

# World Journal of *Clinical Cases*

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## Multimodal treatments of right gastroepiploic arterial leiomyosarcoma with hepatic metastasis: A case report and review of the literature

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### Abstract

Leiomyosarcoma of an artery is very rare, and cases with hepatic metastasis are even rarer. We describe a case of a 70-year-old man who after follow up due to rectal cancer, presented with an intra-abdominal hypervascular mass and a hepatic mass. After surgical resection, it was diagnosed as a leiomyosarcoma of the right gastroepiploic artery with hepatic metastasis. Multiple metastases had recurred at the liver. He has survived more than 53 mo through multimodal treatments (three surgical resections, radiofrequency ablation, transarterial chemoembolization, chemotherapies, and targeted therapy). Multimodal treatments, including active surgical resection, may be



helpful in the treatment of aggressive diseases such as arterial leiomyosarcoma with metastasis.

**Key words:** Multimodal treatments; Intra-abdominal arterial leiomyosarcoma; Hepatic metastasis; Arterial leiomyosarcoma; Surgical resection

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**Core tip:** An arterial leiomyosarcoma (aLMS) is a very rare and aggressive disease. The prognosis is also very poor. A 70-year-old man presented with an intra-abdominal aLMS with hepatic metastasis. He was treated with multimodal treatments that consisted of three surgeries, radiofrequency ablation, transarterial chemoembolization, and targeted therapy. He has survived for 53 mo after these treatments. Multimodal treatments could be helpful treating this kind of disease.

Seo HI, Kim DI, Chung Y, Choi CI, Kim M, Yun S, Kim S, Park DY. Multimodal treatments of right gastroepiploic arterial leiomyosarcoma with hepatic metastasis: A case report and review of the literature. *World J Clin Cases* 2018; 6(8): 219-223 Available from: URL: <http://www.wjgnet.com/2307-8960/full/v6/i8/219.htm> DOI: <http://dx.doi.org/10.12998/wjcc.v6.i8.219>

## INTRODUCTION

The leiomyosarcoma (LMS) is very rare malignant tumor. They usually originate in the smooth muscle of the soft tissues and uterus<sup>[1]</sup>. About 2% of LMS cases occur in the smooth muscle of the vessel wall and 60% occur in the inferior vena cava. The occurrence of LMS involving the veins is about five times higher than that of the arteries<sup>[1]</sup>. The most common site of arterial LMS (aLMS) is the peripheral artery, and the intra-abdominal artery is a rare location for aLMS to occur<sup>[1]</sup>. To the best of our knowledge, this is the first presentation of intra-abdominal aLMS with distant single liver metastasis. We report the clinical course of aLMS that originated from the right gastroepiploic artery with hepatic metastasis during multimodal treatments [three surgical resections, radiofrequency ablation (RFA), transarterial chemoembolization (TACE), chemotherapy, and targeted therapy] and review the literature regarding aLMS.

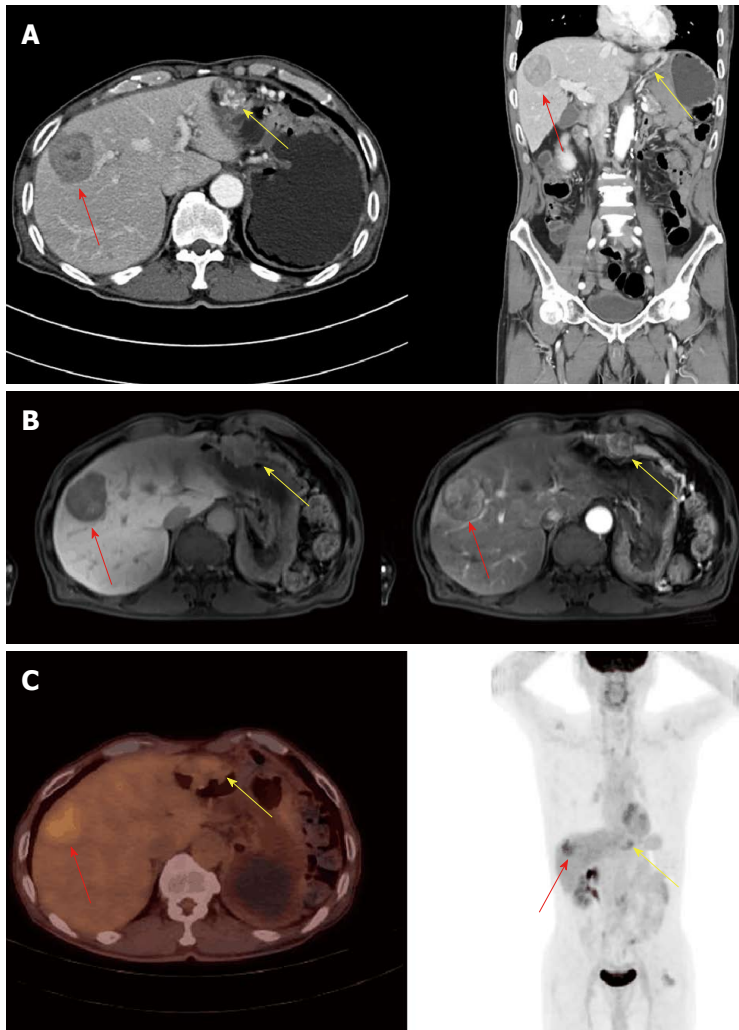
## CASE REPORT

This case involves a 70-year-old man who had a previous operation history due to renal cell carcinoma and rectal cancer (pT2N0M0, stage IIA), ten years and six months ago. Abdominal computed tomography (CT) and magnetic resonance imaging (MRI) performed six months after the low anterior resection revealed a new 47 mm hypodense hepatic mass and a 23 mm hypervascular mass at the great curvature side of sto-

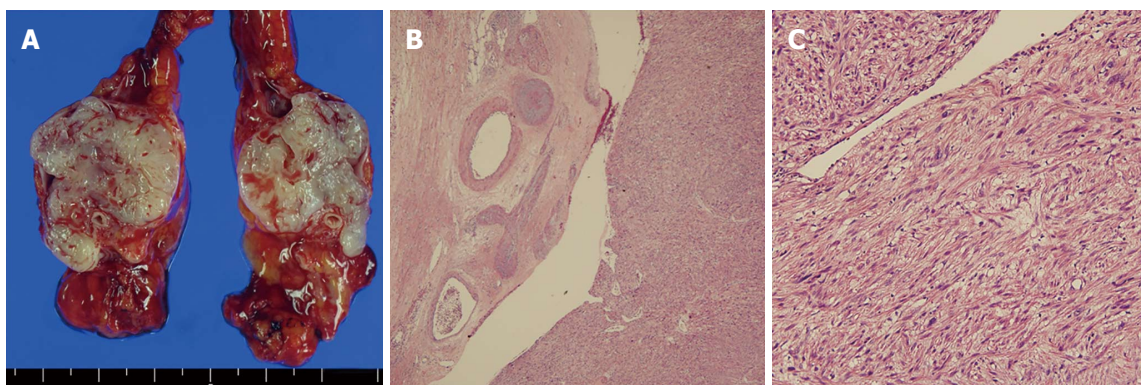
mach (Figure 1A and 1B). It was highly suspected to be a malignant gastrointestinal stromal tumor (GIST) with hepatic metastasis. Fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (FDG PET/CT) demonstrated a hypermetabolic low-density lesion in S8 of the liver and the greater curvature of the stomach with a maximum standardized uptake value (SUVmax) of 3.4 and 2.3, respectively (Figure 1C). For confirmation of the diagnosis, we planned to perform an ultrasonography-guided core needle biopsy. The core needle biopsy specimen of the liver mass showed malignant spindle cell with increased mitosis (7/10 HPFs). Immunohistochemistry results were positive for desmin and smooth muscle actin (SMA) and negative for CD34, c-kit, DOG-1, S-100, and HMB45. These findings suggest that aLMS was the proper diagnosis, not GIST. The mass in the greater curvature of the stomach showed high vascularity upon endoscopic ultrasonography. Therefore, a fine needle aspiration biopsy was not performed because of a bleeding risk. Laparotomy was performed with a diagnosis of the omental GIST and primary hepatic LMS. The omental mass resection and S8 segmentectomy were performed. The omental mass originated from the right gastroepiploic artery on surgical and microscopic field. This mass was a 3.0 cm × 2.7 cm sized aLMS (Figure 2A and 2B). Histopathology showed moderate cellular atypia, high mitotic rate (10/10 HPFs), and 2/3 histologic grade according to the FNCLCC grading system. Ki-67 proliferation index was 4.1% (Figure 2C). Immunohistochemistry results were positive for CD34, CD31, desmin, and SMA and negative for c-kit, DOG-1, and S-100. The liver mass was a 5.0 cm × 3.0 cm × 1.5 cm sized metastatic aLMS with a clear resection margin (free margin: 0.3 cm). Ki-67 proliferation index was 9.3%. The patient was discharged on the nine days after the operation without any complications. It was planned that four cycles of adriamycin monochemotherapy would be administered as an adjuvant treatment. However, treatment was stopped after the third treatment because of neutropenic fever.

Abdominal CT was performed every three months after the operation to check for recurrence. At fourteen months after the first operation, CT and MRI revealed a 2.7 cm, a 5 mm, and an 8 mm sized metastatic masses on of the liver. No extrahepatic metastasis was noted on FDG PET/CT. Right anterior sectionectomy was performed fifteen months after the first operation. There were a 3.0 cm × 2.7 cm and a 1.0 cm × 1.0 cm sized metastatic vascular LMSs. Histopathology and immunohistochemistry showed a high mitotic rate (22/10 HPFs) and a Ki-67 proliferation index of 4.1%. The resection margin was very close to the mass (< 1 mm). Ifosfamide monochemotherapy was administered after the surgery for four cycles.

At eight months after the second operation, CT, MRI and FDG PET/CT revealed a 1.6 cm and a 1.4 cm sized seeding metastatic nodules on the diaphragm and the liver. Diaphragm partial resection and intra-operative



**Figure 1 Diagnosis imaging of the patient.** A: Computed tomography showed a 43 mm hypodense mass at S8 of the liver (red arrow) and a 23 mm sized hypervascular mass at the great curvature side of stomach (yellow arrow). B: Magnetic resonance imaging showed a well-defined encapsulated lesion (red arrow) in S8 of the liver, which showed a strong enhancement during the arterial dominant phase, with wash out during the delayed phase. The mass (red arrow) in the greater curvature of the stomach was accompanied by engorgement of the gastroepiploic vein. C: Fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography images showed a hyper-vascular mass at the great curvature side of stomach (yellow arrow) and a hypervascular metastatic mass at S8 of liver (red arrow).

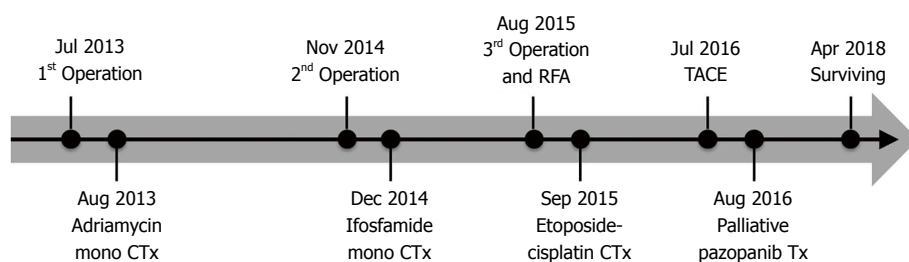


**Figure 2 The gross find of arterial leiomyosarcoma and pathological diagnosis.** The omental mass and A: Gross finding of arterial leiomyosarcoma. A white colored expending nodular mass was present on soft tissue. A medium sized artery was present on adjacent connective tissue. B: Tumor consisted of spindle cells and adjacent to medium sized vessels (H and E staining, × 40). C: Tumor cells showed spindle shaped nucleus with rounded end and eosinophilic cytoplasm (H and E staining, × 200). They formed fascicular pattern and frequently made stage horn shaped vascular spaces.

**Table 1** Review of intra-abdominal arterial leiomyosarcoma literature

Ref.	Sex	Age (yr)	Site	Symptom	Treatment	Metastasis	Follow-up
Hopkins <sup>[2]</sup> (1968)	M	55	Common iliac	Claudication	Surgery	-	Died after 3 wk
Birkenstock <i>et al</i> <sup>[3]</sup> (1976)	M	55	Common iliac	Pain, palpable mass	Surgery	-	No evidence of disease
Stringer <sup>[4]</sup> (1977)	M	49	IMA	Pain, palpable mass	Surgery, RTx, CTx	Lung	Died after 7 yr
Gutman <i>et al</i> <sup>[5]</sup> (1986)	F	55	Common iliac	Claudication	Surgery	-	No evidence of disease
Delin <i>et al</i> <sup>[6]</sup> (1990)	F	72	Common iliac	Claudication, pain	Surgery	-	Died after 7 mo
Gill <i>et al</i> <sup>[7]</sup> (2000)	F	76	Renal	Pain	Surgery	-	Follow up for 9 mo
Rohde <i>et al</i> <sup>[8]</sup> (2001)	F	51	Splenic	Pain, weight loss	Surgery + CTx (adriamycin + ifosfamide)	-	Follow up for 1 yr
Blansfield <i>et al</i> <sup>[9]</sup> (2003)	F	42	Common iliac	Pain	Surgery	-	No evidence of disease
Current case	M	70	Right gastroepiploic	None	Multimodal	Liver	follow up for 36 mo

IMA: inferior mesenteric artery; RTx: radiotherapy; CTx: chemotherapy.

**Figure 3** Timeline of multimodal treatments. CTx: chemotherapy; RFA: radiofrequency ablation; TACE: transarterial chemoembolization

RFA was performed nine months after the second operation (twenty-four months after the first operation). The diaphragm mass was also diagnosed as a metastatic vascular LMS. Etoposide-cisplatin chemotherapy was administered after the surgery for six cycles.

At eleven months after the third operation, there were multiple recurrences in the remnant liver as shown by MRI and TACE (2 mL lipiodol and 10 mg adriamycin). Palliative pazopanib was also started. The patient has been followed for twenty-eight months after the third operation (fifty-three months after the first operation). He is surviving with stable hepatic metastasis controlled by chemotherapy. Treatment modalities are summarized in Figure 3.

## DISCUSSION

Over half of aLMS cases originate from the pulmonary artery, followed by the extra-abdominal peripheral artery. Including this case, only nine cases of aLMS originating from the intra-abdominal arteries, excluding the aorta, have been reported (Table 1)<sup>[2-9]</sup>.

Compared to the other origin of LMS, the case of vascular LMS has a worse prognosis. In the case of LMS with metastasis, metastatic vascular LMS shows similar results as metastatic LMS of other origins<sup>[1]</sup>. However,

the prognosis is not well-known owing to the low number of cases in aLMS. It is presumed that aLMS may be more aggressive than LMS, and hence, the prognosis is expected to be about the same or worse<sup>[1]</sup>. Because aLMS is more aggressive than LMS, and directly seed to artery, it has a higher possibility of metastasis<sup>[10]</sup>.

Clinical signs of aLMS are diverse depending on the area of origin and most of them are due to the mass effect<sup>[9,10]</sup>. In this case, the 3 cm primary mass was located in the intra-abdominal area. The patient had no symptoms owing to aLMS. Although, the size of the primary cancer of the right gastroepiploic artery was 3 cm, which was small, it was accompanied by a distant metastasis at the time of diagnosis.

For diagnostic confirmation, a biopsy is needed, and radiological assisted core needle biopsy is preferred over open biopsy because there is a low risk of complications<sup>[10]</sup>. However, like this case, if the mass is located in the intra-peritoneal region and shows hypervascularity, bleeding could occur after the core needle biopsy. Moreover, bleeding control could be difficult in this location. PET-CT showed SUVmax values of 3.4 and 2.3 each, which had relatively low uptake at first, and uptake was not noted at the lesion recurrence. More precise imaging studies are needed to overcome this limitation.

There are reports of treating LMS cases with surgical



resection, radiotherapy, and chemotherapy<sup>[1,4,6,9]</sup>. Chemotherapy or chemoembolization has been the main treatment of LMS with hepatic metastasis<sup>[10]</sup>. Recently, RFA also shows a good result for metastatic LMS<sup>[10]</sup>. However, recently, just like in other cases of metastatic cancer, liver resection shows better results<sup>[11]</sup>. If a resection of metastasis is possible, surgical treatment and additional treatment including chemotherapy can lead to a good response.

Although aLMS showed aggressive clinical features, multimodal treatment (resection, chemotherapy, RFA, chemoembolization, and targeted therapy) might be helpful to manage this kind of disease.

## ARTICLE HIGHLIGHTS

### Case characteristics

A 70-year-old man presented with an intra-abdominal mass and a hepatic mass during a follow visit for rectal cancer surgery.

### Clinical diagnosis

After CT and magnetic resonance imaging (MRI), it was diagnosed as a malignant gastrointestinal stromal tumor (GIST) with hepatic metastasis.

### Differential diagnosis

After the core needle biopsy of the liver, it was diagnosed as a leiomyosarcoma (LMS). Before the surgery, these were omental GIST and hepatic LMS.

### Imaging diagnosis

At first, it was diagnosed a omental GIST and hepatic metastasis in CT and MRI.

### Pathological diagnosis

The surgical specimen diagnosed as an aLMS with hepatic metastasis.

### Treatment

Multimodal treatments were done (three surgeries, chemotherapy, transarterial chemoembolization, radiofrequency, and targeted therapy).

### Related reports

There were only nine reports about intra-abdominal arterial leiomyosarcoma (aLMS). This is the first report of intra-abdominal aLMS with hepatic metastasis.

### Term explanation

aLMS is a very rare and aggressive disease. The prognosis is very poor. There

were few reports of this disease. So the treatment is also not established.

## Experiences and lessons

Active treatments using multiple modalities may be helpful for these kinds of patients.

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