


CASE REPORT

Rare cause of cystic liver lesions: Liver metastasis of gastrointestinal stromal tumors

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Abstract

Gastrointestinal stromal tumors (GISTs) are rare mesenchymal neoplasms of the gastrointestinal tract with an annual incidence of 1–2 per 10 000 people. Although most GISTs are solid, they may present with predominantly cystic components. A 69-year-old Japanese woman presented with a recently elevated gamma-glutamyl transpeptidase level. Computed tomography revealed multiple space-occupying lesions (SOLs) in the liver. These SOLs appear cystic on magnetic resonance imaging and abdominal ultrasound and are associated with thick walls at the margins. In addition, these thick walls showed high intensity on diffusion-weighted images. She was diagnosed with liver metastasis of GIST by diagnostic biopsies from the thick parts of the cystic liver lesion (thick walls at the margins). The primary lesion was thought to be located along the medial side of the descending part of the duodenum, but a duodenal biopsy was initially undiagnosed. Liver metastases due to GISTs are known to cause cystic changes after treatment, such as imatinib mesylate. However, to the best of our knowledge, only six cases where hepatic GIST with predominantly cystic changes (prior to any treatment) have been reported. It should be noted that GISTs appear cystic in all organs.

Introduction

Gastrointestinal stromal tumors (GISTs) are rare mesenchymal tumors of the gastrointestinal tract with an incidence of 1–2 per 10 000 people annually. The most common sites of occurrence are the stomach (59%), jejunum-ileum intestine (29.2%), and duodenum (4.5%).¹ The most common recurrence site of metastases is the liver (70–77%), and the rate of liver metastases in patients with a primary GIST was reportedly only 15.9%.² Cystic changes may also be observed in hepatic tumors, either due to necrosis or cystic degeneration of rapidly growing hypervascular tumors or as a manifestation of mucinous colonic or ovarian adenocarcinomas.³ Although most GISTs are solid, they may present with predominantly cystic components.

Case report

A 69-year-old Japanese woman presented with a recently elevated gamma-glutamyl transpeptidase level. Physical examination showed no symptoms or abnormalities. She had smoked 15 cigarettes daily for 45 years and had no history of alcohol consumption.

Laboratory tests showed the following: white blood cell count, 7500/ μ L (neutrophils, 71.3%; eosinophils, 0.7%); C-reactive protein level, 0.26 mg/dL; serum creatinine, 2.30 mg/dL; estimated glomerular filtration rate, 17 mL/min/1.73 m²; serum carcinoembryonic antigen level, 5.54 ng/mL; serum carbohydrate

antigen 19-9 level, 7.71 U/mL; serum alpha-fetoprotein level, 4.81 ng/mL; and serum des-gamma-carboxy prothrombin level, 14 mAU/mL.

Computed tomography (CT) revealed multiple space-occupying lesions (SOLs) in the whole liver and a 9-cm large lesion in the medial side of the descending part of the duodenum (Fig. 1a). Magnetic resonance imaging (MRI) revealed multiple cystic SOLs in the liver, showing high intensity on T2-weighted images (Fig. 1b). Liver SOLs also appear cystic on abdominal ultrasound (US) and are associated with thick walls at the margins, as well as MRI (Fig. 1b,e). Color Doppler imaging of US revealed no signal flow at the thick parts of the cystic lesions. In addition, these thick walls and mass lesions in the vicinity of the duodenum showed high intensity on diffusion-weighted images (Fig. 1c,d). Upper gastrointestinal endoscopy revealed an irregularly raised surface adjacent to the duodenal papilla (Fig. 1f).

The patient underwent diagnostic biopsies from the duodenum and thick parts of the liver lesion (thick walls at the margins). Histopathology revealed complicated spindle-shaped cells in the liver (Fig. S1A, Supporting information, hematoxylin and eosin stain; original magnification, 200 \times) and duodenal lesions (Fig. S1B, hematoxylin and eosin stain; original magnification, 200 \times). These tumors were c-Kit- and CD34-positive (Fig. S1C; liver, S1D; duodenum, original magnification, \times 200), but negative for S-100, cytokeratin AE1/AE3, and alpha-smooth muscle actin. Mitotic rate was almost unrecognizable in the 0–1 $>$ 50

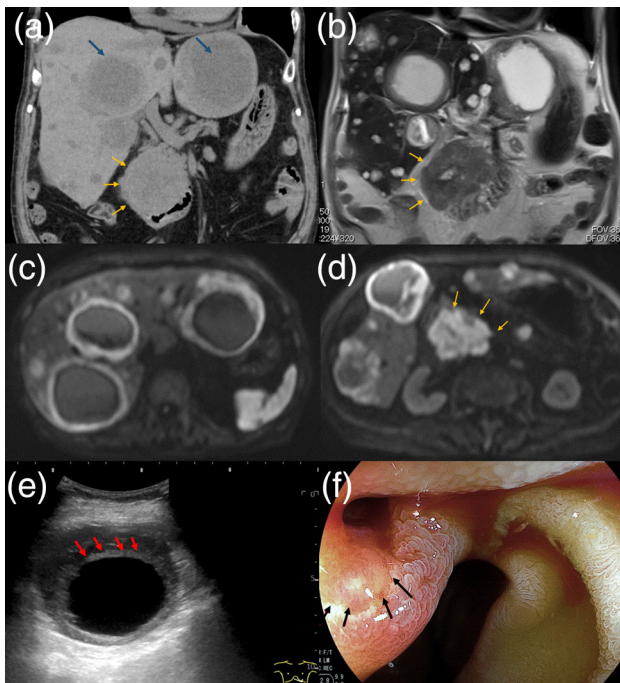


Figure 1 (a) Plain computed tomography revealing multiple space-occupying lesions (SOLs) in the liver (blue arrows). The densities of the central cystic area and marginal thickening area are 20 Hounsfield unit (HU) and 40 HU, respectively. (b) A 9-cm large lesion in the medial side of the descending part of the duodenum (yellow arrows). (c) Magnetic resonance imaging revealing multiple cystic SOLs in the liver that showed high intensity on T2-weighted images. (c,d) These thick walls and mass lesions in the vicinity of the duodenum show high intensity on diffusion-weighted images. (e) Liver SOLs also appear cystic on abdominal ultrasound. (f) Upper gastrointestinal endoscopy revealing an irregularly raised surface near the duodenal papilla.

high-power fields and showed a high Ki-67 labeling index (>5%). There was no evidence of necrosis or neovascularization in the collected tissues.

Discussion

Liver metastases of GISTs are known to cause cystic changes following treatment, such as imatinib mesylate.⁴ However, to the best of our knowledge, only six cases of hepatic GIST with predominantly cystic changes (prior to any treatment) have been reported (Table S1).^{5–10} In this case, a duodenal biopsy was initially undiagnosed; after a liver biopsy, which was performed with a risk for bleeding, we identified a GIST after examining the duodenal tissue. The unique radiological feature of this case is that there was a higher marginal attenuation area compared to that of the central area, which was different from a simple hepatic cyst. Although this case did not perform a contrast-enhanced CT and MRI because of renal dysfunction, neovascularization of the marginal thickening area of cystic lesions might help to diagnose cystic changes in GISTs.^{8–10} This patient does not wish to receive treatment and is being followed

up. It should be noted that GISTs appear predominantly cystic in all organs.

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Supporting information

Additional supporting information may be found in the online version of this article at the publisher's website:

Figure S1 (A; liver, B; duodenum) Histological examination with HE Staining revealed complicated spindle-shaped cells (original magnification, $\times 200$). (C; liver, D; duodenum) These tumors were c-Kit- and CD34-positive (original magnification, $\times 200$).

Table S1 Cystic hepatic metastases due to gastrointestinal stromal tumors prior to any treatment.