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A case of multiple inflammatory hepatic pseudotumor protruding from the liver surface after colonic cancer



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ABSTRACT

INTRODUCTION: Inflammatory hepatic pseudotumor (IHPT) is an important benign liver disease because it is difficult to clinically and radiologically distinguish from malignant tumors.

PRESENTATION OF CASE: Here, we describe a case of a 67-year-old male patient diagnosed with multiple inflammatory hepatic pseudotumors. The patient had undergone left hemicolectomy for descending colonic cancer (T3 N0 M0 stage IIA) 2 years prior. He underwent segment 6 and segment 7 partial hepatectomy because of suspected liver metastasis. The patient had an unremarkable postoperative course and was discharged 7 days after surgery. Marked infiltration of inflammatory cells was observed on histological examination. The patient was finally diagnosed with IHPT of the fibrohistiocytic type.

DISCUSSION: Repeated imaging studies over 1 month showed the spontaneous regression of the hepatic tumors. Therefore, knowledge regarding this condition is necessary to allow for treatment, even in the absence of experience. During examination, it may be important to ascertain lesion size. Moreover, percutaneous needle biopsy and follow-up examinations are necessary for cases of suspected IHPT.

CONCLUSION: Hepatectomy should be considered if the lesion is suspected to be an IHPT.

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1. Introduction

An inflammatory hepatic pseudotumor (IHPT) is a rare benign lesion whose etiology is unclear. IHPT may mimic malignant tumors when found in a patient with a history of malignancy. Some cases of IHPT have been associated with malignant tumors. We, herein, report a case of resected multiple IHPT incorrectly diagnosed as metastatic liver tumors, occurring after colonic cancer. Our case report was based on the SCARE Guidelines [1].

2. Presentation of case

The patient was a 67-year-old man who had undergone left hemicolectomy for descending colonic cancer (T3 N0 M0 stage IIA) 2 years prior to presentation. He underwent adjuvant chemotherapy with oral uracil and tegafur plus leucovorin for 6 months. One year after the hemicolectomy, the patient was found to have cholelithiasis and required cholecystectomy. He was otherwise clinically well without obvious tumor (colonic cancer) recurrence.

Blood chemistry analysis determined the levels of hepatobiliary enzymes.

The laboratory findings on admission were as follows: aspartate aminotransferase 34 U/L, alanine aminotransferase 29 U/L, alkaline phosphatase ALP 392 U/L, and γ -glutamyltransferase 37 U/L. However, the carcinoembryonic antigen level was marginally elevated (7.0 ng/mL) (Table 1). Sonazoid-enhanced ultrasonography showed that the tumors were hypoechoic at the post-vascular phase and were detected at the Kupffer phase. Computed tomography (CT) revealed two irregular peripherally enhanced S6 tumors protruding from the liver surface (Fig. 1a and b). Moreover, the tumors appeared to be growing extrahepatically. Magnetic resonance imaging (MRI) could not be performed because the patient had a tattoo. Fluorodeoxyglucose positron emission tomography (PET-CT) confirmed abnormal metabolic activity in the S6 lesion, with a high standardized uptake value of 4.27 (Fig. 2). Percutaneous needle biopsy under ultrasonic guidance was not attempted owing to a risk of tumor cell dissemination.

This patient was admitted to the Division of Surgery of the Gastroenterological Center in our hospital and underwent S6 partial hepatectomy because of suspected malignancy. He had an unremarkable postoperative course and was discharged in remission from our hospital 7 days after surgery.

Abbreviations: IHPT, inflammatory hepatic pseudotumor.

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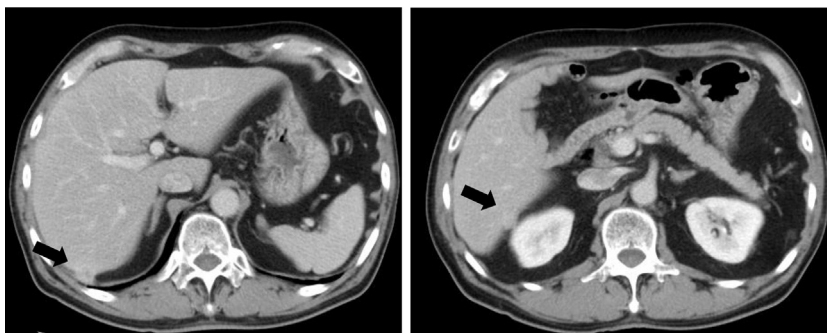


Fig. 1. a and b. Preoperative abdominal CT findings. Abdominal CT revealed two irregular peripherally enhanced S6 tumors protruding from the liver surface (black arrow). CT, computed tomography.

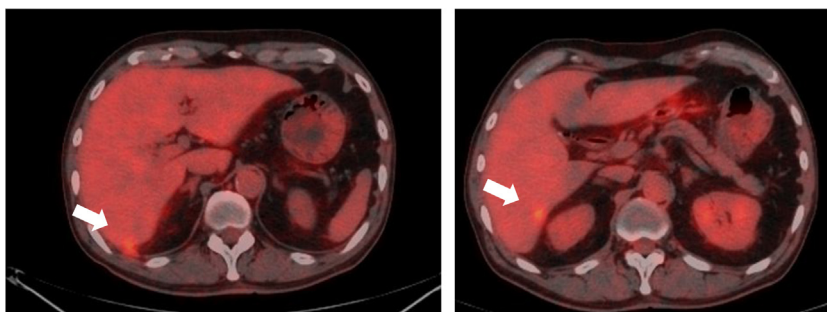


Fig. 2. Preoperative PET-CT. Fluorodeoxyglucose positron emission tomography confirmed abnormal metabolic activity in the S6 lesion, with a high standardized uptake value of 4.27 (white arrow). PET-CT, fluorodeoxyglucose positron emission tomography.

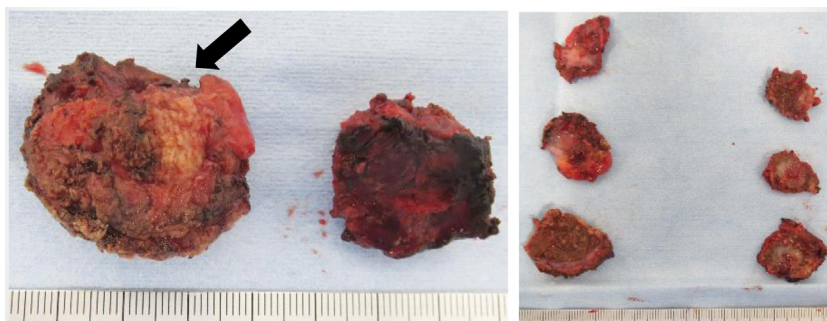


Fig. 3. Macroscopic findings of the resected specimen. The resected specimen showed that the tumor was located in the S6 area of the liver. The cut surface was yellowish and the white tumor measured 15 mm × 13 mm in diameter (black arrow).

The resected specimen showed that the tumor was located in the segment 6 area of the liver. The cut surface was yellowish and the white tumor measured 15 mm × 13 mm (Fig. 3). Histological examination of the tumor did not reveal any malignant cells. However, a remarkable infiltration of inflammatory cells was observed (Fig. 4).

The patient was finally diagnosed with IHPT of the fibrohistiocytic type. The patient was managed with adjuvant chemotherapy for 6 months, and to date, no recurrence or metastasis has been noted after 4 years.

3. Discussion

Inflammatory pseudotumors are benign lesions mostly occurring in the lungs [2] and are also referred to as inflammatory myofibroblastic tumors. They are characterized by localized fibrous proliferations with chronic inflammatory cell infiltration. Histologically, inflammatory pseudotumors are characterized by bland spindle cells and numerous inflammatory cells with a background

of collagenous, hyalinized, or myxoid stroma [3]. The liver is the second most common organ for inflammatory pseudotumors. However, the pathogenesis of IHPT remains largely unknown [4].

IHPT is also called inflammatory myofibroblastic tumor or plasma cell granuloma and was first described in 1953 by Pack and Becker [5]. Abdominal pain, intermittent fever, jaundice, and weight loss are the main symptoms observed in IHPT patients [6]. IHPT typically presents with non-specific constitutional symptoms such as fever (66%), abdominal pain (51%), and weight loss (21%) [7,8]. However, in this case, physical examination demonstrated no symptoms such as abdominal pain.

According to Kose [9] and Lopez et al. [10], IHPT is three times more common in men than in women and is frequently seen in the non-European population. Sixty-one percent of lesions are located in the right hepatic lobe, near the gallbladder, or related to the biliary tree. Yoon et al. [11] reported that IHPT might result from cholangitis because of the degeneration and necrosis of the bile duct wall with subsequent periductal abscess caused by cholangitis and calculi-associated bile stasis.

Table 1
Laboratory findings.

Variable	Range	On admission
Peripheral blood		
WBC (/ul)	3900–9800	8200
RBC (/ul)	430–570	585 × 10 ⁴
Hb (g/dl)	13.5–17.6	17.4
Hct (%)	40.0–52.0	51.2
Plt (/ul)	12.0–34.0	23.2 × 10 ⁴
Tumor markers		
CEA	0–5	7.0 ng/ml
CA19-9	0–37	4U/ml
Blood chemistry		
TP (g/dl)	6.5–8.3	8.0
ALB (g/dl)	3.8–5.2	4.5
T.Bil (mg/dl)	0.2–1.2	0.8
AST (IU/l)	10–40	34
ALT (IU/l)	5–45	29
ALP (IU/l)	110–340	392
γ-GTP (U/l)	12–87	37
LDH (IU/l)	107–230	169
BUN (mg/dl)	8.0–20.0	16.0
Cr (mg/dl)	0.61–1.04	.070
Na (mEq/l)	135–147	142
K (mEq/l)	3.3–5.0	4.1
Cl (mEq/l)	98–108	103
CPK (IU/l)	45–190	71
Serological tests		
CRP (mg/dl)	0–0.30	0.11
HBsAg		(-)
HBsAb		(-)
HCVAb		(-)
Coagulation		
PT (sec)	10.5–13.5	10.8
PT (%)	70–130	104.6
apTT(sec)	25–40	35.5

Thus, the symptoms and characteristics of IHPT are non-specific and its diagnosis is difficult and may be impossible except by percutaneous needle biopsy preoperatively.

Most importantly, it is difficult to differentiate IHPT from other malignant diseases such as hepatocellular carcinoma, intrahepatic cholangiocarcinoma, and metastatic tumor [12]. Findings on CT, MRI, and PET-CT of IHPT may mimic malignant liver tumors because of non-specific findings. In particular, for patients with malignant tumors and history of malignancy, IHPT is clinically and radiologi-

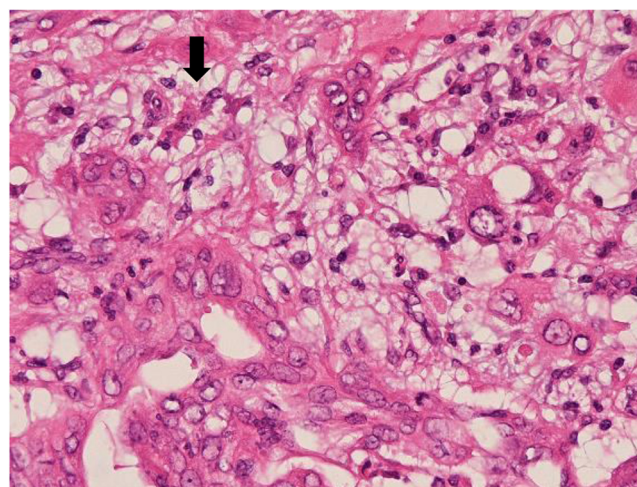


Fig. 4. Histological appearance (hematoxylin-eosin staining ×40). No malignant cells were identified in the tumor. However, a remarkable infiltration of inflammatory cells was observed (black arrow).

cally difficult to differentiate and may mimic malignant metastatic liver tumors.

We performed a literature search for “inflammatory hepatic pseudotumor” in the Ichushi-Web (Japan Medical Abstracts Society) and PubMed databases from April 2000 to October 2016. During this period, 193 cases of IHPT were reported. Of these, 44 patients (22.8%) had malignant tumors and a history of malignancy, 16 patients (8.3%) had multiple type tumors, and 9 patients (4.7%) had both (Table 2).

Five of 10 (50%) patients had post-biliary diseases such as cholangitis, cholecystolithiasis, and primary biliary disease, showing that IHPT might result from cholangitis as shown above (10). Over-treatment (resection or percutaneous transhepatic portal embolization) occurred in 7/10 (70%), showing that biopsy before surgery might be important; however, biopsy was performed in only 3 cases. The importance of percutaneous needle biopsy has been reported [13]. Therefore, percutaneous needle biopsy seems necessary to avoid over-treatment. Because this lesion was located on the surface of the S6 segment, we did not attempt percutaneous needle biopsy because of the risk of tumor cell dissemination.

Table 2
Cases of multiple inflammatory hepatic pseudotumors with malignant tumors and history of malignancy.

Case	Underlying cancer	Location (segment)	therapy	IgG4- related	Post-biliary disease	US	CT	MRI	PET	biopsy
1. 75 male	Gastric cancer	S3,S5,S6	observation	none	cholangitis	NA	done	done	NA	done
2. NA	Cholangio carcinoma	multiple	Partial resection of the liver	NA	NA	NA	done	done	NA	NA
3. 62 male	Rectal cancer	S5,S7	Partial resection of the liver	none	none	done	done	NA	done	NA
4. 73 male	Gastric cancer GIST	S2,S6,S7	Partial resection of the liver	related	cholangitis	done	done	done	NA	NA
5. 48 female	Pancreatic islet cell tumor	S4,S8	Partial resection of the liver	none	none	done	done	done	NA	done
6. 57 female	Sigmoid colon cancer	S5,S8	Portal vein embolization	NA	none	done	done	done	NA	NA
7. 77 male	Gastric cancer	S2,S3	observation	NA	none	NA	done	done	NA	NA
8. 63 male	Cholangio carcinoma	S6,S6	observation	NA	cholangitis	done	done	NA	NA	done
9. 75 male	Gastric cancer	S3,S5,S6	Partial resection of the liver	NA	Primary biliary cirrhosis	NA	done	done	NA	NA
Our case10. 67 male	Descending colon cancer	S6,S6	Partial resection of the liver	NA	cholecystolithiasis	done	done	NA	done	NA

Moreover, it seems infeasible to perform percutaneous needle biopsy in case of multiple liver tumors. Preoperative examination involved at least two examinations (such as CT and MRI). During examination, it might be important to ascertain lesion size. Repeated imaging studies over the course of a month showed the spontaneous regression of the hepatic tumors. This enabled us to make a diagnosis of IHPT without surgical resection. However, hepatectomy should be considered when a hepatic lesion is suspected to be malignant, in order to rule out malignancy, even if the lesion may be an inflammatory pseudotumor, especially in case of multiple hepatic masses.

4. Conclusion

IHPT is an important benign liver disease because it is difficult to clinically and radiologically distinguish from malignant tumors. Therefore, knowledge concerning this lesion is important so that treatment can be administered, even by physicians without experience. Percutaneous needle biopsy and adequate follow-up examinations are necessary examinations for cases of suspected single IHPT.

Conflict of interest

None of the authors has any conflict of interest to declare.

Funding source

None of the authors has any conflict of interest to declare.

Ethical approval

Authors has not gotten the ethical approval of this study by our ethics committee.

Consent

We were explained to the patient and relatives, and informed consent was obtained.

And we got permission from patients.

Author contributions

Obviously we surgeons performed this operation as a team.
Pathologist checked.

This team combination of our hospital could perform these therapies.

Registration of research studies

We got registered the Research Registry.
researchregistry2422.

Guarantor

Author Toshikatsu Nitta.

Acknowledgment

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