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## Interventional Radiology

# Percutaneous radiofrequency ablation for a recurrent metastasis after resection of liver metastases from an ileal clear-cell sarcoma: Long-term local tumor control

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### ABSTRACT

Clear-cell sarcomas (CCSs) in the gastrointestinal tract are extremely rare and aggressive tumors. We present the first case of a CCS arising in the ileum and metastasizing to the liver; our patient was a 60-year-old man. After the resection of the CCS and the liver metastases, a new liver metastasis developed, which was treated via percutaneous radiofrequency ablation only. At the 5-year follow-up, the ablated region was stable without local tumor progression. Percutaneous radiofrequency ablation is a viable local treatment option for recurrent metastases from an ileal CCS if they are detected when small and at an early stage in follow-up studies.

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## Introduction

Clear-cell sarcoma (CCS), also termed malignant melanoma of soft parts, is a rare soft tissue sarcoma with morphologic, immunohistochemical, and ultrastructural characteristics similar to those of malignant melanoma. CCS of the gastrointestinal tract is an extremely rare and aggressive disease. Here we present the first report of a CCS arising in the ileum and metastasizing to the liver. After the resection of the CCS and liver metastases, a new liver metastasis was treated via percutaneous radiofrequency ablation (RFA) only. At the 5-year follow-up, the ablated region was stable without local tumor progression.

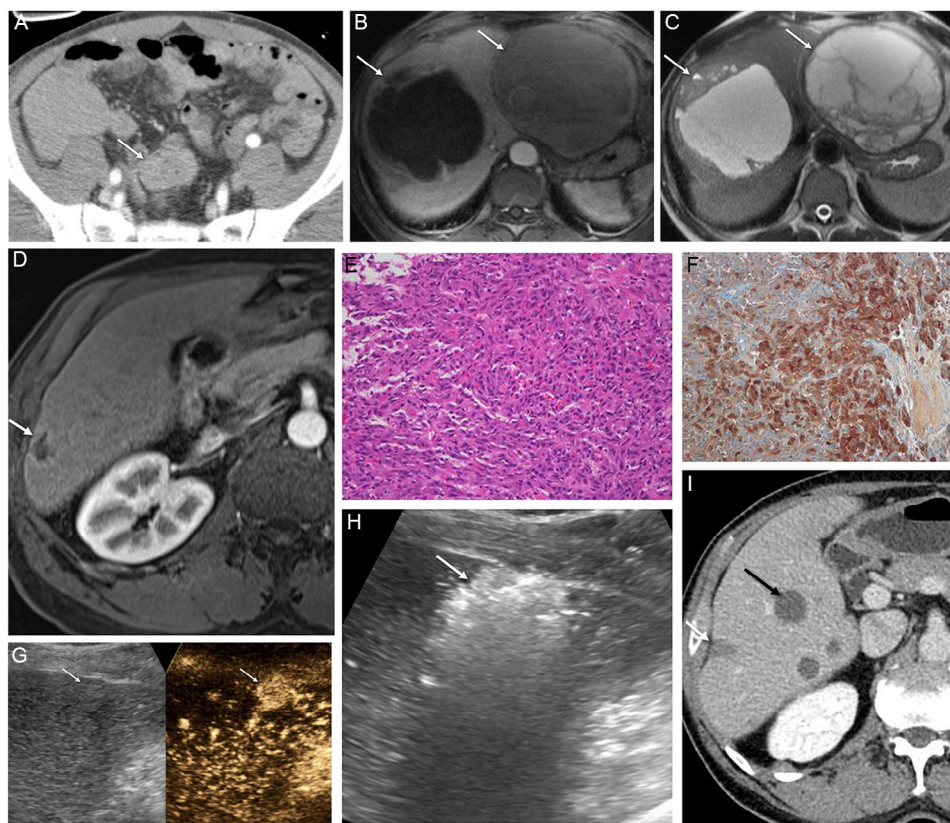
## Case presentation

A 60-year-old man was admitted to our hospital after experiencing melena for 7 days and hematochezia for 3 days. Contrast-enhanced abdominopelvic computed tomography (CT) showed a possible abnormal eccentric enhancing wall thickening in the ileal loop (Fig. 1A), and the colon was filled with high-density hemorrhagic fluid. Subsequent contrast-enhanced T1-weighted magnetic resonance imaging (MRI) revealed 2 large cystic tumors with enhancing papillary regions and septa in the hepatic lobes that appeared to be necrotic metastases or primary malignant biliary cystic tumors (Fig. 1B). T2-weighted MRI showed high-signal intensity masses with

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**Fig. 1 – Ileal clear-cell sarcoma with multiple hepatic metastases: (A)** Contrast-enhanced abdominopelvic computed tomography shows a possible abnormal eccentric enhancing wall thickening (arrow) in the ileal loop. **(B)** T1-weighted liver MRI reveals 2 large cystic tumors (arrows) with enhancing papillary portions and septa in the hepatic lobes. **(C)** T2-weighted MRI reveals high-signal intensity masses (arrows) with solid portions and multiple septa. **(D)** Contrast-enhanced T1-weighted liver MRI at the 2-year follow-up shows a 2.0-cm peripheral enhancing nodule with low signal intensity in segment 6 of the liver (arrow). **(E)** Hematoxylin- and eosin-stained tumor sections obtained via ultrasonography-guided biopsy have a nested or pseudoalveolar pattern ( $\times 100$ ). **(F)** Immunohistochemistry of tumor sections obtained as in panel E shows diffuse expression of S-100 protein in the tumor cells ( $\times 200$ ). The recurrent metastatic clear-cell sarcoma was confirmed histopathologically. **(G)** Contrast-enhanced ultrasonography for RFA planning reveals an ill-defined isoechoic lesion (arrow), with arterial hypervascularity appearing within 15 seconds after the injection of the contrast agent SonoVue (arrow) and washout at 30 seconds. **(H)** Percutaneous RFA was performed by using a 200-W generator in the impedance-controlled mode and a monopolar single internally cooled electrode with a 3-cm active tip, with hydrodissection for 12 minutes (arrow). **(I)** Five years after RFA, the ablated tumor region is stable without local tumor progression (white arrow). However, multiple new liver metastases are seen in other segments of the liver (black arrow). MRI, magnetic resonance imaging; RFA, radiofrequency ablation.

solid portions and multiple septa (Fig. 1C). On conventional superior mesenteric arteriography, there was some leakage of the contrast medium from a branch of the ileocecal or terminal ileal artery.

Active bleeding in the eccentric ileal mass was noted. Although superselective embolization was attempted, an autologous clot and arterial spasm developed and the bleeding stopped. Because the patient's hemoglobin level was below 6.9 g/dL (normal range, 13.0-17.0 g/dL), a small bowel resection of the enhancing eccentric mass in the ileum and a bisegmentectomy of the 2 large hepatic metastases were performed. Based on the final pathology examination of surgical specimens, a primary CCS arising from the ileum was diagnosed, and the hepatic masses were thought to be metastases of the CCS. To further treat the CCS, the patient underwent 6

cycles of combination chemotherapy consisting of cyclophosphamide, vincristine, adriamycin, and dimethyl-triazeno imidazole carboxamide.

Positron emission tomography-CT at the 2-year follow-up showed a focal uptake (standardized uptake value of 2.5) in segment 6 of the liver, and contrast-enhanced T1-weighted liver MRI revealed a 2.0-cm nodule with peripheral enhancing low-signal intensity in segment 6 (Fig. 1D). Ultrasonography-guided liver biopsy of the nodule was performed because a recurrent liver metastasis was suspected. The tumor was composed of sheets and bundles of cytologically uniform epithelioid cells with small nucleoli and eosinophilic cytoplasm. Microphotographs of hematoxylin- and eosin-stained tumor sections showed a characteristic nested or pseudoalveolar pattern (Fig. 1E). Immunohistochemical staining showed a diffuse

expression of S-100 protein in the tumor cells (Fig. 1F). Metastatic CCS was confirmed histopathologically.

We decided to treat the 2-cm hepatic metastatic tumor via percutaneous RFA because the patient refused additional hepatic segmentectomy, which was challenging owing to postoperative adhesion. For RFA planning, contrast-enhanced ultrasonography (CEUS) using SonoVue (Bracco, Milan, Italy) as the contrast agent was performed. CEUS showed an ill-defined isoechoic lesion in the B-mode, with rapidly appearing arterial hypervascularity (within 15 seconds after SonoVue injection) (Fig. 1G) and early washout (within 30 seconds). This description is characteristic of malignant hepatic tumors.

Percutaneous RFA was performed by using a 200-W generator in the impedance-controlled mode and a monopolar single internally cooled electrode with a 3-cm active tip (Valleylab, Burlington, MA), with hydrodissection for 12 minutes (Fig. 1H). There were no RFA-related complications, and immediate postablation evaluation via CEUS showed no evidence of a residual viable tumor. No additional cycles of chemotherapy were administered after RFA. Liver CT at the 1-month follow-up showed a hypodense lesion but no residual enhancing viable tumor. At the 5-year follow-up, the ablated region was stable without local tumor progression (Fig. 1I). However, multiple metastases in other segments of the liver and in the mesentery were observed 48 months after RFA.

## Discussion

CCSs have similar morphologic, immunohistochemical, and ultrastructural features as malignant melanomas, but a distinct molecular profile. CCSs usually develop in the tendons in the limbs and at the bottom layer of the skin. CCSs in the gastrointestinal tract are rare and aggressive [1,2] and usually originate in the ileum [1-3]; several cases have been reported recently. CCS is thought to result from a chromosomal translocation, and its symptoms usually reflect the aggressiveness of tumor invasion into other organs. Advanced CCSs may cause weight loss, appetite loss, and fatigue. The treatment of CCS includes surgery, radiation therapy, and chemotherapy.

The ileal CCS in the current case metastasized to the liver; after the resection of the primary tumor and liver metastases, another liver metastasis developed. In our patient, a 60-year-old man, the recurrent metastasis was treated via percutaneous RFA, and there was no local tumor progression in the following 5 years. Huang et al. used transcatheter arterial chemoembolization to treat diffuse hepatic metastases from a pancreatic CCS [4]. Iwamuro et al. used both transcatheter arterial chemoembolization and RFA to treat liver metastases from an intracranial hemangiopericytoma, an uncommon soft tissue sarcoma [5]. In that case, there was no local recurrence in the liver at the 5-year follow-up. In a patient with liver metastases from a primary malignant fibrous histiocytoma of the pancreas, Jarry et al. achieved an above-average survival rate by using a combination of chemotherapy, percutaneous RFA, and hepatectomy [6].

As suggested by Jones et al., inclusion of RFA in the multimodality management of patients with gastrointestinal stromal tumors may be useful when there is only a single

hepatic metastasis and the disease is otherwise stable [7]. In a single-center study of sarcomas that metastasized to the liver, Pawlik et al. found that RFA (with or without resection) and no adjuvant chemotherapy predicted shorter survival times [8]. To prolong the survival of patients with sarcoma-derived liver metastases, Pawlik et al. emphasized the need for both adjuvant chemotherapy and multidisciplinary patient evaluation. We agree with their recommendations, especially in terms of the aggressive case presented here. As imaging modalities such as CT and MRI improve, early detection of small recurrent sarcoma-derived liver metastases should increase. Thus, local ablation is likely to be more helpful in the future, especially when combined with surgery and chemotherapy.

In our case, the patient underwent small bowel resection for an ileal mass and bisegmentectomy for 2 large liver metastases, after which he underwent 6 cycles of combination chemotherapy (cyclophosphamide, vincristine, adriamycin, and dimethyl-triazeno imidazole carboxamide). Within 2 years, another hepatic metastasis, about 2 cm in size, developed and was treated via percutaneous RFA only. At the 5-year follow-up, the ablated region was stable without local tumor progression. After RFA, there was a second recurrent hepatic metastasis, as well as extrahepatic mesentery metastases 48 months after RFA. We thought that combined chemotherapy after RFA might prolong the recurrence-free period, but the patient refused additional chemotherapy. However, percutaneous RFA did effectively treat the initial recurrent liver metastasis, preventing local tumor progression.

RFA is an effective and safe local treatment for small hepatic metastases (3 cm or less) from colorectal cancers. We thought it appropriate for the treatment of the small hepatic CCS-derived metastasis in our case, especially considering the difficulty of additional surgery. RFA may aid in inhibiting recurrence when combined with other procedures such as surgery, radiotherapy, and chemotherapy. Moreover, RFA may effectively treat recurrent hepatic metastases from an ileal CCS if they are detected when small and at an early stage in follow-up studies.

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