

[CASE REPORT]

Intratumor Abscess in a Posttraumatic Hepatic Inflammatory Pseudotumor Spreading Out of the Liver

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Abstract:

A 45-year-old woman with abdominal pain after minor trauma was referred to our hospital. Computed tomography (CT) showed a hypovascular tumor in the left liver lobe. A tumor biopsy revealed granuloma, although no findings indicated malignancy or infection. A follow-up imaging study showed spread of the hepatic tumor. Her abdominal pain worsened after a second minor trauma. CT revealed an intratumor abscess, and pus overflowed from the patient's umbilicus. The abscess was improved by antibiotics and drainage therapy. In this case, unusual imaging findings and an atypical disease course of a hepatic inflammatory pseudotumor were observed.

Key words: inflammatory pseudotumor, abscess, trauma, liver

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Introduction

Inflammatory pseudotumor (IPT) was first described in 1954 as a tumor clinically and radiologically mimicking malignancy (1). IPT has been described by several names in previous reports, including inflammatory myofibrohistiocytic proliferation, and inflammatory myofibroblastic tumor. IPT is observed most commonly in the lung but can develop in several organs of the body.

Although the cause of IPT is unknown, trauma, surgical inflammation, infection, or immune-autoimmune conditions are thought to be responsible in some cases of IPT (2). Hepatic IPT is an uncommon entity and exhibits heterogeneous clinical findings. Overall, the presenting symptoms and imaging findings of hepatic IPT are nonspecific (1-3). Furthermore, the natural clinical course of hepatic IPT is incompletely understood.

We herein report a case of hepatic IPT after minor trauma spreading from the liver to the intraperitoneal tissue. After a second minor trauma, the patient developed an abscess in the tumor.

Case Report

A 45-year-old woman visited a local hospital because of upper abdominal pain. She had accidentally hit her upper abdomen on a refrigerator handle two weeks previously, and upper abdominal pain developed. Her abdominal pain spontaneously improved the day after minor trauma, but the pain recurred two weeks later. She did not have any specific medical history, nor did she have any history of smoking or excessive alcohol consumption.

Her laboratory data were as follows: white blood cell (WBC), 6,000/ μ L; aspartate transaminase (AST), 12 U/L; alanine aminotransferase (ALT), 9 U/L; alkaline phosphatase (ALP), 252 U/L; γ -glutamyl transpeptidase (GTP), 15 U/L; and C-reactive protein (CRP), 0.94 mg/dL. Contrast computed tomography (CT) revealed a hypovascular tumor in the left liver lobe (Fig. 1A, B). The tumor size was 40 \times 33 mm. Magnetic resonance imaging (MRI) showed a low intensity on T1-weighted imaging, slightly high intensity on T2-weighted imaging, and high intensity on diffusion-weighted imaging (Fig. 1C-E). A heterogeneous hypochoic tumor was indicated by ultrasonography (US) (Fig. 1F). Her

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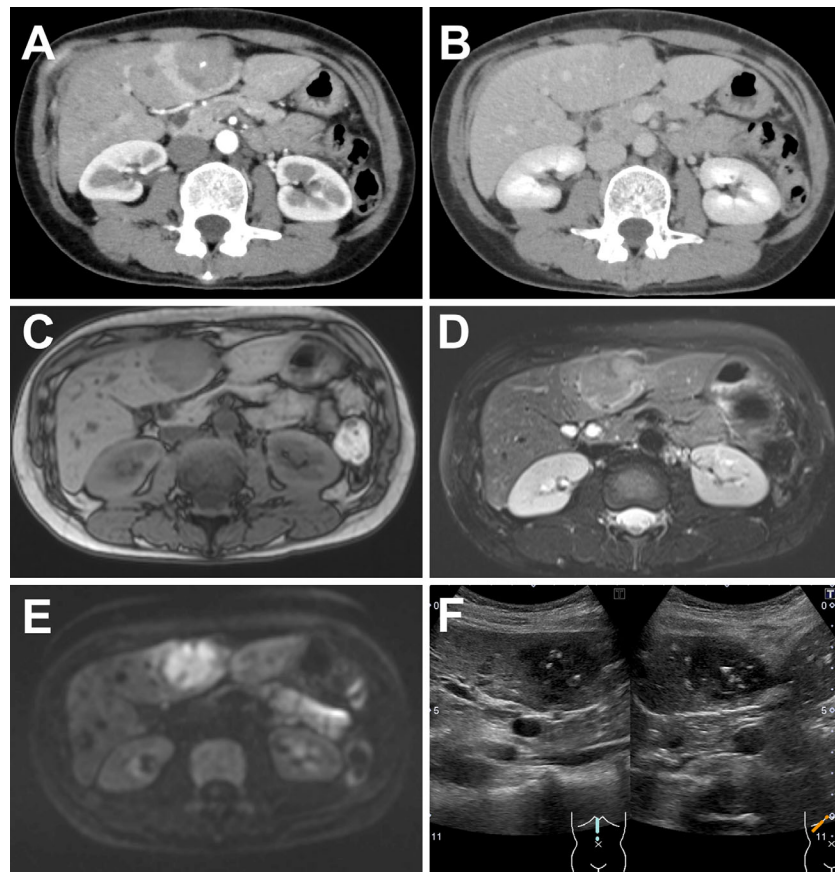


Figure 1. The imaging findings of the hepatic inflammatory pseudotumor. Arterial phase (A) and venous phase (B) imaging obtained using contrast computed tomography. T1-weighted phase (C), T2-weighted phase (D), and diffusion-weighted (E) imaging obtained using magnetic resonance imaging techniques. B-mode ultrasonographic imaging (F).

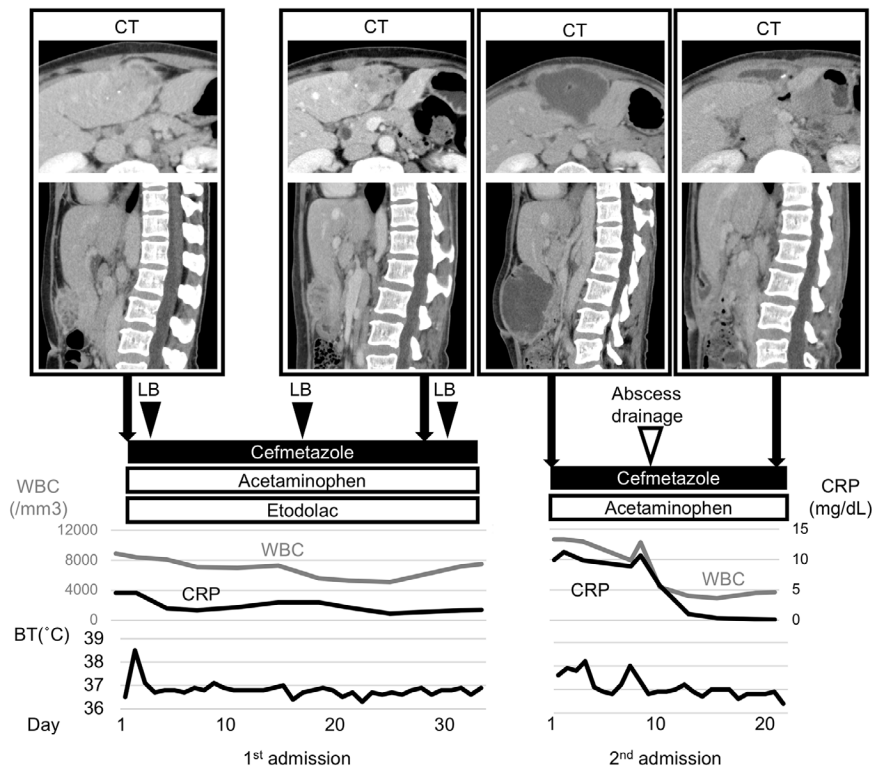


Figure 2. Clinical course of the first and second admissions. BT: body temperature, CRP: C-reactive protein, CT: computed tomography, LB: liver biopsy, WBC: white blood cell

Table. Laboratory Findings.

Complete blood count	
White blood cell count (/mm ³)	8,900
Red blood cell count (×10 ⁴ /mm ³)	341
Hemoglobin level (g/dL)	10.8
Platelet count (×10 ⁴ /mm ³)	34.7
Coagulation status	
Prothrombin activity (%)	81.5
Activated partial thromboplastin time (s)	30.6
Biochemistry	
Aspartate transaminase (U/L)	7
Alanine aminotransferase (U/L)	6
Lactate dehydrogenase (U/L)	101
Alkaline phosphatase (U/L)	265
γ-Glutamyltranspeptidase (U/L)	25
Total protein (g/dL)	6.9
Albumin (g/dL)	3.4
Total bilirubin (mg/dL)	0.5
Blood urea nitrogen (mg/dL)	3
Creatinine (mg/dL)	0.61
Ammonia (μg/mL)	27
IgG (mg/dL)	1,217
IgG4 (mg/dL)	40.5
IgM (mg/dL)	259
C-reactive protein (mg/dL)	4.54
Hepatitis B surface antigen (IU/mL)	0.00
Anti-hepatitis C antibody (S/CO)	0.1
Antinuclear antibody	<80
α-Fetoprotein (ng/mL)	4.3
Des-γ-carboxy prothrombin (mAU/mL)	19
Carcinoembryonic antigen (ng/mL)	1.0
Carbohydrate antigen 19-9 (U/mL)	7.6
Soluble interleukin-2 receptor (U/mL)	487

abdominal pain improved with acetaminophen.

Six weeks after the first minor trauma, the patient developed a mild fever (37.0°C), and her abdominal pain worsened. Contrast CT showed findings of tumor spread from the liver (Fig. 2). She was referred to our hospital because she was considered to have a risk of tumor rupture.

Her body temperature was 36.4°C, blood pressure was 109/63 mmHg, heart rate was 75/min, and SpO₂ was 98%. She presented with abdominal tenderness without rebound or guarding. The laboratory findings are provided in Table. The WBC and CRP values were elevated, whereas the levels of liver enzymes, IgG, and IgG4 were within normal ranges. Tumor markers, such as alpha-fetoprotein, des-gamma-carboxy prothrombin, carcinoembryonic antigen, carbohydrate antigen 19-9, and soluble interleukin-2 receptor, were also within normal ranges. She showed mild anemia, but there were no findings of gastrointestinal bleeding or hemolytic anemia. Contrast-enhanced US revealed an isovascular lesion at the arterial phase that was hypoechoic at the Kupffer phase.

An initial needle biopsy (21 G) was performed because we suspected that the tumor was malignant, and histology demonstrated a granuloma without malignant cells

(Fig. 3A, B). We also detected proliferation of spindle-like cells that were negative for hepatocyte paraffin-1 and p53 by immunohistochemistry (Fig. 3C, D). We considered her tumor to be an IPT and/or an actinomycosis based on the imaging findings and liver biopsy histology. A second needle biopsy (18 G) was performed, followed by tissue culture and 16S rDNA sequencing to detect bacterial infection. However, no infection in the tumor was found. The results of both a T-SPOT test for diagnosing tuberculosis and blood cultures were also negative.

Although the tumor histology did not suggest malignancy or infection, CT showed that the hepatic IPT had gradually spread from the liver (Fig. 2). A third needle biopsy (18 G) was performed to decide the next treatment after obtaining the patient's consent. Liver histology revealed a granuloma with lymphocytic and eosinophilic infiltration (Fig. 4E, F). There were no findings of infection by Gram staining, Grocott staining, or Ziehl-Neelsen staining. We performed a second round of 16S rDNA sequencing on the tumor tissue, with no infection detected. We diagnosed the patient with hepatic IPT, and IPT was observed without resection. Intravenous antibiotics (cefmetazole) were administered, and her fever and abdominal pain were controlled by nonsteroidal anti-inflammatory drugs (etodolac 400 mg/day and acetaminophen 1,000 mg/day). The tumor size decreased slightly, and she was discharged one month after admission. The patient was followed up in our hospital.

Five months after the first hospitalization, she hit her upper abdomen again on a refrigerator handle. Abdominal pain occurred and then improved without treatment. However, her abdominal pain worsened 1 week after the second minor trauma. Laboratory findings showed the elevation of WBC (11,700/mm³) and CRP (9.92 mg/dL). CT suggested an abscess in the tumor. According to CT and MRI findings, the dilated bile duct communicated with the abscess, with disruption of the bile duct between the dilated portion and that in the left lobe (Fig. 4). Intravenous antibiotics (cefmetazole) were started. Ten days after admission, her abdominal pain worsened, and pus overflowed from her umbilicus (Fig. 5). *Klebsiella pneumoniae* was detected in the pus. Abscess drainage was combined with antibiotic therapy, and she was discharged 25 days after the second admission. After the improvement of the abscess, there was leakage of serous-yellow liquid-like bile from her umbilicus, although this leakage spontaneously improved. The abscess had not relapsed at the four-month follow-up visit.

Discussion

We encountered a case of hepatic IPT after minor trauma. Our patient showed unusual radiographic findings and an atypical clinical course. CT revealed tumor spread from the liver, similar to tumor rupture, although the patient's symptoms and vital signs did not deteriorate. Furthermore, the tumor spread from the liver to the intraperitoneal tissue. After the patient developed an intratumor abscess, pus overflowed

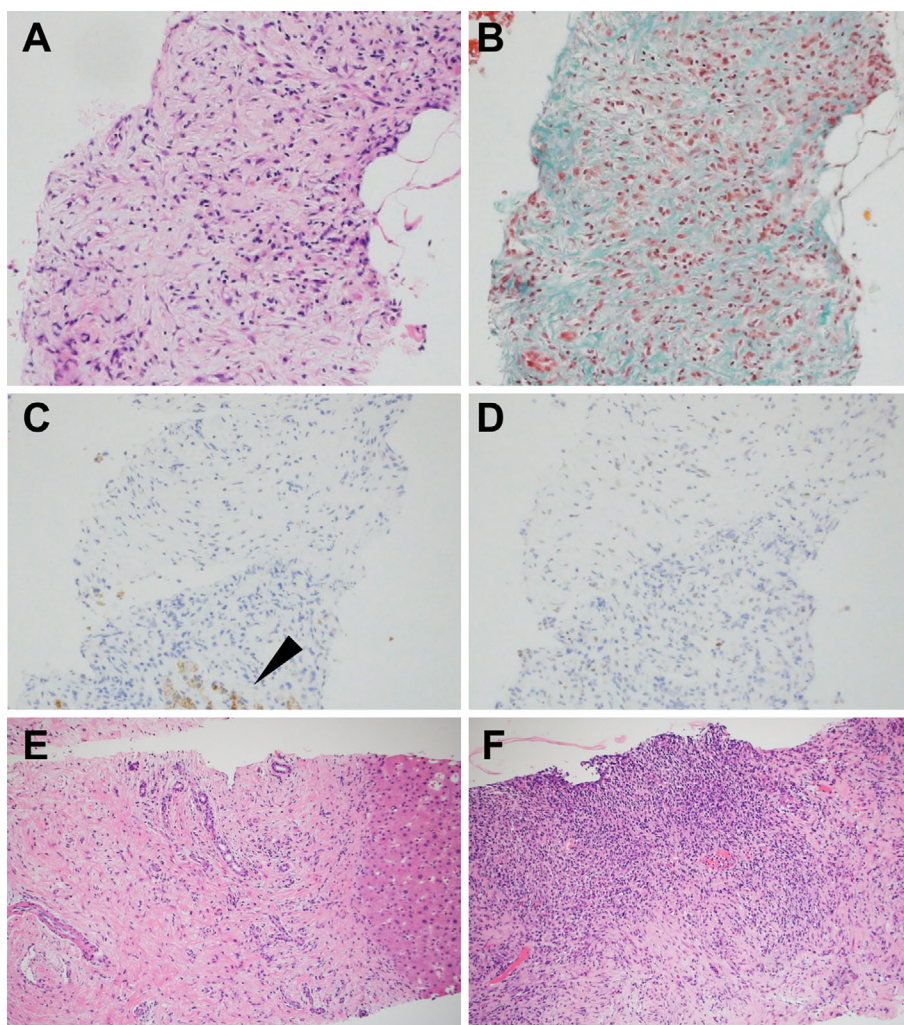


Figure 3. Histological findings of the hepatic inflammatory pseudotumor at the first biopsy (A-D) and third biopsy (E and F). (A) A granuloma showing proliferation of spindle-like cells without malignant cells [Hematoxylin and Eosin (H&E) staining, $\times 200$]. (B) Abundant collagen bundles surround spindle-like and inflammatory cells (Elastica-Masson staining, $\times 200$). (C) A few hepatocytes in the tumor (black arrowhead) and negative staining for hepatocyte paraffin-1 in spindle-like cells (hepatocyte paraffin-1 staining, $\times 200$). (D) Negative staining for p53 in spindle-like cells (p53 staining, $\times 200$). (E) Abundant collagenous tissues in the tumor without malignant cells (H&E staining, $\times 100$). (F) Lymphocytic and eosinophilic infiltration (H&E staining, $\times 100$).

from her umbilicus. These findings suggested the transfer of an inflammatory reaction via the round ligament of the liver. Indeed, abscess formation by inflammation of the round ligament of the liver has been reported (4, 5). Hepatic IPT in the left lobe developed because of an inflammatory reaction induced by minor trauma, and the inflammatory reaction was transmitted from the liver to the umbilicus via the round ligament of the liver.

At her first admission, we had suspicions regarding the association between the IPT and minor trauma. Furthermore, we did not know why the abscess developed in the IPT, as the IPT appeared to exist outside of the liver. *K. pneumonia* was detected in the pus, but the infection route was unclear. Nonetheless, when she developed an abscess, we detected communication between the abscess and the intrahepatic bile duct. Disruption of the bile duct between the dilated portion

and that in the left lobe was also suggested. This finding was made after the second minor trauma, and no event other than this second minor trauma had occurred. These findings suggested that the second minor trauma had caused the hepatic injury with bile duct disruption and laceration. We considered then that her first minor trauma might have caused hepatic injury, subsequently causing IPT, because the patient had hit her abdomen on the same refrigerator handle before the first and second admissions. Furthermore, we speculated that the abscesses in the IPT had been infected by the bile duct. A case of hepatic IPT that developed an abscess in the tumor has been reported (6), and several biliary diseases are thought to be the main cause of liver abscess (7). Fortunately, the abscess in our patient improved following treatment with antibiotics and drainage. However, treatment failure may result in peritonitis or sepsis. In gen-

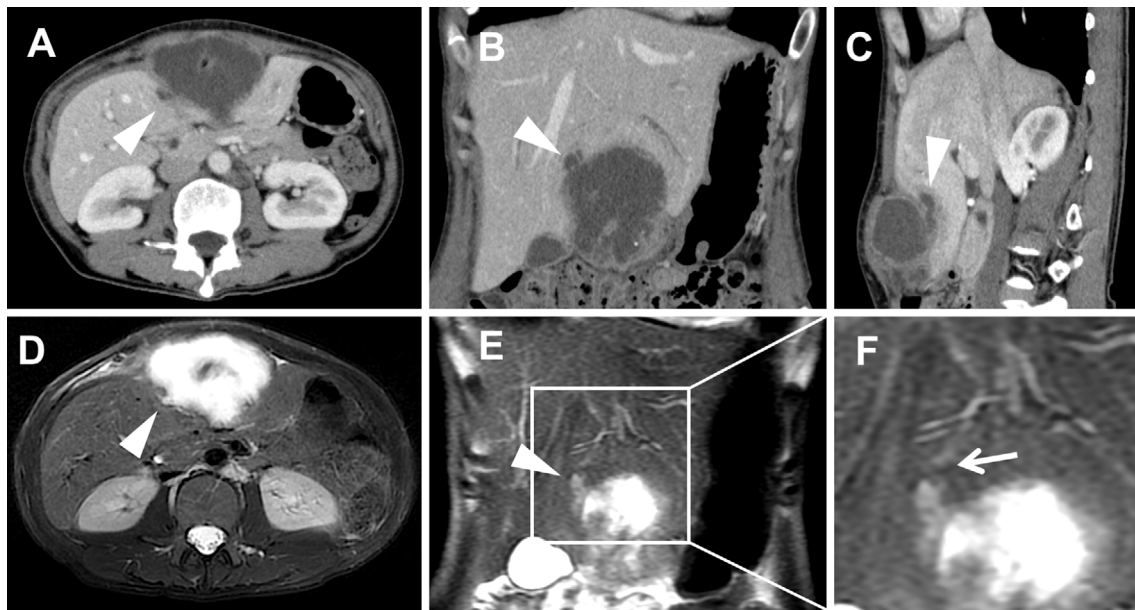


Figure 4. Imaging findings of the abscess in the hepatic inflammatory pseudotumor. Axial (A), coronal (B), and sagittal (C) slices in the portal venous phase of contrast computed tomography. Axial (D) and coronal (E, F) T2-weighted magnetic resonance imaging. The dilated bile duct communicated with the abscess (white arrowhead in A-E). Disruption of the bile duct between the dilated portion and intrahepatic portion (white arrow in F).



Figure 5. Pus overflowing from the patient's umbilicus.

eral, avoiding repeated trauma to an IPT may be important for preventing the development of complications.

As the biological characteristics of IPT vary, the appropriate treatment for IPT may differ among patients (1-3). Hepatic resection is typically performed in IPT patients who have symptoms or malignant features. If malignancy is excluded by a pathological analysis, IPT may be observed without invasive treatment. Our patient underwent two tumor needle biopsies, and there were no findings of malignancy. The size of the tumor was decreased by the administration of antibiotics and nonsteroidal anti-inflammatory drugs. Spontaneous regression of IPT has been reported (8). Furthermore, the p53 expression was low in our patient, a characteristic associated with the clinical course of inflammatory myofibroblastic tumors (9, 10). Our case suggests that hepatic IPT without malignant features can be observed with careful follow-up.

It was difficult to distinguish IPT with or without actino-

mycosis after the first biopsy because actinomycosis can exhibit local tissue invasion; IPT due to actinomycosis has also been reported (11, 12). Second and third tumor biopsies were performed with an 18-G needle because we worried that the tumor samples obtained by the 21G needle had been insufficient to detect infection or malignancy. A tumor biopsy was safely performed and useful for deciding the next treatment in our case. However, multiple biopsies may result in the dissemination of malignant cells or bleeding (13, 14). Thus, efforts should be made to decrease the number of biopsies in order to reduce the risk of complications.

In summary, we report a case of posttraumatic hepatic IPT that showed unusual radiographic findings and an atypical clinical course. Our case may facilitate an understanding of the clinical course of IPT and prevent the development of complications during the follow-up period.

Informed consent was obtained from the patient for inclusion in the study.

The authors state that they have no Conflict of Interest (COI).

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