Inflammatory Pseudotumor of the Liver: A Case Report

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ABSTRACT. We report a surgical case of inflammatory pseudotumor of the liver. A 66-year-old man was admitted to our hospital to evaluate a high grade fever and a mass in the right hepatic lobe. We performed hepatic subsegmentectomy because of possible malignancy. The cut section of the resected tumor was yellowish white and firm. It was well circumscribed without encapsulation, measuring 4.0×2.5 cm. Microscopic examinations revealed proliferation of fibroblasts as well as chronic inflammatory cells. It was diagnosed as an inflammatory pseudotumor (xanthogranulomatous type). It is difficult to differentiate between inflammatory pseudotumor and malignant tumor of the liver preoperatively despite the progress in diagnostic tools. Most cases are diagnosed postoperatively by a pathologic examination. Our case indicates that a fever should raise the possibility of inflammatory pseudotumor of the liver in non-cirrhotic patients with a solitary mass and an aspiration biopsy may be helpful.

Key words: inflammatory pseudotumor — liver — fever — non-cirrhotic liver — aspiration biopsy

Inflammatory pseudotumor has been reported in various organs such as the lung, heart, uterus, urinary bladder, and spleen.[1-5] Such tumors have also been called plasma cell granuloma, histiocytoma, fibroxanthoma, or xanthogranuloma. However, only less than 70 cases have been reported in the liver[6-15] since the first report by Pach and Baker in 1953.[16] Although the exact pathogenesis is not clear, it is thought to be an inflammatory process rather than neoplastic. In most cases, it is difficult to differentiate it from malignant tumors and is often diagnosed after surgery.[6,11]

CASE REPORT

A 66-year-old man was admitted to our hospital to evaluate a mass in the liver. He had a one week history of high grade fever (39 to 40°C) which was treated with antibiotics at other hospital. On physical examination, liver and spleen were not palpable and lymphadenopathy was not found. Laboratory data on admission were within normal limits. Ultrasonography (US) of the liver revealed a hypoechoic mass in the right posterior lobe (Fig 1). Computed

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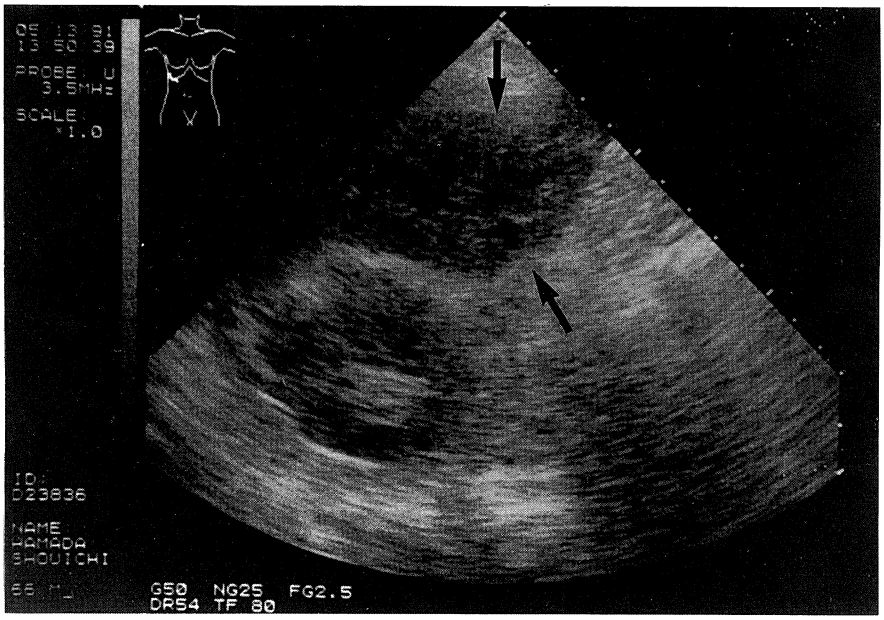

Fig 1. Ultrasonography of the liver demonstrates a hypoechoic mass.

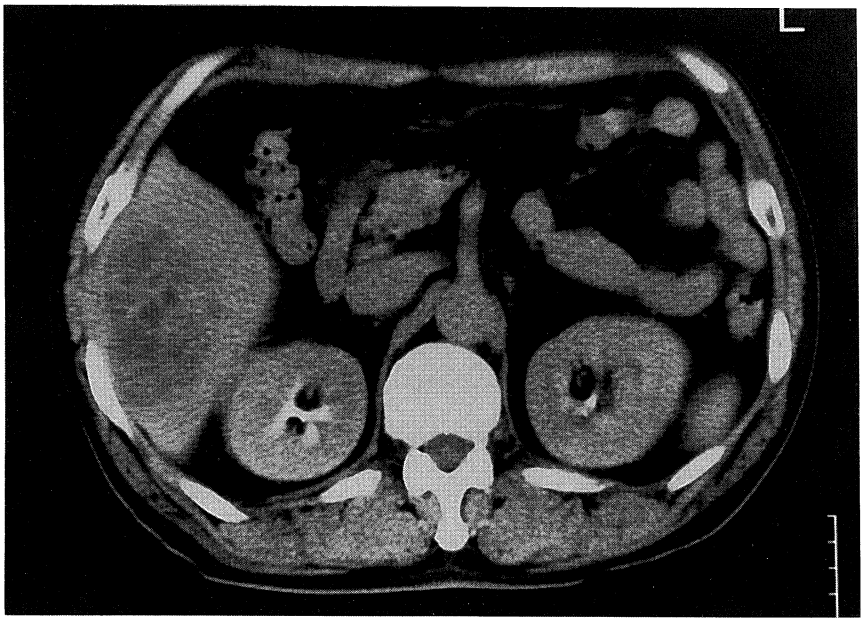

Fig 2. Computed tomography showing a 8.0×5.0 cm low density mass with inhomogenous pattern.

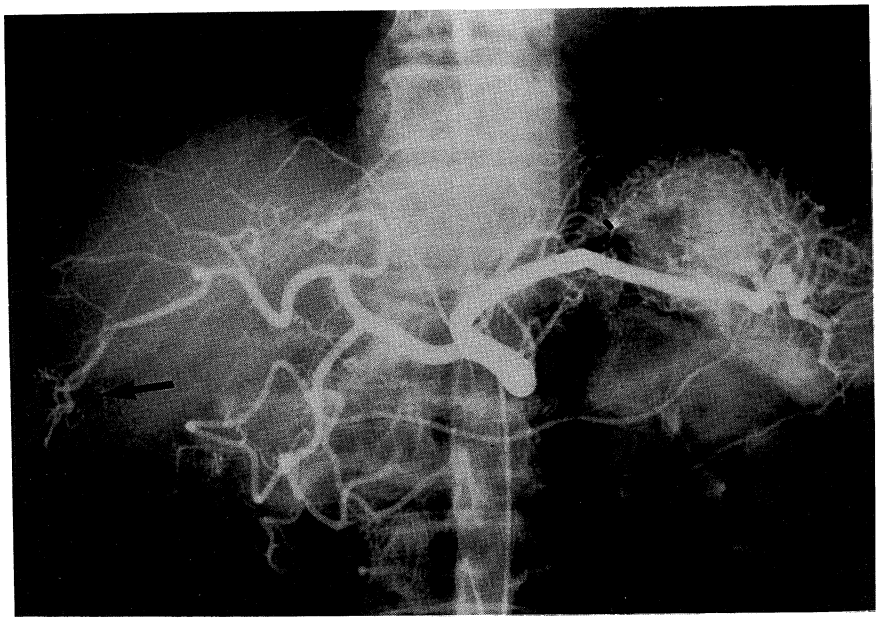

Fig 3. Selected hepatic angiography reveals a slightly hypervascular tumor in the right posterior lobe of the liver.

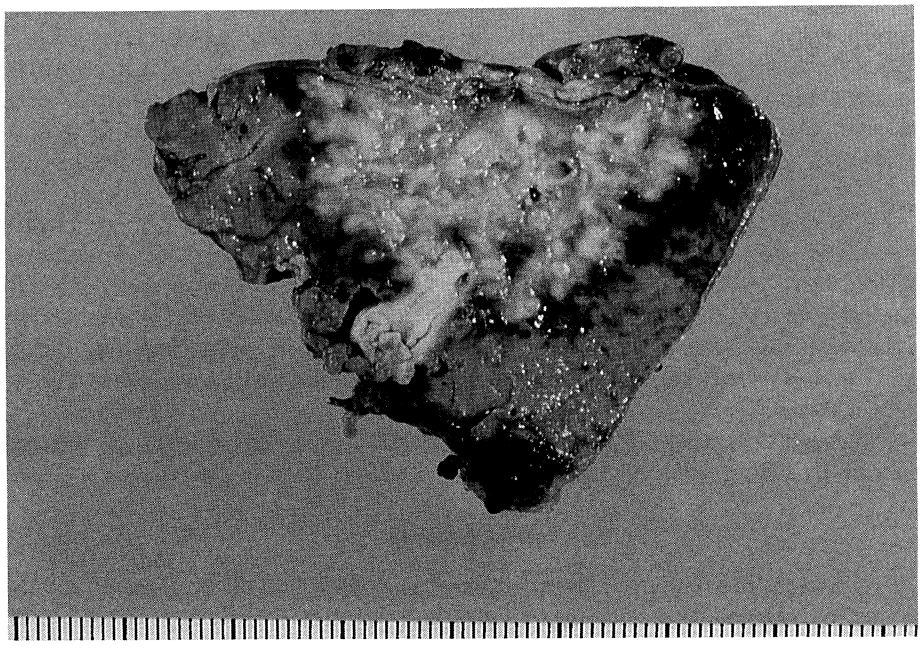

Fig 4. Resected mass measuring 4.0×2.5 cm. No capsule is observed.

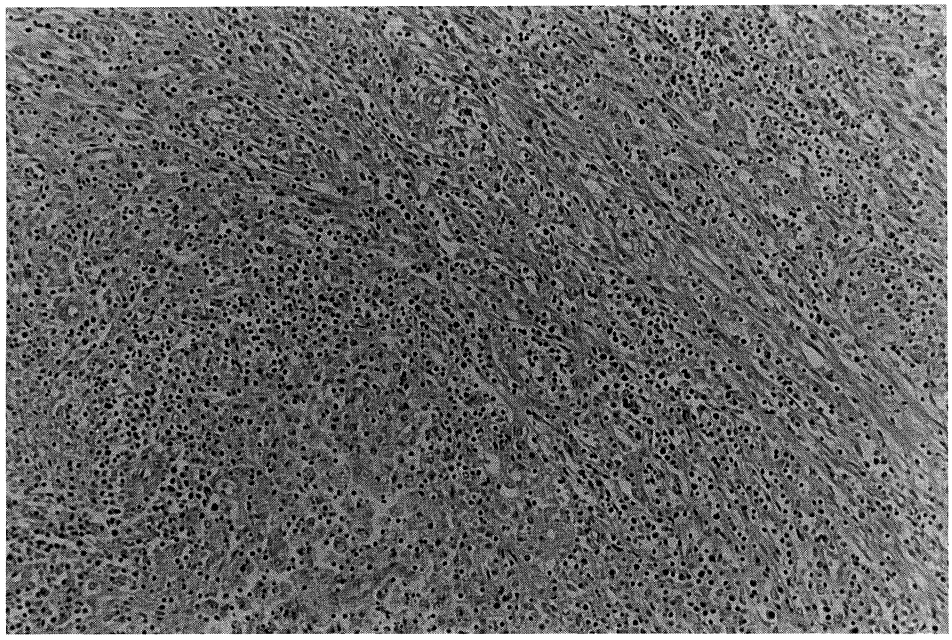

Fig 5. Microscopically, tumor is composed of proliferation of fibroblasts with foamy histiocytes, plasma cells, and lymphocytes.

tomography (CT) showed a mass measuring 8.0×5.0 cm. The mass was a low density area and its inner part was inhomogenous pattern (Fig 2). There was no evidence of liver cirrhosis. Selected hepatic angiography demonstrated a slightly hypervascular tumor in the right posterior lobe of the liver (Fig 3). Under these findings, either abscess or hepatocellular carcinoma was considered the preoperative diagnosis. We performed subsegmentectomy of the hepatic posterior lobe. Immediately after resection of the tumor, aspiration biopsy was performed. It showed normal looking hepatocytes and fibroblasts with lymphocytes. No evidence of malignancy was noted.

The resected mass was yellow and firm, measuring 4.0×2.5 cm (Fig 4). There was no capsule. Microscopically, it was composed of proliferating fibroblasts with foamy histiocytes, plasma cells and lymphocytes (Fig 5). Foreign body type giant cells and Russel body were occasionally seen, but no microorganisms were observed by special stains, such as periodic acid-Schiff and Ziehl-Neelsen stains. The diagnosis of inflammatory pseudotumor was made.

The postoperative clinical course was excellent. The patient has been well without any symptoms or recurrence for 5 years since his discharge.

DISCUSSION

Although inflammatory pseudotumor is a well known entity and reported in various organs, its occurrence in the liver is rare despite recent progress in diagnostic imaging.[6-15] According to Shek *et al* extensive review of the

literature,[6] the male-to-female ratio was 2.9 and the average age was 37 years. The pathogenesis is still not clear. The common symptoms were upper abdominal pain, fever, general fatigue, and weight loss. Jaundice and abdominal mass were observed in some cases.[6] Concerning laboratory findings, some cases of inflammatory pseudotumor showed leukocytosis, elevated erythrocyte sedimentation rate, and hyper-gamma globulinemia suggesting the inflammatory nature of this lesion.[17,18] However, our case only developed a high grade fever; laboratory findings were within normal limits. Therefore, it is better to remember that a fever may be the only clue for diagnosis of inflammatory pseudotumor of the liver in non-cirrhotic patients with liver mass. From this point of view, we analyzed the Shek's review[6] and found 28 out of 51 cases (55%) showing fever. Past history of traveling to Southeast Asia and India,[19] which is not found in our case, is also important.

On diagnostic imaging, the lesion in most cases was observed as a low density area on CT and a mosaic pattern on US and even by magnetic resonance imaging (MRI) it is not specific.[8,9,13,14] The correct preoperative diagnosis is often difficult, and diagnosed as a malignant tumor is common. Therefore, hepatectomy is performed in about 60% of reported cases.[6] Pathologic findings are yellowish elastic firm mass macroscopically, and infiltration of chronic inflammatory cells mainly consisting of foamy histiocytes and plasma cells, microscopically.[6] Occlusive phlebitis has been reported in some cases.[19] Someran[20] classified the following three histologic types: 1) hyalinized sclerosing type, 2) xanthogranulomatous type, 3) plasma cell granuloma type. The different histologic types may reflect the variation of the disease process or period. Our case was diagnosed as a xanthogranulomatous type. Concerning the diagnosis, aspiration biopsy may be useful,[6] since our aspiration biopsy immediately after resection of tumor revealed no evidence of malignancy. The prognosis for inflammatory pseudotumor of the liver is fair.[6]

Since Shek *et al* extensive review in 1993,[6] about 20 new cases has been reported for these three years.[7–15] This is because the number of patients diagnosed with inflammatory pseudotumor of the liver has improved with progress in imaging techniques at least about its presence of tumor. A relative longer course of disease, fewer clinical symptoms, good general condition are said to be supporting features.[21] In addition, our case indicated that inflammatory pseudotumor should be considered, if the patient with a hepatic mass in non-cirrhotic liver is febrile and in such a case intraoperative or preoperative aspiration biopsy may be useful to make a correct diagnosis before resection of the tumor. Finally, recent report mentioned that some cases of inflammatory pseudotumor, particularly those of the liver and spleen, are EBV-positive inflammatory follicular dendritic cell tumors.[22,23] This findings has to be evaluated in the future study.

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