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Case Report
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Inflammatory Pseudotumor of the Liver Misdiagnosed Malignancy: A Case Report

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Abstract

Inflammatory Pseudotumor of Liver (IPL) is a rare disease, which characterized by heterogeneous population of acute and chronic inflammatory cell. The etiology of inflammatory pseudotumor is unclear, and it is often confused with hepatic malignant tumors due to atypical clinical manifestation. Because inflammatory pseudotumor can regress spontaneously or be managed conservatively, it is important to differentiate IPL from malignant hepatic tumor before operation, in order to avoid unnecessary surgery. Although IPL may spontaneously regress or regress following antimicrobial and steroidal treatment, surgery is the first choice of treatment yet. Herein, we reported a case of IPL and reviewed the literature.

Introduction

Inflammatory Pseudotumor of Liver (IPL) is a rare disease, which characterized by heterogeneous population of acute and chronic inflammatory cell. It is often confused with hepatic malignant tumors due to atypical clinical manifestation [1-5]. Because inflammatory pseudotumor can regress spontaneously or be managed conservatively, it is important to discriminate it from malignant hepatic tumor before operation, in order to avoid unnecessary surgery [4,6-8]. Herein, we reported a case of IPL and reviewed the literature.

Case Presentation

A 57-year-old man was admitted because of dull pain and discomfort in the right upper quadrant of the abdomen. He had been treated at another hospital for the presumptive diagnosis of liver tumor. Sonographic examination at that hospital disclosed a hepatic mass for which he was transferred to our hospital for further evaluation. He had no remarkable familial or personal medical histories, such as pulmonary tuberculosis and viral B hepatitis.

Physical examination on admission revealed no abnormal findings. Initial laboratory tests revealed the following: white blood cell 9.0 × 10^9/L, Hemoglobin (Hb) 97 g/L, Erythrocyte Sedimentation Rate (ESR) 25 mm/hr, serum alkaline phosphatase concentration 61 u/L (53 u/L to 128 u/L), serum aspartate aminotransferase level l20 u/L (5 u/L to 55 u/L), serum alanine aminotransferase level 28 u/L (5 u/L to 60 u/L), and serum bilirubin level 8 μmol/L (0 μmol/L to 12 μmol/L), Alpha-Fetoprotein (AFP) 2.6 μg/L (0 μg/L to 20.0 μg/L), Carcinoembryoneal Antigen (CEA) 3.0 μg/L (0 μg/L to 5.0 μg/L), CA19-9 16 μg/L (0 μg/L to 37.0 μg/L). Antibodies for hepatitis B virus and HIV were negative. Ultrasonography and abdomen CT scan revealed that an heterogeneous lesion of ellipse-like isodense with peripheral rim like hypodense was in the left lobe of the liver (Figure 1A), and the peripheral rim like hypodense became clearer after enhanced scan, compared with the internal low density area (Figure 1B).

On laparotomy, a tumor of 6.5 cm in diameter, gray in color, was found (Figure 1C) in the left lobe of the liver. The patient underwent irregular resection of the tumor around the outside 2 cm for the presumptive diagnosis of hepatocellular carcinoma. On microscopic examination, the mass consisted of collagenous fibrous tissue infiltrated with chronic inflammatory cells such as plasma cells, lymphocytes, and histiocytes. The postoperative course was uneventful, and the patient is alive without evidence of recurrence five years later.

Discussion

Inflammatory pseudotumor of liver is a rare benign disease, which is characterized by a prominent inflammatory infiltrate, namely, plasma cells, lymphocytes, and eosinophils as the predominant

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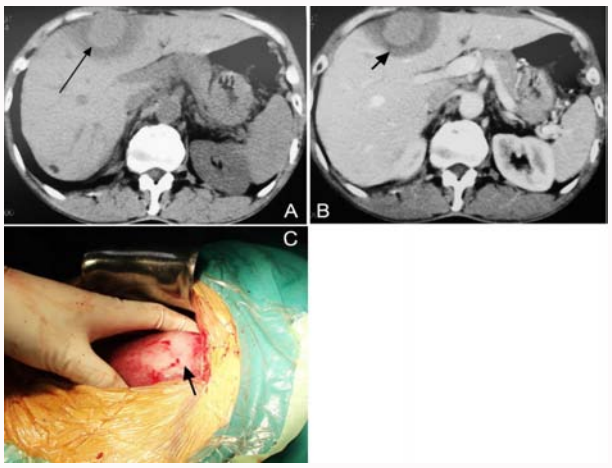

Figure 1: Inflammatory pseudotumor of the liver. **A:** Plain CT scan showed an irregular heterogeneous ellipse-like lesion of isodense with peripheral rim like hypodense in the segment 4 of the liver; **B:** Enhanced CT scan showed that the peripheral ring-shaped hypodense became clearer; **C:** The photo taken during the procedure showed a lesion of 6.5 cm in diameter.

cellular component [4-5,9]. The etiology and pathogenesis of this disease is not clear, and underlying factors such as infection, IgG4 sclerosing cholangitis, radiation, and chemotherapy, have been reported to relate to this disease [8,9,10]. Torzilli et al., [1] have reported 3 cases (0.7%) of inflammatory pseudotumor that were found among 403 patients who had undergone liver resection. The etiology of inflammatory pseudotumor has not yet been very clear, and may be related to infection and immunological compromise. Yang et al., [3] retrospective analyzed 114 Hepatic Inflammatory Pseudotumors (HIPT) cases with surgical treatment, the mean age was 53.14 ± 10.98 years, and most presented symptoms were abdominal pain (59/144, 41.0%), fever (48/114, 42.1%), abdominal distension (35/144, 24.3%), and weight loss (12/144, 8.3%). Our patient only had nonspecific dull pain and discomfort in the right upper quadrant of the abdomen.

IPL is often confused with the malignant tumors, the accuracy of the pre-surgical diagnosis was low [1-5,9,11,12]. According to the report from Yang et al., [3], most of the tumors were located in the right lobe (79/114, 69.3%), 33 in the left lobe, and 2 in the caudal lobe. Radiological features of inflammatory pseudotumor of liver are atypical, it is often demonstrated have hypo- or isodensity on CT, hypo- or isointense on T1WI and hyper- or isointense on T2WI on magnetic resonance imaging [4-5,11]. Most cases were normal in laboratory examinations, and some cases may with leukocytosis or increased CRP. Our patient had normal laboratory test results, and nonspecific imaging of the occupying lesion of the liver.

Numerous studies have shown that the inflammatory pseudotumor may regress naturally [6-8]. Once the diagnosis of inflammatory pseudotumor has been confirmed with biopsy, patients with inflammatory pseudotumor can simply be observed and regular follow-up is performed until the condition resolves itself, or the patient can be medically treated with antibiotics, anti-inflammatory drugs and steroid [4-8,11]. Because of the potential risk of tumor dissemination and hemorrhage, percutaneous needle biopsy was not performed in this patient, and final diagnosis is made by post pathologic examination.

Although IPL may spontaneously regress or regress following antimicrobial and steroidal treatment, nowadays, surgery is the first choice of treatment, especially for patients with the following situation: 1. the cases that is resistant to non-surgical therapy, those systemic symptoms with fever do not resolve, for example; 2. the cases whose liver mass are markedly grow, with or without symptoms, on serial examinations and imaging studies; 3. the cases whose liver mass involve the hepatic hilum, subsequently causes biliary obstruction and portal hypertension; 4. the cases are with high suspicious of liver malignancy [2,4-7,9,10].

It has been reported that inflammatory pseudotumor has the potential of malignant transformations and recurrences 4 to 7 years after operation in literature [12,13]. Therefore, long-term follow-up is necessary even for patients who have been successfully treated by surgical resection [14].

Conclusion

In conclusion, inflammatory psuedotumor of the liver is a rare tumor-like lesion, and it is important to differentiate this disease from other liver tumor, the treatment of choice is still surgery because of difficult preoperative diagnosis.

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