

Case Report

Inflammatory Pseudotumor of the Liver with Gastric Cancer : Report of a Case.

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Abstract

We report on a case of hepatic inflammatory pseudotumor (IPT) with gastric cancer. A 65-year-old man complained of upper abdominal pain, and double cancers of the stomach were detected by gastric endoscope, one of which was advanced and the other of which was early gastric cancer. A computed tomography (CT) scan revealed a solitary liver tumor, measuring 1.7 cm, located in the segment III. The CT scan also showed a ring enhancement of the tumor in the early phase and showed nonuniform late enhancement in the center of the tumor during the intermediate and late phases. We performed a dynamic CTHA, the center's staining became rich, and the rim firmly maintained its enhancement as time advanced. Moreover at the CTAP, the tumor showed a perfusion defect. The serum carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) levels were normal, as was α -fetoprotein (AFP), which suggested that the liver tumor was IPT. However, we were unable to rule out a metastatic liver tumor. We performed a total gastrectomy and wedge resection of the liver. IPT of the liver was confirmed by the histopathological findings of the resected specimens, which showed a multifocal invasion with inflammatory cells in the peripheral zone and rich fibrosis in the central zone of the tumor. We assume that the part of intense fibrous tissue part showed a ring enhancement and delayed enhancement.

(Key words : inflammatory pseudotumor, liver, gastric cancer, CT)

Introduction

Inflammatory pseudotumor (IPT) of the liver is a rare, benign tumor-like lesion¹⁾. IPT has sometimes been sometimes visualized as a hypoattenuation on enhanced CT scans, with the higher attenuation corresponding to areas of intense fibrosis, and the areas of lower attenuation corresponding to the predominantly cellular areas.²⁻⁶⁾ For this reason, IPT is sometimes misdiagnosed as a malignant tumor based on the imaging findings.¹⁾ There have been are just a few cases reported where IPT is associate with gastric cancer.

Case Report

A 65-year-old man, with upper abdominal pain and discomfort for two months, came to our hospital and underwent a gastric scope examination, which revealed double cancer of the stomach, one of which was a 3.5-cm disruption tumor beside the esophagocardial junction. The other was an early cancer lying in the mid-part of the stomach. The biopsy was moderately differentiated adenocarcinoma and he tested positive for hepatitis C virus antibody. Serum carcinoembryonic antigen levels, carbohydrate antigen 19-9 levels, α -fetoprotein levels were normal.

Percutaneous ultrasound findings revealed a heterogeneous low echographic mass with blood flow. The blood flow was found on some parts of the peripheral zone in the tumor.

A dynamic CT scan found space occupied in the lesion at the left ventral segment of the liver. During the pre-contrast CT, the entire tumor was a low-density lesion. However, during the early arterial-dominant phase, the tumor was enhanced nonuniformly with ring-like enhancement and during the late arterial-dominant phase, the center of the tumor was enhanced, subsequently revealing a similar density to the surrounding hepatic parenchyma of the liver in the late phase. The border of the tumor was unclear with no apparent peripheral washout signs.

Subsequently, we performed a dynamic CTHA (computed tomography during hepatic arteriography), whereupon the change in the contrast enhancement pattern of the tumor gradually spread from its center to the border. From the intermediate to the delayed phase, the center's staining became rich, and the rim firmly maintained its enhancement. [Fig.1 B. C. D. E].

Moreover at the CTAP (computed tomography during arterial portography) [Fig.1 A], the tumor showed a perfusion defect, which suggested that the tumor was not fed by a portal vein.

Therefore, we had differing diagnoses of liver tumor, metastatic tumor of gastric cancer, IPT, and hem-

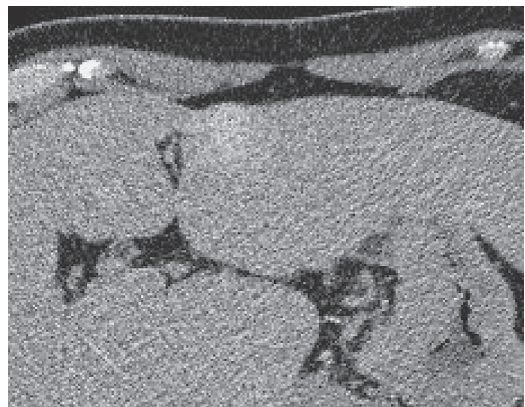
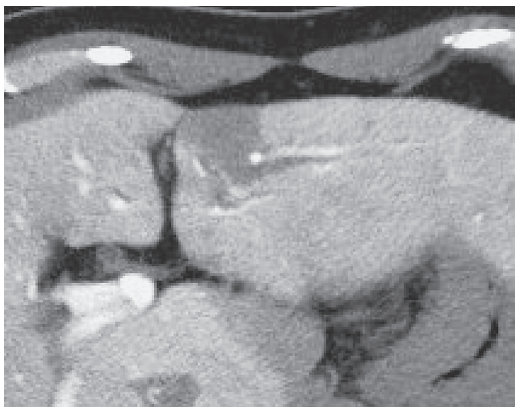
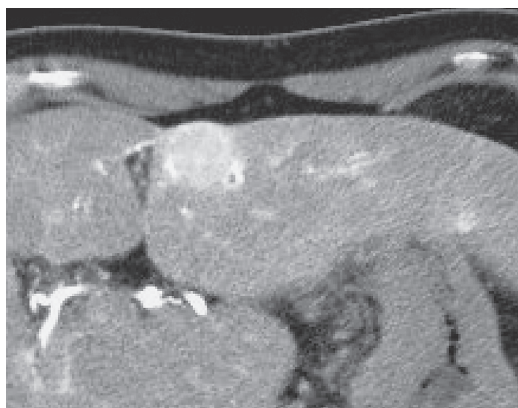


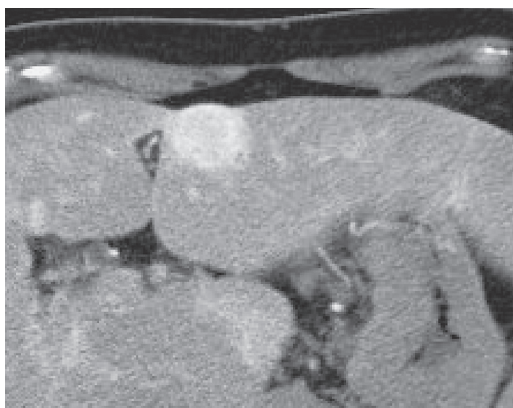
Figure.1

(A) CTAP (computed tomography during arterial portography), the tumor showed a perfusion defect, which suggested that it was not fed by the portal vein.

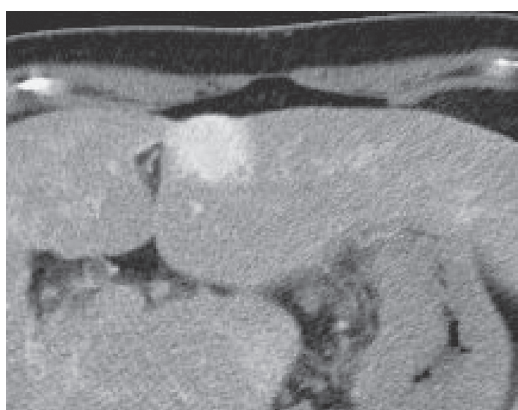
(B) is at the time before injection. The change in the contrast enhancement pattern of the tumor gradually spread from the center of the tumor to the border. During the intermediate



(C) CTHA (computed tomography during hepatic arteriography).



(D) to delayed



(E) phase, the center of the liver tumor became enhanced, and the rim firmly retained this enhancement.

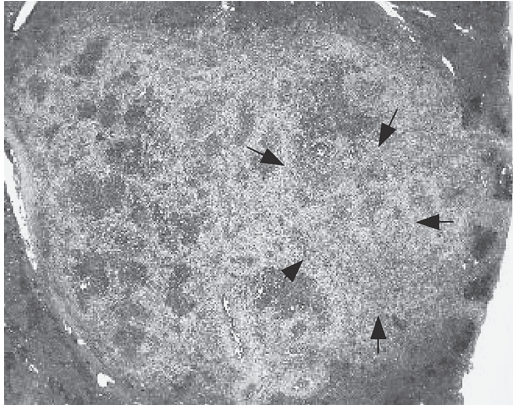
angioma, although there was some conflict with each diagnosis and imaging study.

We subsequently performed a total gastrectomy, and a wedge resection of segment III of the liver. The pathological findings were double cancer of the stomach and each depth was ss and sm, with one lymphatic metastasis (1/40). However, no cancer lesions were identified on the liver tumor and there was no evidence of necrosis or hemorrhage. There was, however, evidence of uneven distribution of invading multifocal inflammatory cells with dense fibril formation [Fig.1 F arrow] on the center of the tumor. The inflammatory cells were mainly composed of neutrophils, plasma cells, and lymphocytes, while the pathological findings confirmed a diagnosis of hepatic IPT. [Fig.1 F. G]

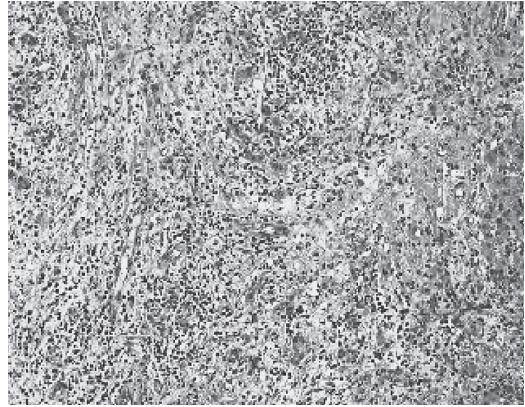
Discussion

The hepatic localization of IPT was first reported in 1953 by Pack and Baker,⁷⁾ and until recently, was considered a rare entity.⁸⁻⁹⁾ However, we were able to find more than 300 cases of inflammatory pseudotumor of the liver in a PubMed search. This lesion usually presents in young adults in their 30s, but can also occur in the elderly. IPT of the liver is frequently associated with symptoms such as fever, abdominal pain, anorexia, jaundice, and weight loss.¹⁰⁾

Sometimes, IPT with primary or secondary liver cancer is revealed in radiological studies.¹⁰⁾ The tumor in this patient showed low echogenicity and early contrast enhancement on dynamic CT without any washout phenomenon in the late phase, with hypervascularity on CTHA, with ring-like enhancement in



(F) Low-power view of the inflammatory pseudotumor of the liver. (X12.5) Uneven distribution of invading inflammatory cells was evident, with a dense fibril formation mainly in the center of the tumor (→).



(G) High-power view of the peripheral part of the tumor, showing granulation with capillary formation. (X200) Distributed foci of the inflammatory cells aggregated unevenly, with fibrosis.

the early arterial phase. These radiological findings are generally suggestive of metastatic tumor.

Based on the imaging findings, IPT is sometimes misdiagnosed as a malignant tumor, hepatocellular carcinoma (HCC), cholangiocellular carcinoma (CCC), metastatic liver tumor, or a liver abscess¹¹⁻¹²⁾. The differing ratios of cellular infiltration and fibrosis observed pathologically in IPT may explain the heterogeneity of CT and MRI findings²⁻⁶⁾. Heterogeneous enhancement of liver tumors on delayed-phase CT scans has been reported in some malignant tumors, liver abscesses and IPT, with the higher attenuation corresponding to areas of intense fibrosis, and areas with lower attenuation corresponding to predominantly cellular areas. This delayed enhancement in fibrous tissue is probably caused by extravascular contrast material due to the slow washout rate of the contrast material¹⁾, and the delayed peripheral rim-like enhancement with an internal low-density area seen in our case and some others reported in the literature may be an IPT finding³⁻⁶⁾.

Hepatic metastasis and CCC with abundant fibrosis also show delayed enhancement on CT scans, but delayed enhancement is seen in the central part of the tumor, while in HCC and liver abscess, delayed enhancement is seen in the peripheral part of the tumor in a ring-like form, and the capsule in HCC is thinner and smoother than in IPT. CT scans of liver abscesses typically show central near-water density, which differs from the solid nature of IPT. However, some liver abscesses that, which show incomplete liquefaction or have granulation within the abscess may mimic IPT on CT scans. CT-pathological correlation of the internal low density area, specified by the existence of blood supply and histological findings, could have been discussed on pre-contrast and arterial-dominant-phase CT scans.¹⁾

In this patient, part of the early enhanced area showed uniform foci of inflammatory cells with capillary formation, whereas part of the late enhanced area showed fibrous agglutination. Based on pathological findings, their distribution coexisted heterogeneously within the tumor. We were unable to clearly explain

the ring-like enhancement during the early phase, because there were differences between the pathological and the CT findings. If we could have the pathological slice conform to the CT's horizontal face, the distribution of inflammatory and fibrosis areas may well correspond with each other.

Uncertainty about the nature of IPT was manifested in an earlier report by the multiplicity of appellations including "plasma cell granuloma," "postinflammatory tumor," and "xanthomatous pseudotumor." Some clarity has come about through the recognition that a combination of histopathology, immunohistochemistry, and molecular and/or cytogenetic studies can differentiate one type of inflammatory pseudotumor from another. Inflammatory pseudotumor with the basic microscopic features of a mixed population of lymphocytes, plasma cells, and histiocytes in a variable cellular background of fibroblasts and myofibroblasts producing a mass lesion(s) can be a manifestation of an infection. On the other hand, a tumor with microscopic features resembling IPT, IMT (inflammatory myofibroblastic tumor), may indeed represent a follicular dendritic cell neoplasm upon immunophenotyping.¹³⁾ In this patient, there is no follicular dendritic cell neoplasm, so we refer to this tumor as IPT.

In conclusion, we have reported a case of IPT of the liver associated with gastric cancer. CTHA showed delayed enhancement, which might present areas of intense fibrosis. Although surgery is not obligatory for hepatic IPT, if suspicion exists surgical resection is carried out to evaluate the possibility of malignancy. If you have a much time to observe the course and wait, serial repeated imaging studies over the course of a month show the spontaneous regression of the hepatic tumor, thus enabling us to make a diagnosis of IPT without surgical resection.

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肝内炎症性偽腫瘍を合併した胃癌の症例

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要 約

65歳男性。心窩部痛を主訴に来院し上部消化管内視鏡にて噴門部に進行胃癌と胃体中部に早期胃癌を指摘され精査加療目的に入院となった。肝 S3に腫瘍を指摘された。肝 S3の病変はdynamic CT で内部が不均一に造影され、辺縁は早期相から造影効果を認め、中央部は遅延相にかけて造影された。CTAP では腫瘍は門脈血流欠損域として描出された。鑑別診断として転移性肝腫瘍、炎症性偽腫瘍、血管腫が挙げられた。胃全摘・肝部分切除術を施行し病理組織学

的検査を行ったところ、肝 S3結節には腫瘍細胞は認められず、多巢性に好中球主体の炎症細胞浸潤を認める線維化病変であった。腫瘍内に線維化を強く認める部分があり、それがCT所見において早期相では濃染が不良で後期相から遅延相にて濃染してくるパターンを取ったものと考えられた。CTと病理組織学的所見での線維化病変の分布の相違はそれぞれの切断面が異なったのが原因であると推察される。