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Case Report

Hepatic Inflammatory Pseudotumor With Elevated Serum CA19-9 Level Mimicking Liver Metastasis From Rectal Cancer: Report of a Case

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Inflammatory pseudotumor (IPT) of the liver is rare, and differential diagnosis from malignant tumors is difficult. We herein report a case of IPT of the liver with marked elevation of carbohydrate antigen 19-9 (CA19-9). A 72-year-old woman, who underwent low anterior resection for rectal cancer, had low-grade fever and epigastric pain. The patient has well-controlled idiopathic thrombocytopenic purpura (ITP) with steroid maintenance treatment. Abnormal laboratory findings included serum carcinoembryonic antigen of 15.9 ng/mL and CA19-9 up to 14,632 U/mL. Computed tomography revealed a low-density lesion, and magnetic resonance imaging demonstrated a high intensity lesion on T1, T2-weighted, and diffusion-weighted images (55 × 22 mm in the Couinaud's segment 4 of the liver). However, 1 month after the appearance of the symptom, serum level of CA19-9 suddenly decreased to 37 U/mL. The tumor decreased spontaneously to 20 × 10 mm. Seven months later, the tumor had reduced to 18 × 11 mm. With a diagnosis of IPT, the patient remains well without regrowth of the tumor at 1 year follow-up after CA19-9 decrease to normal limits.

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Inflammatory pseudotumor (IPT) of the liver is a rare condition characterized by proliferating fibrovascular tissue with infiltration of inflammatory cells.[1] Despite recent improvements in imaging modalities, differential diagnosis of IPT of the liver and malignant liver tumors is still difficult. We herein report such a case of IPT, which is accompanied by remarkably elevated serum levels of carbohydrate antigen 19-9 (CA19-9).

Case Report

A 72-year-old woman, who underwent low anterior resection for rectal cancer (moderately differentiated adenocarcinoma, stage I, T2 M0 N0) in 2006, visited our hospital for low-grade fever and epigastric pain. The patient has well-controlled idiopathic thrombocytopenic purpura (ITP) with oral intake of prednisolone 3 mg/d. On physical examination, the abdomen was unremarkable except for a postoperative scar, and neither tumor nor gallbladder was palpable. Abnormal laboratory findings included a white blood cell count (WBC) of 15,100/μL, C-reactive protein (CRP) of 1.14 mg/dL, serum carcinoembryonic antigen (CEA) of 15.9 ng/mL, and serum CA19-9 of 14,632 (normal, 37 U/mL) U/mL. Abdominal nonenhanced computed tomography (CT) revealed a low-density lesion in the Couinaud's segment 4 of the liver contacting the gallbladder (55 × 22 mm) and cholecystolithiasis (Fig. 1A). Enhanced CT revealed a hypovascular tumor with ring-enhancement (Fig. 1B). Magnetic resonance imaging (MRI) showed a tumor with high intensity on T1- and T2-weighted images. With diffusion-weighted imaging, the intensity was too high with a low B (50 s/mm^2) and a high B value (800 s/mm^2) (Fig. 2). Ultrasonography revealed a hypoechoic tumor, in contact with the gallbladder, having no obvious border with the surrounding hepatic parenchyma (Fig. 3). The blood flow to the tumor was not detected by Doppler scanning. Endoscopic retrograde cholangiography revealed a normal intrahepatic bile duct and a partial defect of the cystic duct as a result of a biliary calculus. The

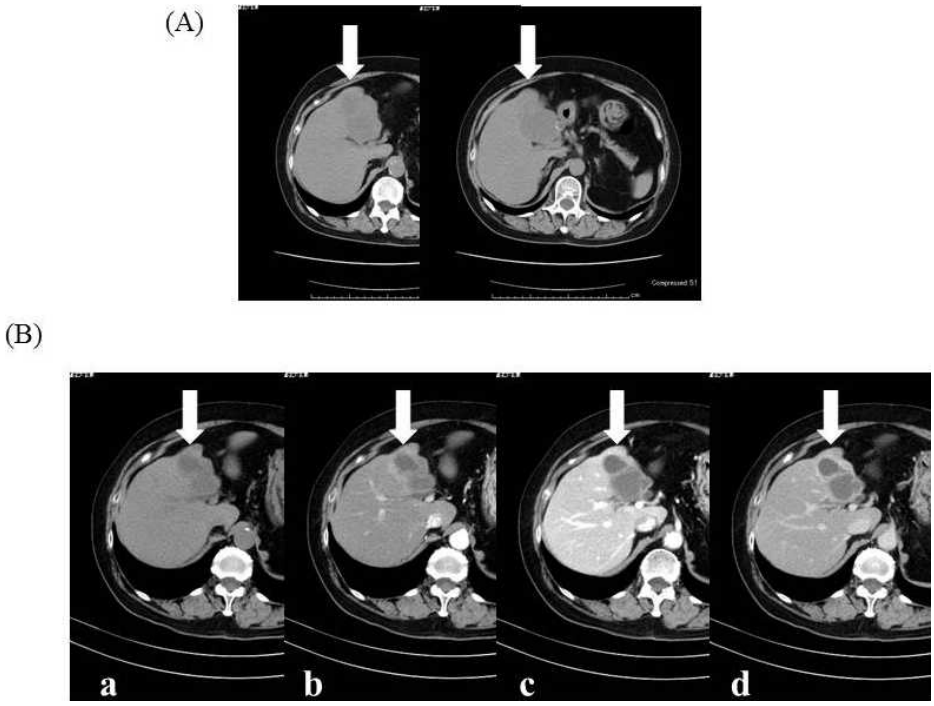

Fig. 1 **(A)** Plain CT revealed a low-density lesion in the Couinaud's segment 4 of the liver (A-a) and a gallstone (A-b). (B) Enhanced CT revealed ring-enhancement and interval hypovascularity of the tumor (B-a: plain, B-b: arterial phase, B-c: portal phase, B-d: venous phase).

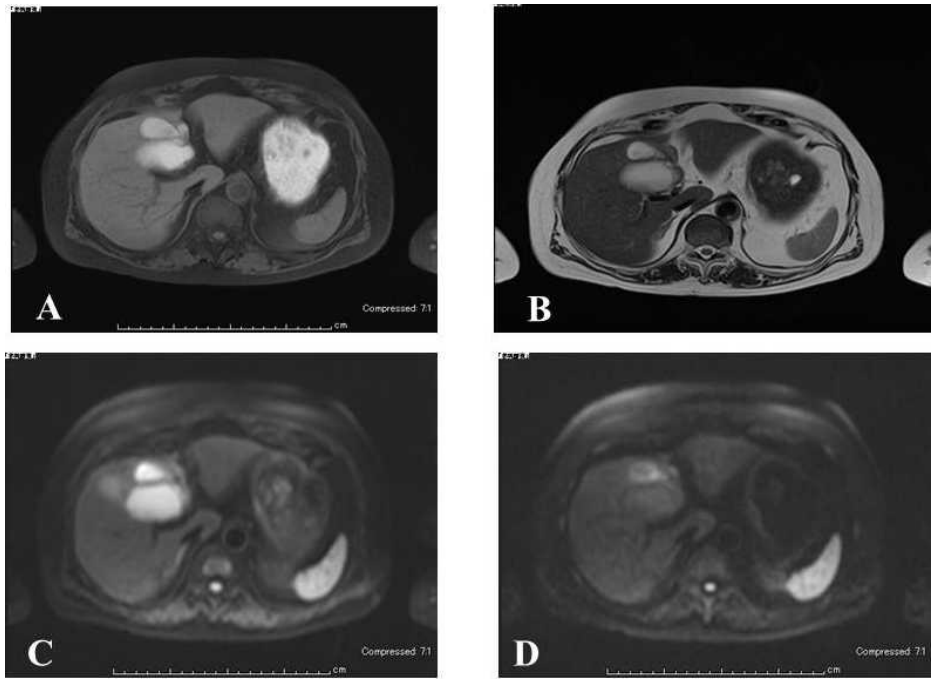

Fig. 2 MRI revealed the tumor to be high intensity on T1-weighted images (A), T2-weighted images (B), and diffusion magnified-weighted images with a low B (C) and a high B value (D).

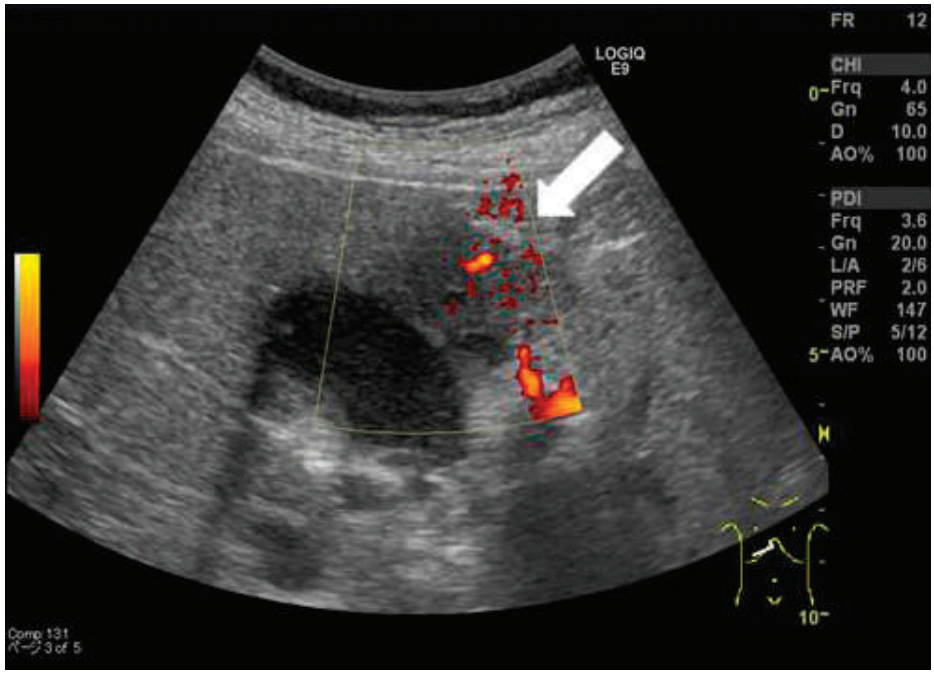

Fig. 3 Ultrasonography revealed a hypoechoic tumor continuity with the neck of the gallbladder.

Fig. 4 FDG-PET revealed several hot nodules in the Couinaud's segment 4 of the liver.

serum CA19-9 level of bile was very high (33,466 U/mL). ^{18}F-fluorodeoxyglucose-positron emission tomography (FDG-PET) revealed several hot nodules in the Couinaud's segment 4 of the liver (Fig. 4). With a diagnosis of liver metastasis from rectal cancer, we planned partial hepatic resection with cholecystectomy.

However, at 1 month after CA19-9 elevation, the serum level of CA19-9 decreased to 37 U/mL without any treatment (Fig. 5). The tumor of the liver regressed spontaneously (20 × 10 mm) and was not enhanced in all phases on gadoxetic acid–enhanced MRI. We decided that the patient should be frequently followed up without surgery. Seven months after CA19-9 elevation, the tumor size had decreased to 18 × 11 mm on MRI (Fig. 6). With a diagnosis of IPT, the patient has been followed up for 1 year after CA19-9 decrease to normal limits and remains well without regrowth of the tumor.

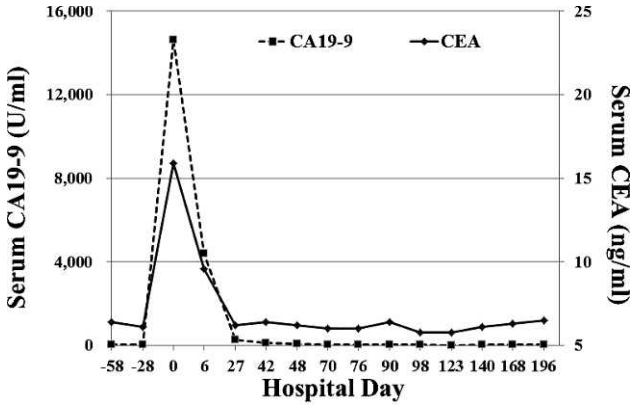

Fig. 5 Changes in serum CA19-9 and CEA.

Discussion

IPT of the liver was first reported by Pack and Backer[2] in 1953, and the liver is now recognized as one of the most frequent sites of IPT. Since then, IPT has been known as a benign lesion characterized by localized proliferation of inflammatory cells within a fibrous supporting stroma of unknown etiology,[1,3] and its spontaneous regression has been reported.[4,5] Therefore, differential diagnosis of IPT from a malignant tumor is very important. However, such a diagnosis can be difficult because of the absence of specific radiologic signs and variable radiologic findings reflecting the evolutionary stage of IPT.[6] Kitajima *et al*[7] have reported that high intensity by diffusion magnified-weighted imaging with low B value is one of the most decisive radiologic features for the diagnosis of IPT by reflecting high perfusion of water. Percutaneous needle biopsy of a suspicious lesion might be useful for making an accurate diagnosis of IPT.[8] However, this procedure is associated with a risk of needle-track and peritoneal seeding. Smith[9] reported that the incidence of seeding cells after fine-needle biopsy is about 0.006%. In addition, this patient has ITP, which requires steroid treatment. Therefore, we chose frequent follow-up by radiologic examinations.

The CA19-9, discovered by Koprowski H *et al*,[10] is a digestive cancer–related antigen and is produced in normal pancreatic and biliary ductal cells.[11] Extremely elevated CA19-9 of more than 1000 U/mL usually indicates a digestive cancer.[12] Nevertheless, false-positive results owing to benign disease such as pancreatitis[13] or acute cholangitis have been reported.[14] In the current case, we

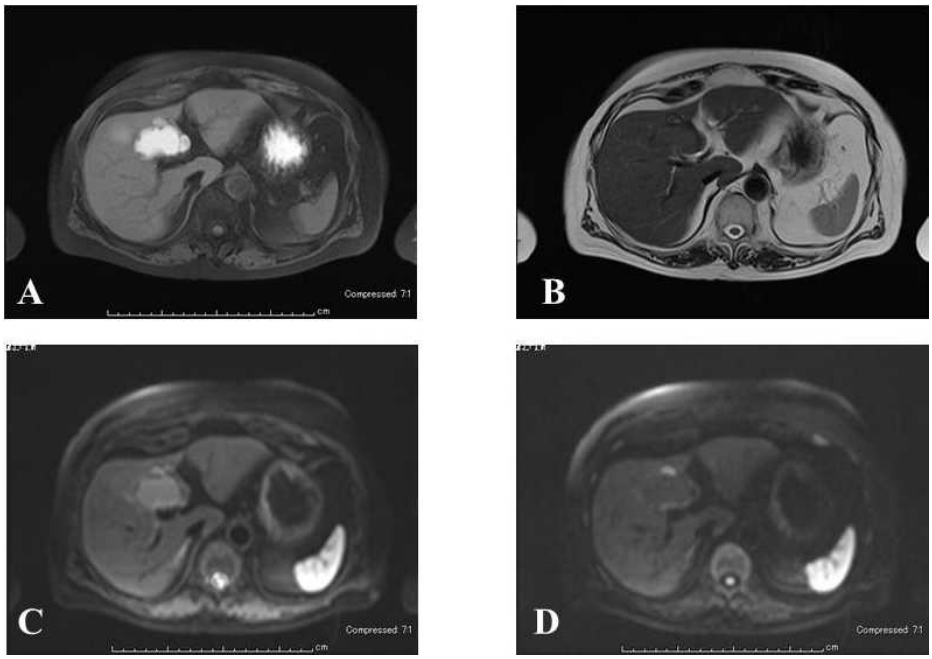

Fig. 6 MRI after 7 months. (A) T1-weighted images. (B) T2-weighted images. (C) Diffusion magnified-weighted images with a low B value. (D) Diffusion magnified-weighted images with a high B value.

diagnosed the liver mass as IPT because of spontaneous regression of the tumor size and reduction in serum CA19-9. The reasons for extreme elevation of the serum CA19-9 elevation in the current case are unclear. Possible reasons are that acute cholangitis with gallbladder stone lead to both serum CA19-9 elevation and IPT of the liver. Yoon *et al* reported that IPT of the liver might result from cholangitis because of degeneration and necrosis of the bile duct wall with subsequent periductal abscess caused by cholangitis and calculi-associated bile stasis.[15] Murohisa *et al*[14] and Sheen-Chen *et al*[16] have reported cases of common bile duct stone with cholangitis and elevated serum CA19-9 levels (60,000 U/mL and 5,673.8 U/mL, respectively). In both cases, the serum CA19-9 level decreased after starting treatments for acute cholangitis and returned to the normal level within 2 months, which is similar to the current case.

The effect of steroid treatment for IPT is controversial. Koea *et al*[17] and Maze *et al*[18] have reported the remarkable reduction of IPT of the liver and improvement in systemic symptoms, such as fever and pain, with steroid treatment. In contrast, Hertzer *et al*[19] reported an IPT case without treatment effect of steroid. Our patient received steroid treatment for ITP, which suggests that steroid treatment may have reduced the size of IPT in the present case.

In this case, we could diagnose the lesion as IPT of the liver by radiologic features and clinical progress, including spontaneous reduction in the tumor size and serum CA19-9.

Despite recent improvements in clinical and imaging modalities, differential diagnosis of IPT with malignant liver tumors is still difficult. Therefore, careful diagnosis is important for IPT.

Acknowledgments

The authors have no conflict of interest.

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