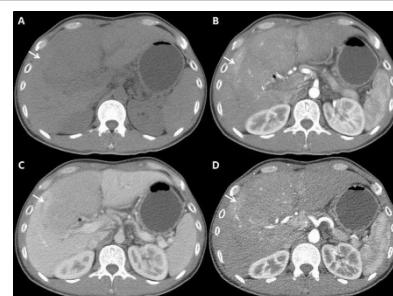


Figure 1: A: Scan phase. Areas with patchy, slightly lower density; B: Arterial phase. Lesions with inhomogeneous enhancement; C: Venous phase. Loss of lesion enhancement; D: Delayed phase. Loss of lesion enhancement.

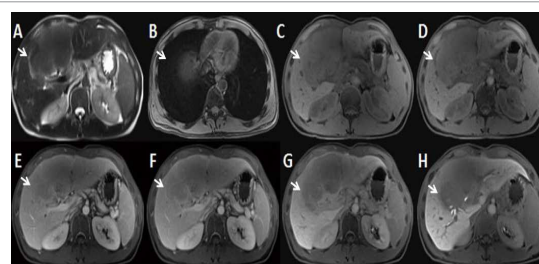


Figure 2: MRI A: T2. Slightly higher signal; B: T1. Slightly low signal; C: Enhanced scanning; D: Arterial phase. Venous phase enhancement lesions with inhomogeneous medium; E: Venous phase; F: Delayed phase at 5 min; G: Delayed phase at 10 min; H: Delayed phase at 30 min.

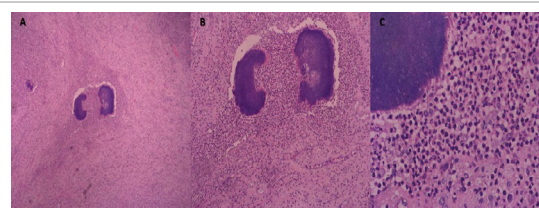


Figure 3: Pathological examination (hematoxylin-eosin staining) A: 4X, multiple abscess formation; B: 10X, large number of clustered bacteria in the abscess and inflammatory cell infiltration; C: 40X, many neutrophilic infiltrates in abscess and surrounding tissues.

abscess and surrounding tissues. There were no malignant tumor cells. Therefore, the final diagnosis was liver IPT. During the hospital stay, the patient received anti-infective, and liver-protective therapy, as well as nutritional support. The patient was discharged after his general condition improved.

In November 2014, he was re-hospitalized. The level of albumin was 20.89 g/L, globulin was 48.76 g/L, A/G was 0.43, and pre-albumin was 38.35 mg/L. The other test results were normal. CT showed no difference compared with previous results. Liver biopsy findings indicated IPT again. The patient received liver-protective therapy and nutritional support, and resumed monthly outpatient monitoring after discharge. On long-term follow-up, the liver lesion gradually remitted. The patient's general condition improved.

Discussion

IPT is a rare benign tumor, and is divided into 3 types: yellow granulomatous pseudotumor, plasma cell granuloma-type pseudotumor, and sclerosing pseudotumor [7]. IPT can be isolated, or appear as several lesions at the same time, with a reported size up to 25 cm. Under a microscope, IPT shows characteristic, shuttle-shaped cells, muscle fiber

cells, and mixed inflammatory cells (plasma cells, lymphocytes, and sporadic tissue cells). The most common IPT symptoms are abdominal pain, fever, and weight loss. IPT is often self-limited, and has a good prognosis [8].

IPT occurs most commonly in the lungs, followed by the liver. Liver IPT was reported for the first time in 1953, with more than 200 reports thereafter [9]. The etiology of liver IPT is unknown, but is associated with bacterial infection [10], abscess formation [11], autoimmune reactions, and virus infection. Because there are no typical laboratory or imaging features, it is difficult to diagnose. The most common symptoms are abdominal pain, fever, fatigue, and weight loss [12,13], and occasionally jaundice and gastrointestinal discomfort. A small number of patients (about 9%) may be asymptomatic [12]. The IPT on Gd-EOB-DTPA-enhanced MRI appeared hypointense on T1 and T2 weighted images, and completely hypointense on Gd-EOB-DTPA-enhanced images [14].

Liver cancer shows a signature finding of arterial-phase enhancement followed by portal or delayed “washout” on contrast-enhanced MDCT and/or contrast-enhanced MRI. Multiple row CT and MRI can identify and determine the nature of liver tumors larger than 2 cm in diameter [4]. Gd-EOB-DTPA-enhanced MRI is useful in the diagnosis of liver cancer. Wang [15] found that Gd-EOB-DTPA-enhanced MRI can increase the accuracy of the diagnosis of liver cancer. For liver cancer, Gd-EOB-DTPA-enhanced MRI and high-row enhanced CT are accurate and effective imaging examinations in clinical diagnosis. Research has shown a high rate of diagnosis of liver cancer by using Gd-EOB-DTPA-enhanced MRI [16,17]. Nonetheless, MDCT and Gd-EOB-DTPA-enhanced MRI could not accurately distinguish between malignant liver tumors and IPT [18,19].

This patient underwent 3 additional image examinations including Gd-EOB-DTPA-enhanced MRI, MDCT, and ultrasound, which indicated liver malignancy. As the patient had a history of chronic hepatitis B, we diagnosed liver cancer with a non-invasive examination. Because of insufficient normal liver volume and abdominal lymph node metastasis, he was not a candidate for surgery.

In contrast with patients with a history of severe acute pancreatitis and abdominal surgery, the present patient's mental outlook was unlike those with malignant tumors; as he tested negative for AFP, we performed a liver biopsy. Histology showed a large number of inflammatory cells, but no malignant cells. Biopsy pathology showed IPT both times. Long-term follow-up confirmed IPT. The patient avoided transhepatic arterial chemotherapy and embolization, which is the preferred treatment for patients with inoperable primary liver cancer.

Conclusion

IPT should be considered in the differential diagnoses when a mass lesion in the liver is encountered, especially in those patients with a history of infection in the abdomen or abdominal surgery, and who are AFP-negative. Pathologic examination may be necessary.

References

- Faraj W, Ajouz H, Mukherji D, Kealy G, Shamseddine A, et al. (2011)

Inflammatory pseudo-tumor of the liver: A rare pathological entity. World J Surg Oncol 9: 5.

- Torzilli G, Inoue K, Midorikawa Y, Hui AM, Takayama T, et al. (2001) Inflammatory pseudotumors of the liver: Prevalence and clinical impact in surgical patients. Hepatogastroenterology 48:1118-1123.
- Tsou YK, Lin CJ, Liu NJ, Lin CC, Lin CH, et al. (2007) Inflammatory pseudotumor of the liver: Report of eight cases, including three unusual cases, and a literature review. J Gastroen Hepatol 22: 2143-2147.
- Yoon JH, Park JW, Lee JM (2016) Noninvasive diagnosis of hepatocellular carcinoma: Elaboration on Korean liver cancer study group-national cancer center Korea practice guidelines compared with other guidelines and remaining issues. Korean journal of radiology (KJR) 17: 7-24.
- Torzilli G, Minagawa M, Takayama T, Inoue K, Hui AM, et al. (1999) Accurate preoperative evaluation of liver mass lesions without fine-needle biopsy. Hepatology 30: 889-893.
- Levy I, Greig PD, Gallinger S, Langer B, Sherman M (2001) Resection of hepatocellular carcinoma without preoperative tumor biopsy. Ann Surg 234: 206-209.
- Ntinis A, Kardassis D, Miliaras D, Tsinoglou K, Dimitriades A, et al. (2011) Inflammatory pseudotumor of the liver: a case report and review of the literature. J Med Case Rep 5: 196.
- Yamaguchi J, Sakamoto Y, Sano T, Shimada K, Kosuge T (2007) Spontaneous regression of inflammatory pseudotumor of the liver: Report of three cases. Surg today 37: 525-529.
- Akatsu T, Wakabayashi G, Tanimoto A, Kameyama K, Kitajima M (2006) Inflammatory pseudotumor of the liver masquerading as hepatocellular carcinoma after a hepatitis B virus infection: Report of a case. Surg today 36:1028-1031.
- White JE, Chase CW, Kelley JE, Brock WB, Clark MO (1997) Inflammatory pseudotumor of the liver associated with extrahepatic infection. Southern Med J 90: 23-29.
- Jover Diaz F, Martin Gonzalez C, Mayol Belda MJ, Cuadrado Pastor JM (2009) Multiple inflammatory liver pseudotumors and *Enterococcus durans* bacteremia. Gastroenterol Hepatol 32: 97-100.
- Goldsmith PJ, Loganathan A, Jacob M, Ahmad N, Toogood GJ, et al. (2009) Inflammatory pseudotumors of the liver: A spectrum of presentation and management options. Eur J Surg Oncol 35: 1295-1298.
- Nagarajan S, Jayabose S, McBride W, Prasad I, Tanjavur V, et al. (2013) Inflammatory myofibroblastic tumor of the liver in children. J Pediatr Gastroenterol Nutr 57: 277-280.
- Saito M, Seo Y, Yano Y, Miki A, Morinaga Y, et al. (2012) Sonazoid-enhanced ultrasonography and ga-EOB-DTPA-enhanced MRI of hepatic inflammatory pseudotumor: A case report. Internal Med 51: 723-726.
- Wang JH, Chen TY, Ou HY, Wang CC, Liu YW, et al. (2016) Clinical impact of gadoxetic acid-enhanced magnetic resonance imaging on hepatoma management: A prospective study. Digest Dis Sci 61: 1197-1205.
- Wu LM, Xu JR, Gu HY, Hua J, Chen J, et al. (2013) Is liver-specific gadoxetic acid-enhanced magnetic resonance imaging a reliable tool for detection of hepatocellular carcinoma in patients with chronic liver disease? Digest Dis Sci 58: 3313-3325.
- Liu X, Zou L, Liu F, Zhou Y, Song B (2013) Gadoteric acid disodium-enhanced magnetic resonance imaging for the detection of hepatocellular carcinoma: A meta-analysis. PLoS one 8: e70896.
- Iguchi H, Yamazaki H, Tsunoda H, Takahashi Y, Yokomori H (2013) A case of inflammatory pseudotumor of the liver mimicking hepatocellular carcinoma on EOB-MRI and PET. Case Rep Med 2013: 594254.
- Saito M, Seo Y, Yano Y, Miki A, Morinaga Y, et al. (2012) Sonazoid-enhanced ultrasonography and Ga-EOB-DTPA-enhanced MRI of hepatic inflammatory pseudotumor: A case report. Intern Med 51: 723-726.