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Case Report

Inflammatory Pseudotumor of the Liver

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Abstract

Inflammatory pseudotumor (IPT) of the liver is a rare benign lesion, characterized by chronic infiltration of inflammatory cells and areas of necrosis mimicking a malignant tumor. Few cases have been reported, and the precise etiology is still unknown. Patients usually present with abdominal pain, fever, and jaundice. Herein, we report the case of a 78-year-old male with a history of diabetes mellitus who had abdominal fullness and body weight loss for 4 months. A computed tomography scan showed a 15-cm liver tumor in segment 2 and 3 and suspected hepatocellular carcinoma. Left hepatectomy was performed, and the pathology showed IPT. After surgery, the symptom of abdominal fullness subsided.

Keywords: Hepatocellular carcinoma, inflammatory pseudotumor, liver

Introduction

Inflammatory pseudotumor (IPT) is a rare condition first described in the lungs in 1939.[1] These lesions most commonly occur in the lungs, followed by the liver.[2] IPT has also been reported in the heart, pancreas, spleen, stomach, small intestine, thyroid, meninges, urinary bladder, and orbits.[3,4] Macroscopically, the lesion may mimic a malignancy and may be alone or several lesions may be present. The lesion may be as large as 25 cm. Microscopically, IPT is characterized by spindle-shaped cells, myofibroblasts, and mixed inflammatory cells (plasma cells, lymphocytes, and sporadic histiocytes)[5,6] and is frequently confused with a malignant tumor.[7] Hepatic IPT (HIPT), first described by Pack and Baker in 1953,[8] is a rare lesion, with <300 cases described in the medical literature.[9,10] HITP most commonly occurs in males in their mid-thirties. The etiology and pathogenesis of HIPT are still unknown, and there are no specific symptoms or laboratory or radiological findings that are useful in diagnosing HIPT. Differentiating between HITP and other focal hepatic lesions remains a major problem. The treatment of choice is still surgical resection, especially for the patient with severe symptoms or an indeterminate diagnosis.[11,12]

Case Report

A previously hypertensive 78-year-old male with a history of diabetes mellitus was referred to our outpatient department due to abdominal fullness, weight loss, and

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fatigue for 4 months. He denied fever and jaundice. On physical examination, hepatomegaly was remarkable. Laboratory findings included Hb 10.5 mg/dl, leukocyte count 25,600/uL with left shift, C-reactive protein (CRP) 7.55 mg/dL (reference range: 0–1 mg/dL), normal hepatic transaminases, alkaline phosphatase, and total bilirubin, hepatitis B surface antigen (-), anti-hepatitis C virus (-), alpha-fetoprotein 2 ng/mL (reference range: 0–9 ng/mL) carcinoembryonic antigen (CEA) 4.5 ng/mL (reference range: 0–5 ng/mL), and CA19-9 22.8 U/mL (reference range: 0.8–35 U/mL).

Abdominal sonography showed a solid lesion of approximately 13 cm occupying segments 2 and 3. Abdominal computed tomography (CT) showed a huge, well-defined heterogeneous and gradually enhancing mass measuring around 14 cm replacing almost the entire left hepatic lateral segment. The associated exophytic component compressed the stomach, part of the pancreas, and left portal vein [Figures 1-3]. A malignant lesion was suspected. Upper gastrointestinal endoscopy confirmed a hepatic lesion with gastric and duodenal compression. The patient's clinical status did not improve after admission, and therefore, left hepatectomy was performed. Histopathological analysis showed a specimen consisting of a segment of the liver measuring 19.5 cm × 12.5 cm × 11.5 cm in size and 1250 g in weight, in a fresh state. The capsule was intact with fibrotic thickening. Cut sections demonstrated a demarcated, solid, and fleshy tumor measuring 15 cm × 12 cm × 11.5 cm in size located 0.2 cm away from the resection margin. Foci of necrosis and hemorrhage were also noted. The small rim of nonneoplastic hepatic tissue included was not cirrhotic. Sections of the specimen labeled as "liver" submitted for a microscopic examination showed an IPT (inflammatory myofibroblastic tumor) composed of fascicles of bland-looking spindle-shaped fibroblasts or myofibroblasts with a mixture of inflammatory cells including many plasma cells, lymphocytes, neutrophils, and even eosinophils. Granulation tissue-like areas and focal necrosis were also seen. The resection margin was free [Figures 4-7]. The final diagnosis was HIPT.

There were no postoperative complications, and the patient was discharged from the hospital on the 14th postoperative day. He is currently being followed up regularly and has remained asymptomatic.

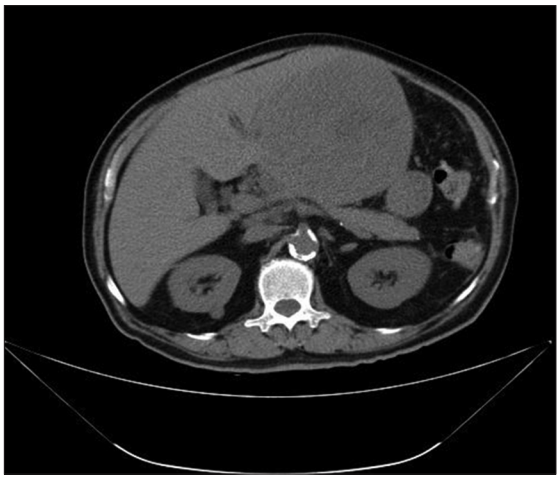

Figure 1: A huge, well-defined heterogeneous attenuating mass measuring around 14 cm replacing almost all of the left hepatic lateral segment, with an associated exophytic component

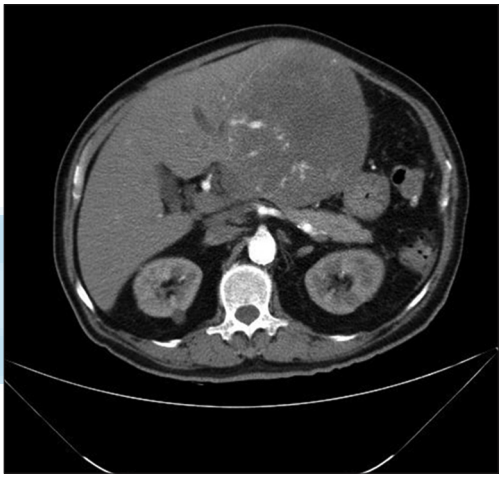

Figure 2: Minimal tumor stain in the arterial phase

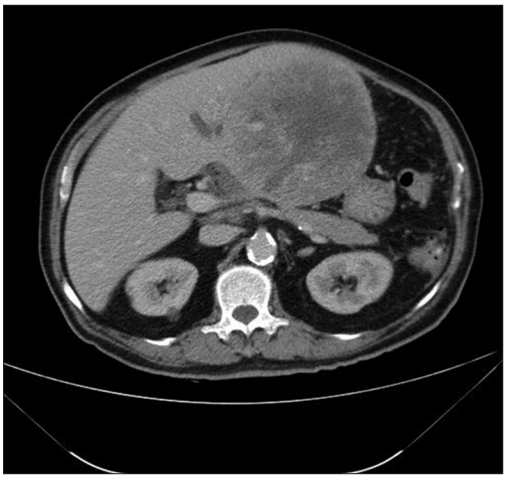

Figure 3: Gradual peripheral enhancement with an obvious central necrotic component in the delayed phase

Discussion

HIPT is a rare disease that lacks specific symptoms and laboratory or radiologic findings. The exact pathogenesis of HITP has not been well characterized; however, the inflammatory pathological pattern and systemic symptoms include fever and malaise. The predominantly inflammatory pattern of the pathology and the associated laboratory findings suggest an underlying infection via the hepatobiliary tract; biliary stones, history of liver resection, cholangitis, liver abscess, and hepatobiliary malignancy have been reported in 68%–80% of cases.[13-16] Abdominal pain; fever; and elevated inflammatory markers including erythrocyte sedimentation rate, CRP, and leukocyte count are common in cases with HIPT;[9,17] however, serum AFP, CA 19-9, and CEA level

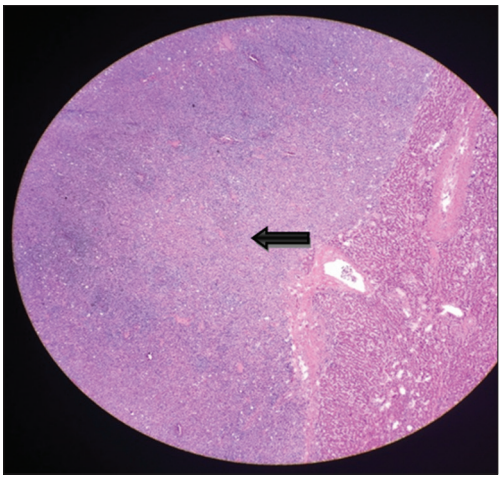

Figure 4: A well-demarcated solid mass over the left half (arrow) and normal liver tissue in the right half (×4)

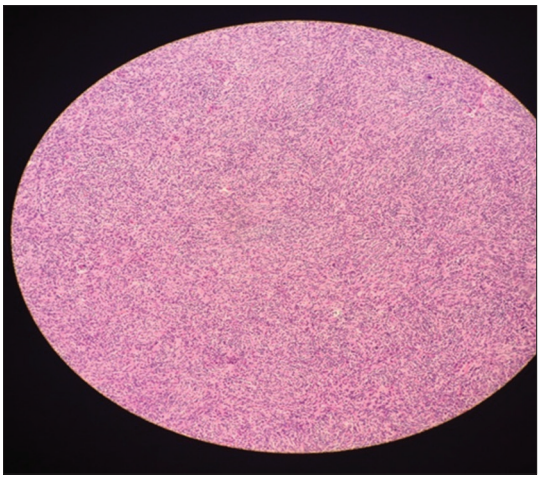

Figure 5: Fascicles of bland-looking spindle cells and inflammatory cell infiltrates (×10)

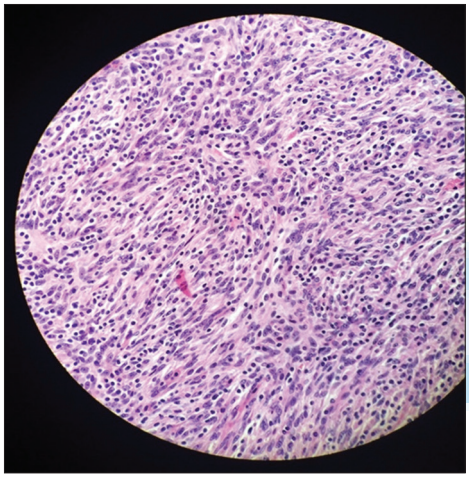

Figure 6: Fibrohistiocytic proliferation (×40)

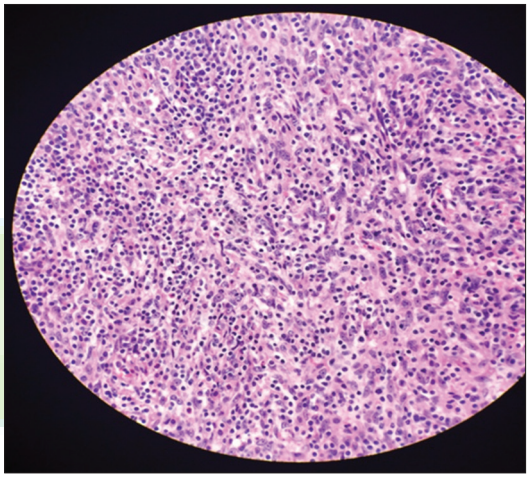

Figure 7: Mixed inflammatory cell infiltration, including lymphoproliferative cells, neutrophils, and eosinophils (×40)

are normal in most patients. The differential diagnosis from a malignant tumor can be difficult because the radiologic findings of HIPT are rather nonspecific.[18] A percutaneous biopsy is the most reliable method, and it enables avoiding unnecessary exploratory laparotomy or hepatectomy when there is an uncertain diagnosis. Numerous studies have shown that the natural history of HIPT is one of the disease regressions. Once the diagnosis of HIPT has been confirmed with a biopsy, the patients can simply be observed with regular follow-up until the condition resolves itself, or the patient can be medically treated with antibiotics, anti-inflammatory drugs, and steroids.[19] The role of a biopsy is more contentious and possibly dangerous in cases of a solitary hepatic mass that is likely to be malignant.[20] Even though HIPT may regress spontaneously or following antibiotic treatment, the treatment of choice is still surgical resection, and this is especially true for patients with severe symptoms or an indeterminate diagnosis.[21] Hepatectomy has become a safer option for noncirrhotic patients over the past 20 years, with the mortality rate falling to 0%.[22] Therefore, the treatment of choice should be surgical resection in such cases.[23] This approach is preferable because it minimizes the risk of biopsy-related complications (dissemination in cases of malignancy) and it eliminates the possibility of HIPT recurring.

Conclusion

HIPT is a rare condition, and differentiating this pseudotumor from hepatic space-occupying lesions is crucial. HIPT may regress spontaneously, and it may mimic other liver tumors. The treatment of choice for patients with severe symptoms or an indeterminate diagnosis is surgical resection.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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