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CASE REPORT

A case of hepatic inflammatory pseudotumor protruding from the liver surface

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Abstract We report a case of a resected hepatic inflammatory pseudotumor (IPT) protruding from the liver surface. A 69-year-old male with diabetes mellitus was admitted to hospital for investigation of an hepatic mass. An irregularly shaped, low-echoic mass measuring 21 × 18 mm was identified by ultrasound in S6. On computed tomography, the tumor appeared to be growing extrahepatically. After contrast enhancement, the lesion showed persistent peripheral enhancement, while the central part was hypoenhanced. On T2-weighted magnetic resonance imaging (MRI), the central portion of the lesion was hyperintense compared with the periphery. EOB-enhanced MRI revealed the mass to be being hypointense in contrast to the surrounding liver parenchyma in the hepatobiliary phase. On diffusion-weighted images, the lesion was hyperintense. Percutaneous biopsy was not attempted to avoid tumor cell dissemination. The patient underwent partial hepatectomy because of suspected malignancy. Histopathological examination of the resected specimen revealed fibrotic tissue and abundant vessels in the periphery, while a massive infiltration of inflammatory cells and fewer vessels were observed in the center. The patient was finally diagnosed with hepatic IPT of the fibrohistiocytic type.

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Introduction

Inflammatory pseudotumor (IPT) of the liver is a relatively rare disease entity accounting for approximately 8 % of extrapulmonary IPT lesions [1]. Preoperative diagnosis is difficult because of the potential variety in imaging findings, and surgical resection has been performed in many cases. We present a case of resected hepatic IPT mimicking an extrahepatic growth pattern.

Case report

A 69-year-old male who had been regularly followed up at a local clinic for diabetes mellitus, hypertension and dyslipidemia presented with fever, and consulted his physician. Although his symptoms had been improving for several days, the laboratory data showed abnormalities, including elevated liver enzymes. He was referred to our institution, and abdominal ultrasonography (US) revealed an irregularly shaped, low-echoic mass measuring 21 × 18 mm in the hepatic segment VI (S6) (Fig. 1). He was therefore admitted for further evaluation of the lesion.

On admission, the patient was non-icteric and afebrile. He was obese (body mass index 31.6), and physical examination showed slight tenderness at the right upper quadrant of the abdomen, where a mass was palpable in the absence of lymphadenopathy.

Laboratory tests revealed slightly elevated levels of aspartate aminotransferase [AST 51 IU/L (normal, 4-43 IU/L)], and C-reactive protein [CRP, 0.67 mg/dL

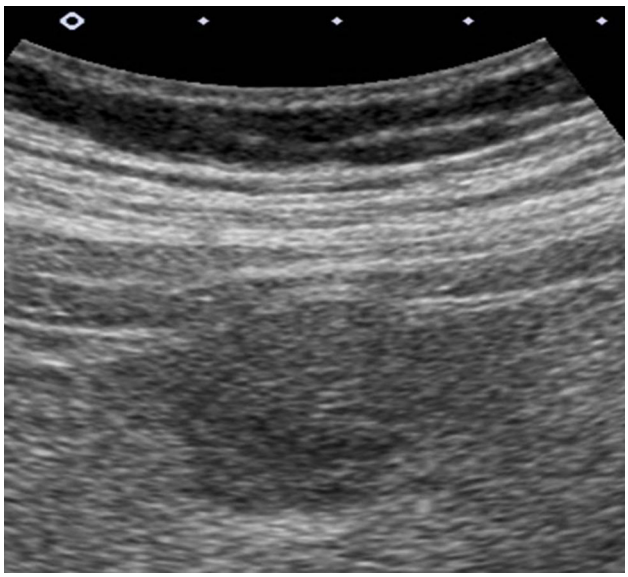

Fig. 1 Ultrasound imaging showing an irregularly shaped, low-echoic mass measuring 21 × 18 mm in S6

(normal, 0-0.3 mg/dL)]. Diabetes control was poor, with a hemoglobin A1c level of 8.3 % (normal, 4.5-5.8). With regard to tumor markers, levels of carcinoembryonic antigen (CEA), alpha-fetoprotein (AFP) and protein induced by vitamin K absence II (PIVKA-II) were within normal ranges. The level of carbohydrate antigen 19-9 (CA19-9) was slightly elevated (48 ng/mL; normal, <37 ng/mL).

A plain computed tomography (CT) scan revealed a slightly low-density mass partially protruding from the liver surface. The lesion was subsequently found to be peripherally enhanced from the early phase to the delayed phase (dynamic CT), while the central portion was hypoenhanced (Fig. 2a–c).

Magnetic resonance imaging (MRI) revealed a homogeneously hypointense lesion on T1-weighted imaging, while the central portion of the lesion was hyperintense compared with the periphery on T2-weighted imaging (Fig. 3a). After contrast enhancement with ethoxybenzyl (EOB), the lesion showed persistent peripheral enhancement, while the central part was hypoenhanced (Fig. 3b). In the hepatobiliary phase, the mass was hypointense in contrast to the surrounding liver parenchyma. Diffusion-weighted imaging revealed the lesion to be hyperintense (Fig. 3c). Neither esophagogastroduodenoscopy (EGD) nor colonoscopy revealed any malignant tumor that could be the primary lesion.

We considered peripheral cholangiocellular carcinoma (CCC) or atypical hepatocellular carcinoma (HCC) as the preoperative diagnosis. Although the results of these investigation could not exclude the possibility of a benign lesion rather than malignancy, percutaneous liver biopsy

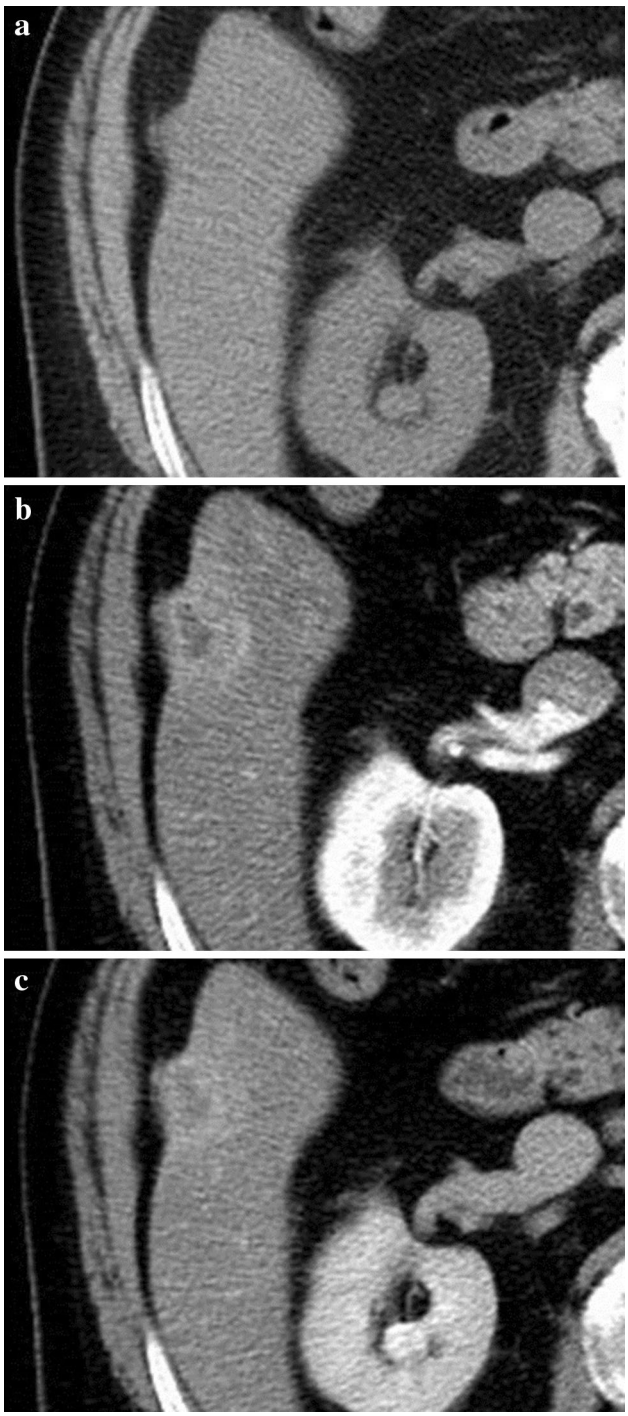

Fig. 2 Abdominal CT images. **a** Precontrast image showing a slightly low-density mass which slightly protrudes from the liver surface. **b** The lesion was peripherally enhanced in the arterial phase, while the central portion was hypoenhanced. **c** Peripheral enhancement persisted until the delayed phase

was thought to entail a risk of tumor cell dissemination because of the protrusion from the liver surface. Eventually, partial hepatectomy was performed 27 days after US examination under detailed informed consent. The findings

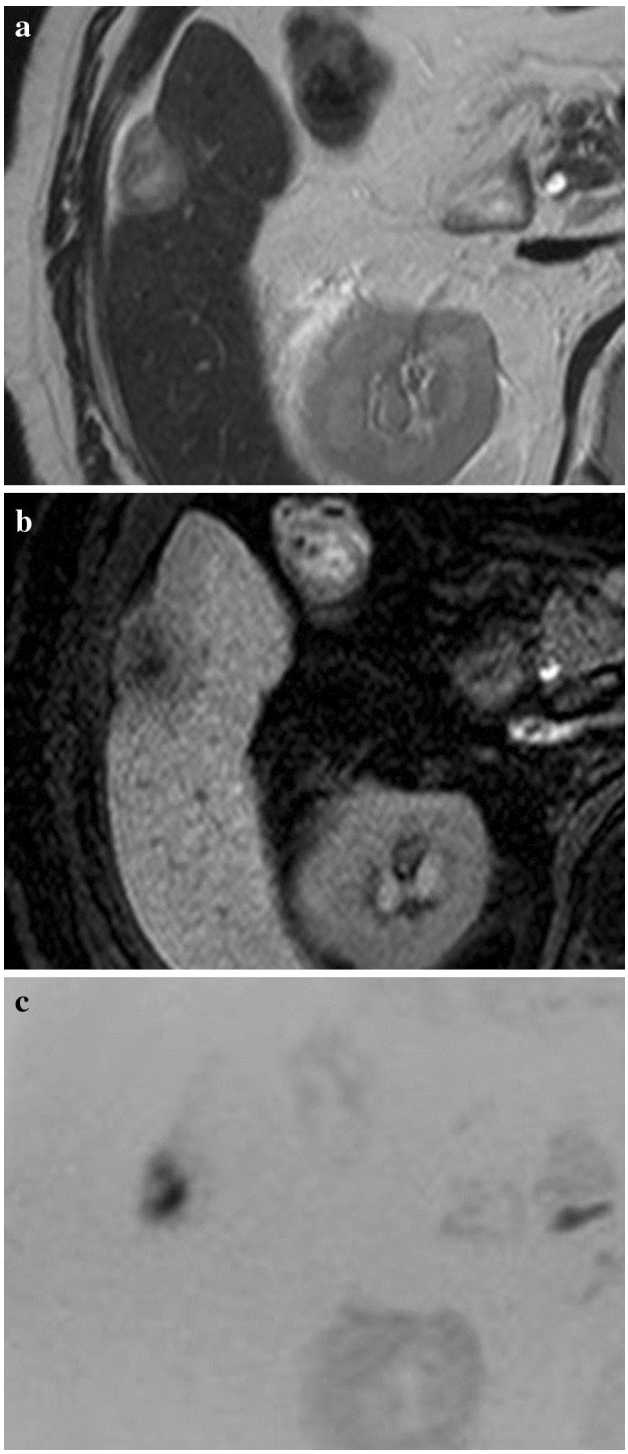

Fig. 3 MRI images. **a** On T2-weighted images, the center of the mass was hyperintense compared with the periphery. **b** On EOB-enhanced images, the lesion was hypointense in contrast to the surrounding liver parenchyma in the hepatobiliary phase. **c** Diffusion-weighted images revealed the lesion to be hyperintense

of intraoperative US were the same as those of preoperative examination. The operative time was 246 min and blood loss was 138 mL.

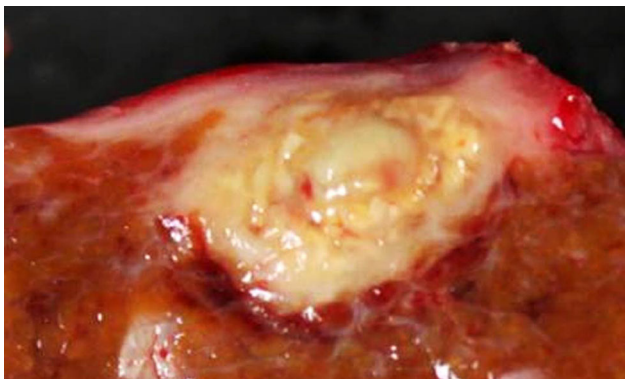

Fig. 4 Resected specimen. The lesion was observed as a whitish-yellow mass 2 cm in size

With respect to the resected specimen, the lesion macroscopically constituted a whitish-yellow mass 2 cm in size (Fig. 4). Microscopically, the protruding edge of the mass was mainly composed of fibrotic tissue along with inflammatory cell infiltrates, while abundant inflammatory cells and cholesterol cleft granuloma with focal abscess were observed in the central compartment (Fig. 5a–c). Hepatocytes were entrapped focally in the central portion of the lesion. Microvessels were more abundant in the periphery than in the center. Malignant cells were not detected. Immunohistochemical staining with anti-IgG4 antibody was negative, and obliterative phlebitis was not observed. Background liver tissue showed steatosis with bridging fibrosis. The final histological diagnosis was hepatic IPT of the fibrohistiocytic type. The postoperative course was uneventful. After discharge, the patient has been well without any sign of recurrence.

Discussion

The most commonly reported site of IPT is the lung; however IPT may develop in other organs, including the liver. Hepatic IPT was first described by Pack and Baker in 1953 [2]. According to available data, the male-to-female ratio ranges between 1:1 and 3.5:1, with a male predominance [3]. Patients present with abdominal pain (36–53 %), fever (24–41 %), malaise (11–20 %) and/or weight loss (3–9 %), but sometimes remain asymptomatic (16–20 %) [4, 5]. Inflammatory reactions, abnormal liver function tests, or sometimes elevated serum CA19-9 levels may be observed [6, 7]. Although several mechanisms have been proposed, such as infection, trauma, vascular diseases, and autoimmune disorders [3], the precise etiology and pathogenesis of IPT of the liver has not been completely elucidated. Hepatic IPT might be a multifactorial disease. Lesions may be single or multiple and sometimes resolve

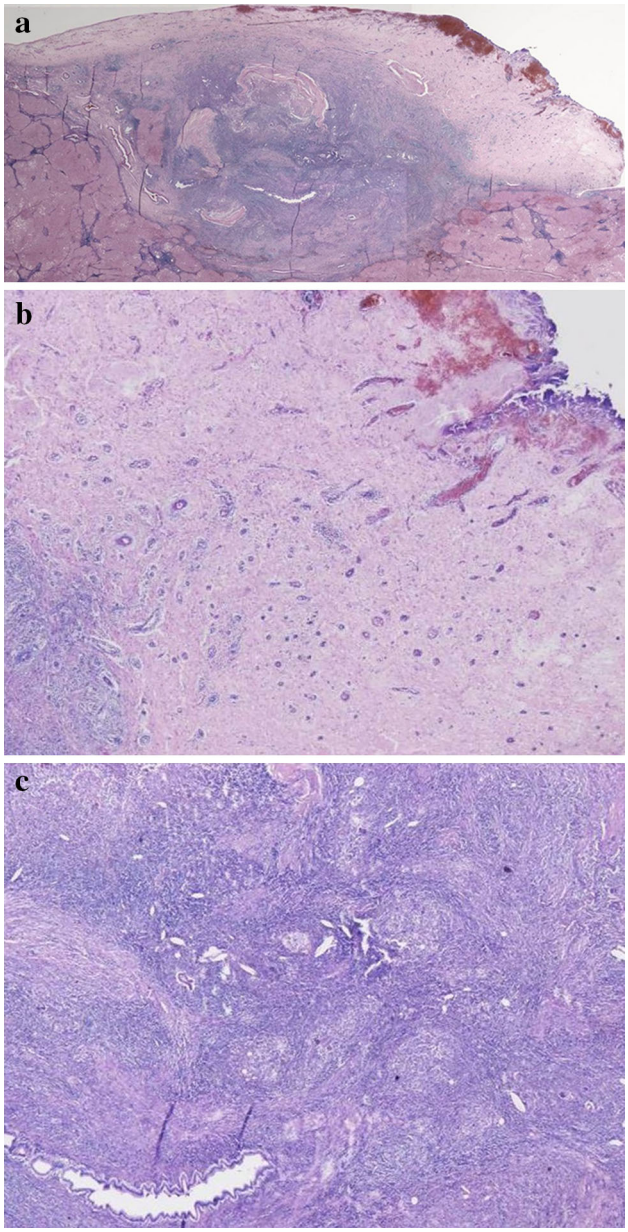

Fig. 5 Pathological findings. **a** The histological features of the center were different from the periphery [hematoxylin–eosin staining (HE), ×1]. **b** The protruding periphery was mainly composed of fibrotic tissue (HE, ×4). **c** Abundant inflammatory cells and cholesterol cleft granuloma were observed in the central portion (HE, ×4)

spontaneously [3, 4, 8, 9]. Preoperative diagnosis is difficult because specific serological markers or imaging findings are lacking. Furthermore, imaging findings may change over time, probably because of the inflammatory processes [10]. Surgical resection has been performed in many cases, but percutaneous needle biopsy could provide a clue to avoid unnecessary operation, if indicated [11]. In this particular case, we did not perform needle biopsy because the edge of the mass appeared to protrude from the liver surface. Any significant reduction in the size of the lesion was not noted during the clinical course.

Considering imaging studies, previous reports have pointed out several characteristics. In a study of 13 cases of hepatic IPT, Yoon et al. [6] reported that contrast-enhanced CT showed central hypoattenuating areas with an iso- or hyperattenuating thickened periphery in four cases and a multiseptate appearance with hyperattenuating internal septa and periphery in the remaining nine cases. The central hypoattenuating area histologically indicated the presence of chronic inflammatory infiltrates, while iso- or hyperattenuating areas in the periphery and internal septa of the mass represented fibroblastic proliferation. Park et al. [4] reported that enhanced CT indicated poorly defined peripheral enhancement (82.5 %) at the arterial phase and poorly defined hyperattenuating lesions with internal hypoattenuating areas at the equilibrium phase (77.0 %). MRI revealed poorly defined peripheral rim-like enhancement (77.8 %). The enhancement pattern of the present case was consistent with previous reports. Histologically, the peripheral portion with rim-like enhancement corresponded to fibrous tissue, whereas the poorly-enhanced central part contained inflammatory cells and less vessels. The difference in histology and vascularity between these compartments is considered to explain the disparity of enhancement patterns.

As for hepatic malignancies which should be differentiated from IPT, early enhancement with delayed washout is the typical finding in cases of HCC. However, Lee et al. [12] reported that the typical enhancement pattern was detected only in 56.4 % of surgically resected HCCs. Protrusion from the liver surface is occasionally observed in HCCs, which may lead to spontaneous tumor rupture [13]. In contrast, peripheral CCC typically shows delayed enhancement on CT and MRI [14, 15]. Likewise, it should be noted that the enhancement pattern varies according to the histological subtype and the degree and distribution of fibrosis within the tumor [16]. In cases of peripheral CCC, retraction of the liver capsule would be more frequently observed than protrusion [17], although this finding has also been reported in benign lesions including IPTs [18, 19]. In the present case, the protruded portion was mainly composed of fibrous tissue. The usefulness of diffusion-weighted MRI has not been examined in a large case series. These facts indicate that imaging studies cannot completely preclude the possibility of HCC or CCC. From a retrospective point of view, the enhancement pattern and absence of hepatic capsular retraction in the present case might have supported a preoperative diagnosis of IPT rather than HCC or CCC.

Pathologically, Zen et al. [20] classified IPT into two categories. The fibrohistiocytic type was characterized by xanthogranulomatous inflammation, multinucleated giant

cells, and neutrophilic infiltration. The lymphoplasmacytic type showed lymphoplasmacytic infiltration with IgG4-positive plasma cells and might be associated with IgG4-related diseases. The present case was compatible with the fibrohistiocytic type. Whether differences in imaging characteristics or clinical behavior exist between the two subtypes needs to be elucidated by further case investigation.

In conclusion, IPT should be considered as one of the differential diagnoses when a mass lesion protruding from the liver surface is encountered.

Compliance with ethical standards

Conflict of Interest: Takashi Obana, Shuuji Yamasaki, Kazushi Nishio, and Yasushi Kobayashi declare that they have no conflict of interest.

Human/Animal Rights: All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and its later amendments.

Informed Consent: Informed consent was obtained from all patients for being included in the study.

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