Surg Today (2003) 33:714–717
DOI 10.1007/s00595-003-2561-z

SURGERY TODAY

Hepatic Inflammatory Pseudotumor Mimicking Intrahepatic Cholangiocarcinoma: Report of a Case

KEISUKE INABA[1], SHOHACHI SUZUKI[1], YOSHIHIRO YOKOI[1], SHIGEYASU OTA[1], TOSHIO NAKAMURA[1], HIROYUKI KONNO[1], SATOSHI BABA[2], YASUO TAKEHARA[3], and SATOSHI NAKAMURA[1]

[1] Second Department of Surgery, [2] Second Department of Pathology, and [3] Department of Radiology, Hamamatsu University School of Medicine, 1-20-1 Handayama, Hamamatsu 431-3192, Japan

Abstract
Hepatic inflammatory pseudotumor (IPT) is a relatively rare lesion comprised of proliferating fibrovascular tissue infiltrated by inflammatory cells. IPT has a potential for recurrence and persistent local growth. We present a case of hepatic IPT mimicking a periductal-infiltrating type of intrahepatic cholangiocarcinoma (ICC) in a patient whose serum carbohydrate antigen 19-9 was slightly elevated. We performed a left hepatic lobectomy with resection of the extrahepatic bile duct and regional lymph node dissection under a preoperative diagnosis of ICC. However, histlogical examination of the resected tumor revealed granuloma tissue with lymphocyte infiltration, mainly by plasma cells, and proliferation into the surrounding connective tissue, and the lesion was ultimately diagnosed as hepatic IPT. This case points out the difficulties in differentiating between hepatic IPT extending along Glisson's sheath and ICC, based on imaging findings alone.

Key words Liver · Inflammatory pseudotumor · Intrahepatic cholangiocarcinoma · Hepatic lobectomy

Introduction

Inflammatory pseudotumor (IPT) arises most commonly in the lungs, but it can also develop in the stomach, the intestinal mesenterium, and the liver.[1–3] This tumor is generally defined as a localized mass consisting of proliferating fibrovascular tissue infiltrated by inflammatory cells.[2–4] Although hepatic IPT is generally considered to be a relatively rare lesion, the number of case reports is increasing with progress in medical imaging modalities. We recently treated a patient initially thought to have intrahepatic cholangiocarcinoma (ICC) based on laboratory data and medical imaging, but who was later found to have hepatic IPT. This case is reported because of the difficulty in differentiating hepatic IPT from ICC.[5]

Reprint requests to: K. Inaba
Received: January 5, 2002 / Accepted: January 21, 2003

Case Report

A 72-year-old Japanese man was referred to our department after an abdominal computed tomography (CT) scan showed a dilated intrahepatic bile duct in the lateral segment of the liver and disappearance of the left branch of the portal. He had undergone a laparoscopic cholecystectomy for cholecystolithiasis 5 years earlier, followed 3 years later by surgical resection of advanced rectal carcinoma. Since then, abdominal CT scans had been performed every 6 months as follow-up, but no abnormal findings were detected until just prior to this presentation.

Abdominal magnetic resonance imaging (MRI) showed a slight low-intensity lesion with dilatation of the intrahepatic bile duct on T1-weighted imaging (Fig. 1A) and a slight high-intensity lesion on T2-weighted imaging (Fig. 1B) extending along Glisson's sheath of the lateral segment of the liver. Thus, he was admitted to our hospital for further investigations. On admission, he was free from symptoms and in good general health, without pyrexia or jaundice. The liver and spleen were not palpated and there was no sign of any abdominal mass. Abnormal laboratory findings included glutamic oxaloacetic transaminase, 43 IU/l (normal range: 11–30:IU/l); glutamic pyruvic transaminase, 64 IU/l (5–42 IU/l); γ-guanosine triphosphate, 208 IU/l (12–73 IU/l); and carbohydrate antigen (CA) 19-9, 56 U/ml (3–34 U/ml). There was no elevation of C-reactive protein (<0.01 mg/dl). Serology for hepatitis B and hepatitis C virus were negative. Abdominal ultrasonography (US)

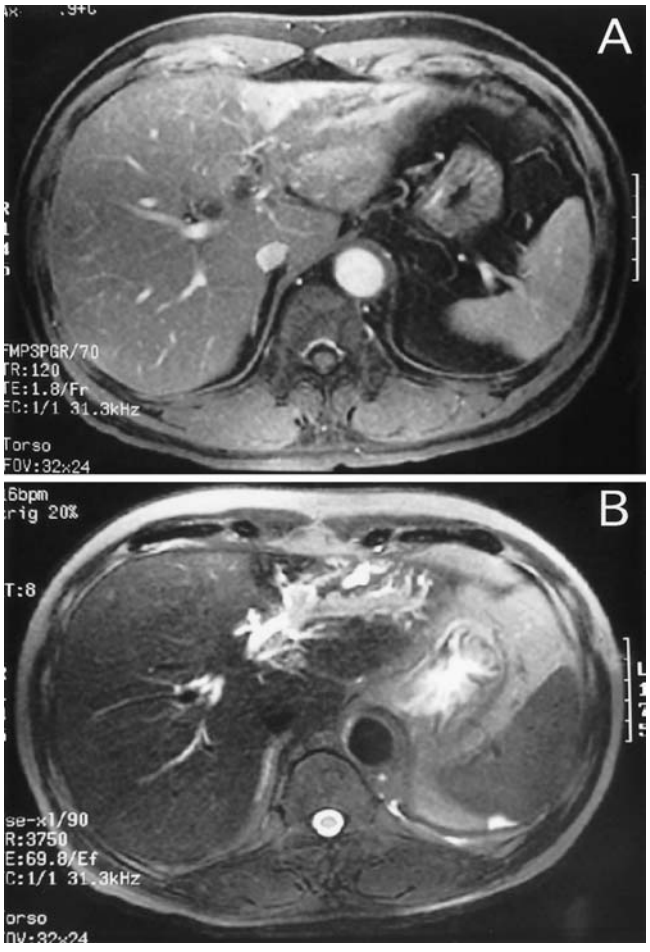

Fig. 1. Magnetic resonance imaging showed a low-intensity lesion extending laterally with dilatation of the intrahepatic bile duct on T1-weighted imaging (**A**) and a slight high-intensity lesion mimicking intahepatic cholangiocarcinoma on T2-weighted imaging (**B**)

showed a high echogenic tumor at the hepatic hilum of the left lobe with dilated intrahepatic bile ducts. The blood flow signal of the left branch of the portal vein was not detected by Doppler scanning. Segment 3 of the intrahepatic bile duct was not visualized on endoscopic retrograde cholangiography (ERC) (Fig. 2). A hepatic arteriogram showed stretching of the lateral segment of the artery, without any sign of a hypervascular tumor (Fig. 3A). The left branch of the portal vein was stenotic with irregularity in the venous phase of a selective superior mesenteric arteriogram (Fig. 3B). We did not perform US-guided biopsy of the lesion because of the risk of needle-track or intraperitoneal seeding of tumor cells. The possibility of a hepatic malignancy such as the periductal-infiltrating type ICC was suspected from the combination of medical imaging findings and the elevated CA19-9 level. A laparotomy was performed 3 weeks after admission. A tumor extending to the hepatic hilum was palpated in the left hepatic lobe

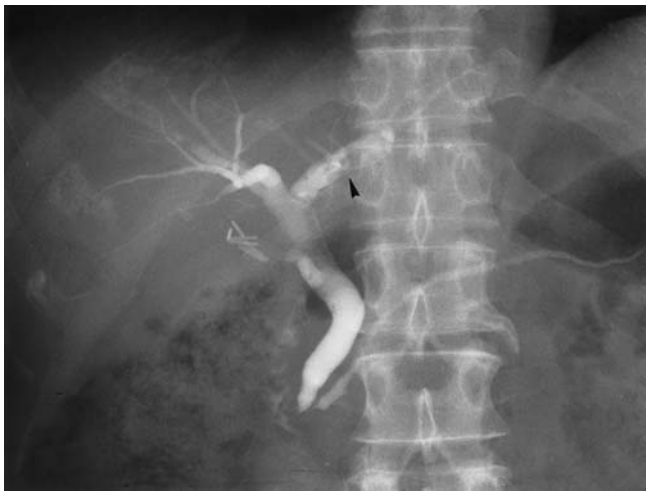

Fig. 2. Endoscopic retrograde cholangiography showed no image of segment 3 of the intrahepatic bile duct (*arrowhead*)

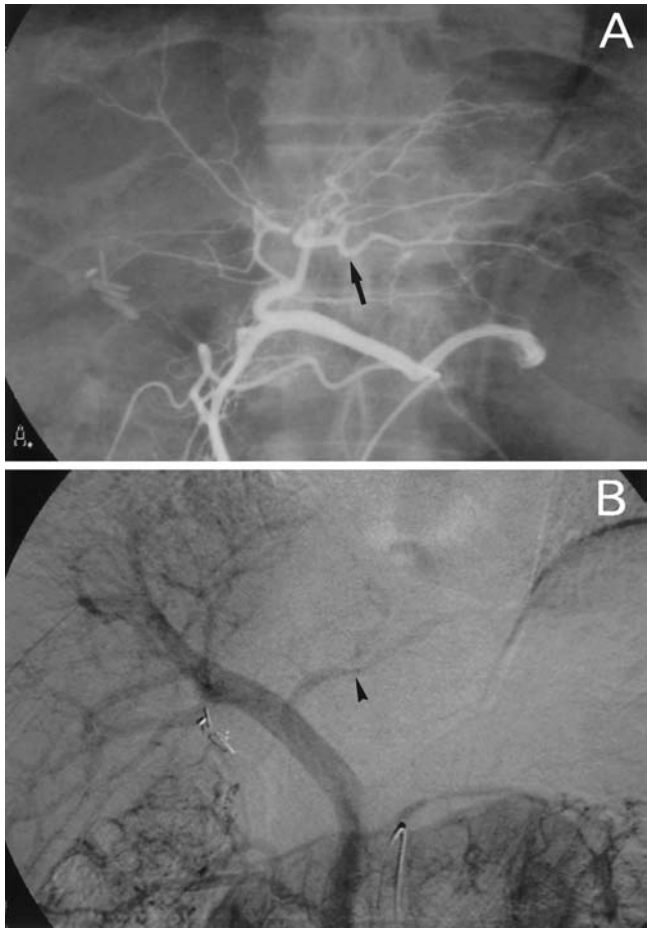

Fig. 3. **A** Hepatic arteriogram showed stretching of the lateral segment of the artery without any evidence of a hypervascular tumor (*arrow*). **B** The left branch of the portal vein was stenotic with irregularity in the venous phase of a selective superior mesenteric arteriogram (*arrowhead*)

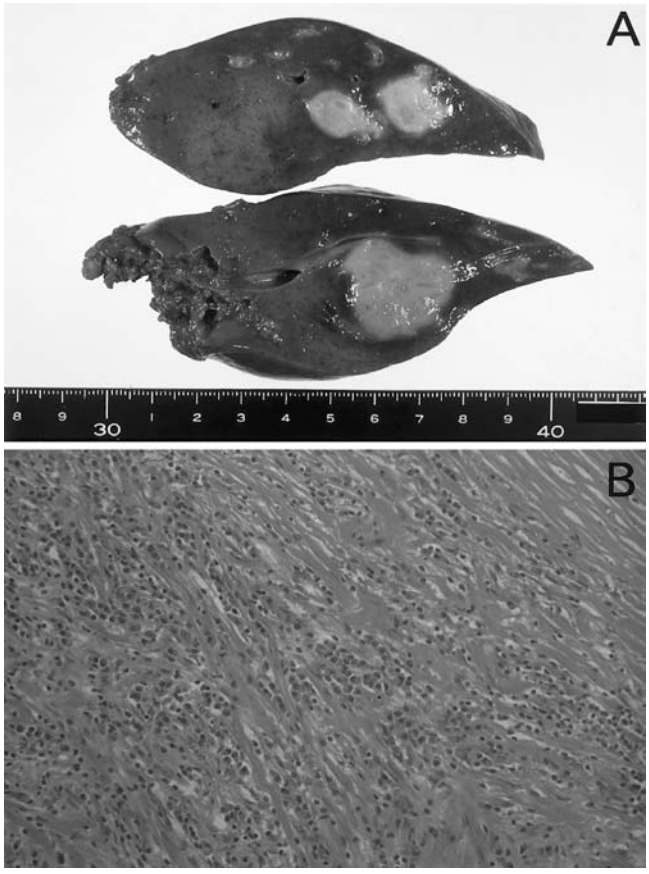

Fig. 4. A Sagittal cut surface of the resected specimen revealed well-circumscribed yellowish-white tissue growing along Glisson's sheath of the left lateral hepatic lobe. **B** Histological examination showed granuloma tissue with lymphocyte infiltration, mainly by plasma cells, and proliferation into the surrounding connective tissue. These histological findings were consistent with a diagnosis of hepatic inflammatory pseudotumor (plasma cell type) (H&E, ×200)

and the lymph nodes in the hepatoduodenal ligament were remarkably enlarged. Therefore, a left hepatic lobectomy with extrahepatic bile duct resection and lymph node dissection was performed. The right hepatic duct was anastomosed with a Roux-en-Y jejunal loop. The resected specimen contained yellowish-white tumors (25 × 23 × 50 mm in size) growing along Glisson's sheath of the left hepatic lobe. These tumors were firm and clearly circumscribed, despite the absence of capsules. The adjacent hepatic parenchyma was unremarkable (Fig. 4A). Tumor histology revealed granuloma tissue with lymphocyte infiltration mainly by plasma cells and proliferation into the surrounding connective tissue. These histological findings confirmed a diagnosis of hepatic IPT of the plasma cell type (Fig. 4B). The lesions revealed infiltrative growth along Glisson's sheath causing occlusion of the portal vein. The patient had an uneventful postoperative course and was discharged 1 month after surgery, by which time his serum CA19-9 level had decreased to within the normal range (20 U/ml). He has been well for 2 years since the operation, with no sign of recurrence.

Discussion

Since the first case report by Pack and Baker in 1953,[4] more than 200 cases of hepatic IPT have been documented.[6] Hepatic IPT has been reported to account for 0.4% of all focal liver lesions treated by hepatectomy.[7] IPT has been identified as a lesion associated with a local inflammatory reaction, and recurrent bacterial infection has been postulated as its etiology. Since degeneration and necrosis of the bile duct wall with subsequent periductal abscess is caused by chronic cholangitis and calculi-associated bile stasis,[8] hepatic IPT may result from recurrent cholangitis. Other studies have indicated that hepatic IPT could develop secondary to portal venous infection, and an inflammatory mass may be produced in a gradually expanding fashion together with obliterating phlebitis.[8,9] The latter might explain the etiology according to the histological findings in our patient, although he was asymptomatic and the IPT was incidentally discovered by abdominal CT and MRI during follow-up after radical resection of rectal cancer. The pathogenesis of IPT is still unclear because microorganisms are not always detected in resected specimens or blood cultures.[3,4,8] Bacterial culture of the resected specimen was not performed in this case, but no bacterial infiltration was found in the resected specimens histologically.

There are no specific radiologic findings in hepatic IPT.[9–11] Most of the reported cases were detected as solitary or multiple nodular lesions in the liver on imaging studies. MRI did not show an obvious mass in our patient, but rather a lesion extending along Glisson's sheath in the lateral segment of the liver, indicative of a periductal-infiltrating type of ICC.[5] Therefore, it was impossible to make a diagnosis of hepatic IPT based on the radiographic appearance alone. Percutaneous needle biopsy of a suspicious lesion may be useful for making an accurate diagnosis of IPT,[12] but this procedure carries the risk of needle-track and peritoneal seeding. Based on the results of a multicenter questionnaire, Smith[13] estimated that the incidence of seeding after fine-needle biopsy was about 0.006%. Considering the possibility of tumor cell seeding, this diagnostic procedure was not performed in our patient whose serum CA19-9 level was slightly elevated.

Regarding the serum levels of tumor markers, serum α-fetoprotein (AFP) levels are within the normal range in patients with hepatic IPT,[14] although this tumor

marker is elevated in many patients with hepatocellular carcinoma (HCC). Thus, serum AFP levels would be helpful in differentiating between hepatic IPT and HCC. Our patient had a normal serum AFP level and a slightly elevated serum CA19-9 level. Of the 27 patients with hepatic IPT reported in Japan, 5 had slightly elevated serum CA19-9 levels.[15] Since an increase in serum CA19-9 levels is occasionally observed in patients with benign diseases of the hepatobiliary tract,[15] it is impossible to differentiate hepatic IPT from other tumors, including ICC.

Spontaneous regression occurs in some cases of hepatic IPT. Okamoto et al.[16] reported that IPTs disappeared in 14 of 62 (22.6%) patients after conservative therapy. Therefore, most patients are treated with simple observation or conservative therapy if an accurate diagnosis is made by diagnostic imaging or biopsy. There is one case report of IPT involving the hepatic hilum, which was treated with percutaneous transhepatic biliary drainage for 2 years and 9 months to manage obstructive jaundice.[17] Conservative therapy, consisting of steroid and anti-inflammatory drugs, was ineffective in this patient, and major hepatectomy was subsequently performed to remove the hepatic IPT. Thus, IPT involving the hepatic hilum mimics hilar cholangiocarcinoma or ICC, and treatment for obstructive jaundice resulting from compression of the biliary tract is required. IPT has a potential for recurrence and persistent local growth.[2] Kaneko and associates[17] reviewed 14 cases of IPT involving the hepatic hilum and indicated that complete surgical resection is desirable to prevent recurrence. The hepatic IPT in our patient, who did not have obstructive jaundice, presented as an infiltrative growth along Glisson's sheath, causing the portal vein occlusion. We report this case to point out the difficulties in differentiating between hepatic IPT extending along Glisson's sheath and ICC based on the findings of medical imaging scans. Even if the diagnosis of IPT is made by biopsy, success is not always achieved by conservative therapy for hepatic IPT with infiltrative growth along Glisson's sheath, as reported by Kaneko et al.[17] Considering the difficulties in the differential diagnosis and the tumor characteristics of IPT mimicking ICC, an immediate surgical strategy would minimize the impact on the liver volume that might be sacrificed in the treatment of this type of IPT.

References

1. Bahadori M, Liebow A. Plasma cell granuloma of the lung. Cancer 1973;31:191–208.
2. Coffin CM, Watterson J, Priest JR, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor). A clinicopathologic and immunohistochemical study of 84 cases. Am J Surg Pathol 1995;19:859–72.
3. Chen KT. Inflammatory pseudotumor of the liver. Hum Pathol 1984;15:694–6.
4. Pack GT, Baker HW. Total right hepatic lobectomy. Report of a case. Ann Surg 1953;138:253–8.
5. Liver Cancer Study Group of Japan Classification of primary liver cancer. 1st English ed. Tokyo: Kanehara; 1997. p. 6–7.
6. Gluszek S, Kot M, Czerwaty M. Inflammatory pseudotumor of the liver treated surgically. Hepatogastroenterology 1999;46: 2959–60.
7. Torzilli G, Inoue K, Midorikawa Y, Hui AM, Takayama T, Makuuchi M. Inflammatory pseudotumors of the liver: prevalence and clinical impact in surgical patients. Hepatogastroenterology 2001;48:1118–23.
8. Yoon KH, Ha HK, Lee JS, Suh JH, Kim MH, Kim PN, et al. Inflammatory pseudotumor of the liver in patients with recurrent pyogenic cholangitis: CT-histopathologic correlation. Radiology 1999;211:373–9.
9. Someren A. Inflammatory pseudotumor of liver with occlusive phlebitis: report of a case in a child and review of the literature. Am J Clin Pathol 1978;69:176–81.
10. Heneghan MA, Kaplan CG, Priebe CJ Jr, Partin JS. Inflammatory pseudotumor of the liver: a rare cause of obstructive jaundice and portal hypertension in a child. Pediatr Radiol 1984;14:433–5.
11. Yan FH, Zhou KR, Jiang YP, Shi WB. Inflammatory pseudotumor of the liver: 13 cases of MRI findings. World J Gastroenterol 2001;7:422–4.
12. Zamir D, Jarchowsky J, Singer C, Abumoch S, Groisman G, Ammar M, et al. Inflammatory pseudotumor of the liver – a rare entity and a diagnostic challenge. Am J Gastroenterol 1998;93: 1538–40.
13. Smith EH. Complications of percutaneous abdominal fine needle biopsy. Radiology 1991;178:253–8.
14. White JE, Chase CW, Kelley JE, Brock WB, Clark MO. Inflammatory pseudotumor of the liver associated with extrahepatic infection. South Med J 1997;90:23–9.
15. Ogawa T, Yokoi H, Kawarada Y. A case of onflammatory pseudotumor of the liver causing elevated serum CA19-9 levels. Am J Gastroenterol 1998;93:2551–5.
16. Okamoto K, Yoshimi F, Shida D, Oka D, Hagihira T, Shioyama Y, et al. A case of inflammatory pseudotumor of the liver which presented difficulty in differential diagnosis of cholangiocellular carcinoma (in Japanese with English abstract). Nihon Syokakigeka Gakkaizasshi (Jpn J Gastroenterol Surg) 2000;33:1900–4.
17. Kaneko K, Ando H, Watanabe Y, Seo T, Nagino M, Kamiya J, et al. Aggressive preoperative management and extended surgery for Inflammatory pseudotumor involving the hepatic hilum in a child. Surgery 2001;129:757–60.