

Complete resection and arterial reconstruction for primary sarcoma arising from superior mesenteric artery

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ABSTRACT

In the treatment of retroperitoneal sarcoma involving major vessels, complete resection with vascular reconstruction is challenging. We describe the case of a 72-year-old man who presented with 8 months of abdominal pain. Diagnostic workup revealed occlusion of the celiac trunk and the origin of the superior mesenteric artery due to a soft tissue sarcoma. Radical resection of the tumor and vessels was performed. Guided by intraoperative angiography, arterial reconstruction was performed without mesenteric ischemia. All arterial margins were negative. At the 6-year follow-up, the patient was alive with no evidence of recurrence. (*J Vasc Surg Cases Innov Tech* 2022;8:70-4.)

Keywords: Pleomorphic sarcoma; Arterial reconstruction; Complete resection

Complete resection with a sufficient surgical margin is fundamental for the treatment of retroperitoneal soft tissue sarcomas. The involvement of major vessels may become a limitation for complete resection, which makes the decision regarding vascular reconstruction difficult. Few studies have reported a complete resection with vascular reconstruction. We present a case that underwent resection with arterial reconstruction guided by imaging modalities. The patient provided written informed consent for this case report.

CASE

A 72-year-old man presented with 8 months of intermittent abdominal pain. His medical history was significant for hypertension and cholecystectomy. Laboratory data were normal. Abdominal ultrasound examination revealed a retroperitoneal mass. Computed tomography angiography (CTA) suggested that the mass might be a superior mesenteric artery (SMA) aneurysm. The patient was referred to our hospital for further evaluation of the mass. CTA showed a 30-mm mass centered within an SMA with wall enhancement, a partial filling defect in the distal SMA (Fig 1, A and B), and occlusion of the celiac artery (CA) due to direct compression by a median

arcuate ligament. On the basis of these CTA findings, we suspected a neoplastic lesion.

Magnetic resonance imaging showed a round mass with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. ¹⁸F-fluorodeoxyglucose positron emission tomography revealed high uptake lesions in the mass around the origin of the SMA, with a maximal standardized uptake value of 28.5 (Fig 1, D).

Transfemoral angiography showed occlusion of the origin of the celiac trunk and a short segmental stenosis of the SMA. The distal CA was compensated for by collateral circulation from the inferior mesenteric artery. The flow of the SMA was partially defective from the origin to the proximal part of the jejunal artery.

A biopsy specimen was obtained from the mass using endoscopic ultrasound-guided fine-needle aspiration. The histologic examination findings were inconclusive, but consistent with high-grade sarcoma.

We decided to perform surgical resection of the tumor and arterial reconstruction. Surgery was performed 3 months after biopsy.

The tumor was approached using median laparotomy. After Kocherization and dissection of the space behind the pancreas body, the origin of the SMA was identified, which was not pulsatile.

Intraoperative arterial ultrasound examination revealed an occlusion of the SMA from the origin to the level of the ileocolic artery (ICA) and the extent of the tumor distal to the ICA (Fig 2, A). Intraoperative angiography showed that the inferior mesenteric artery supplied blood flow to the small intestine through the ICA and to the celiac trunk through the dorsal pancreatic artery because of occlusion of the celiac trunk and the origin of the SMA. After transection of the SMA proximal to the ICA, a saphenous vein graft (SVG) was anastomosed to the end of the SMA (Fig 2, B). We supplied the blood flow through the anastomosed SVG from a sheath

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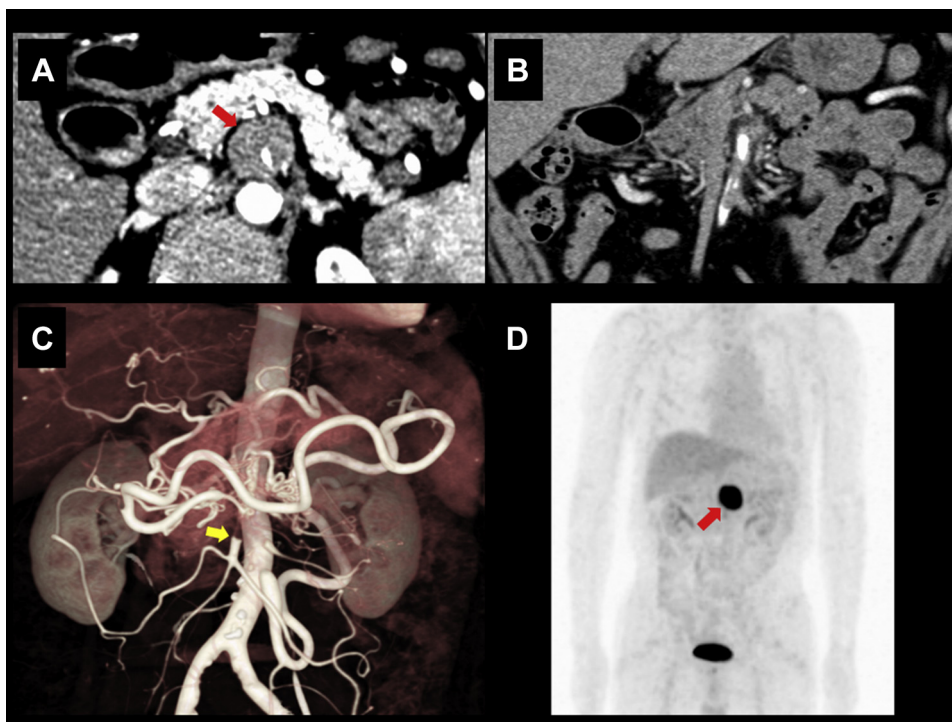


Fig 1. Preoperative computed tomography images. The *red arrows* indicate a round mass centered into the superior mesenteric artery (SMA). **A**, Axial image demonstrating a mass centered within an SMA with wall enhancement. **B**, Coronal image demonstrating a partial filling defect in the distal SMA. **C**, Three-dimensional computed tomography angiography (CTA). The SMA was occluded to the range indicated by a *yellow arrow*. **D**, ¹⁸F- fluorodeoxyglucose (FDG) positron emission tomography/computed tomography on coronal images. FDG uptake (a maximal standardized uptake value = 28.5) was shown, suggestive of a malignant lesion.

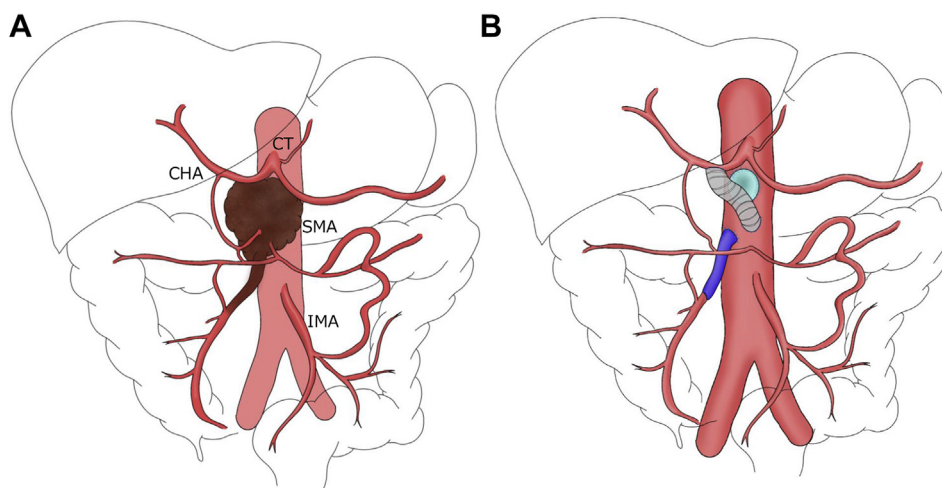


Fig 2. **A**, Illustration of a tumor and visceral perfusion. **B**, Illustration of final arterial reconstruction. *CHA*, Common hepatic artery; *CT*, celiac trunk; *IMA*, inferior mesenteric artery; *SMA*, superior mesenteric artery.

inserted in the right femoral artery after an anastomosis to the distal end of the SMA. The aorta was cross-clamped at two levels above the celiac trunk and above the renal arteries under systemic heparinization. Radical tumor resection was performed, which included a part of the aortic wall around the origin of the SMA to secure a

resection margin, although aortic invasion was unclear. The defect of the aorta was reconstructed using a 20 × 30 mm Dacron patch. The proximal end of the SVG was anastomosed to the aorta, and the aortic cross-clamp was removed. The aortic cross-clamp time was 60 minutes. We found hardly a pulse in the common

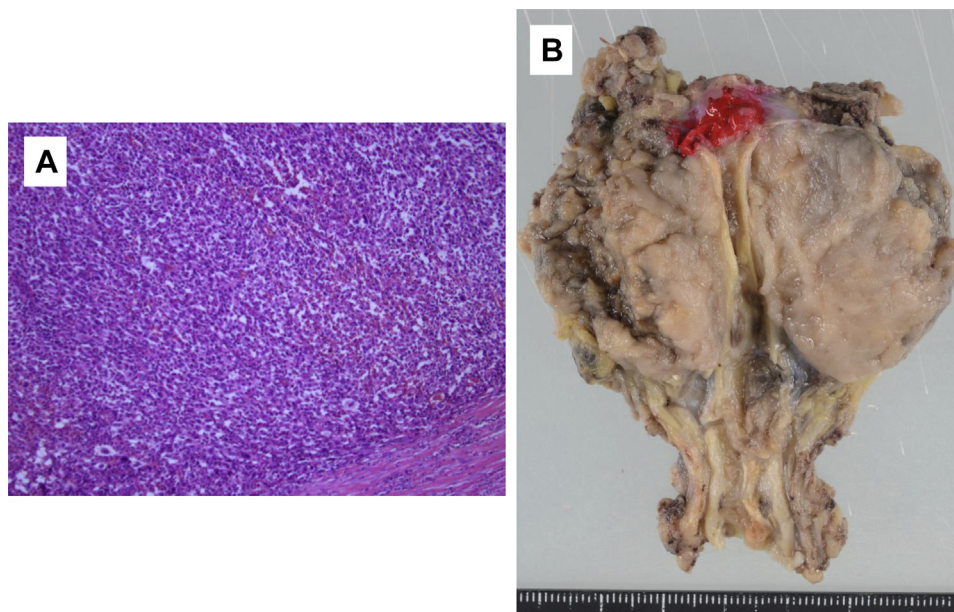


Fig 3. **A.** Hematoxylin and eosin staining of the tumor. The tumor was composed of spindle-shaped cells arranged in fascicles. **B.** Resected sarcoma tissue. The origin of a superior mesenteric artery (SMA) was marked with red ink.

hepatic artery (CHA) and splenic artery. This could be caused by reducing the collateral source from the SMA to the CA after the resection of the SMA. Therefore, we decided to perform an arterial bypass for the CHA. The bypass was performed without incision of the median arcuate ligament because the CHA was sufficiently exposed. An 8-mm ringed polytetrafluoroethylene graft was anastomosed to the CHA. After aortic cross-clamping, the end of the graft was anastomosed to the infrarenal aorta. All the arterial margins were negative. Postprocedural angiography revealed satisfactory results. The operation required 14 hours. The total blood loss was 2700 mL.

The patient experienced diarrhea postoperatively. The stool bacterial samples tested negative for pathogens. Diarrhea was considered to be associated with the resection of parasympathetic nerve fibers around the SMA. The patient was discharged on postoperative day 25 with aspirin.

Follow-up CTA revealed a local recurrence around the site of the reconstruction with a patch 6 months postoperatively.

After pathological diagnosis, adjuvant chemotherapy was administered including adriamycin, fixed-dose rate gemcitabine, docetaxel, and gemcitabine. Although the regimen of gemcitabine was stopped because of severe fatigue, he is alive with no evidence of recurrence at the 6-year follow-up by an interdisciplinary team at our institution. The duration of adjuvant chemotherapy was 8 months.

HISTOLOGIC FINDINGS

Immunohistochemistry showed that the tumor was negative for cytokeratins, S-100, desmin, CD34, C-kit, HHF35, and α -SMA. Histopathological analysis of the resected tumor revealed an undifferentiated pleomorphic sarcoma (Fig 3). The final resection lines and the resected aorta were tumor-free.

DISCUSSION

Retroperitoneal sarcomas are extremely rare, especially when they involve the major vessels. Schwarzbach et al¹ experienced 141 surgeries for retroperitoneal soft tissue sarcoma, and 17.7% of them involved major retroperitoneal blood vessels (Supplementary Table). Furthermore, only one patient had a primary sarcoma with isolated arterial involvement. Few studies have described a primary sarcoma originating from the arterial vessels, except for the aorta.²

The long encasement of the SMA is generally considered a nonresectability criterion.^{3,4} In our case, we were able to perform a complete resection and an anatomical reconstruction because the tumor encased a proximal portion despite its considerably long segment. Intraoperative angiography led to the accurate assessment of tumor extension and intestinal perfusion, and we determined the range of the resection and the anastomotic site.

Poultides et al⁵ reported a poor prognosis in patients with retroperitoneal sarcomas involving major vessels, even if vascular reconstruction was performed.

Stojadinovic et al⁶ reported that 77% of retroperitoneal sarcoma-related deaths were caused by local recurrence. Especially in the case of an undifferentiated pleomorphic sarcoma, a poor prognosis has been reported. Metastatic rates of 50% have been previously reported,⁷ and the reported 5-year survival rate was 4% in patients with metastasis.⁸ Therefore, we believe that complete resection of the tumor, including invaded major vessels, offers a better prognosis. A previous study reported that complete resection with a negative margin was a significant prognostic factor for survival.¹ Our patient was still alive with no evidence of recurrence during the 6-year follow-up because a complete resection with microscopically negative resection margin was achieved. Although the role of adjuvant chemotherapy is of no study-proven value,³ it may be considered for individual patients with potentially chemosensitive subtypes.⁹

Extensive dissection of the SMA and subsequent diarrhea are considered to be associated with nerve damage.^{10,11} In our case, the origin of the SMA, which was encased by the tumor, had to be fully exposed. Accordingly, this complication is unavoidable and usually improves gradually over time.

CONCLUSIONS

On the basis of preoperative diagnosis by interdisciplinary studies, we aggressively approached the tumor with the aim of complete resection.

Accurate evaluation guided by imaging modalities, including intraoperative angiography, led to the

achievement of necessary and sufficient arterial reconstruction without mesenteric ischemia.

REFERENCES

1. Schwarzbach MHM, Hormann Y, Hinz U, Leowardi C, Bockler D, Mechtersheimer G, et al. Clinical results of surgery for retroperitoneal sarcoma with major blood vessel involvement. *J Vasc Surg* 2006;44:46-55.
2. Fatima J, Duncan AA, Maleszewski JJ, Kalra M, Oderich GS, Gloviczki P, et al. Primary angiosarcoma of the aorta, great vessels, and the heart. *J Vasc Surg* 2013;57:756-64.
3. Trans-Atlantic RPS Working Group. Management of primary retroperitoneal sarcoma (RPS) in the adult: a consensus approach from the Trans-Atlantic RPS Working Group. *Ann Surg Oncol* 2015;22:256-63.
4. Dimitri T, Bouhadiba T, Gaignard E, Bonvalot S. Major vascular resections in retroperitoneal sarcoma. *J Surg Oncol* 2018;117:42-7.
5. Poultsides GA, Tran TB, Zambrano E, Janson L, Mohler DG, Mell MW, et al. Sarcoma resection with and without vascular reconstruction: a matched case-control study. *Ann Surg* 2015;262:632-40.
6. Stojadinovic A, Yeh A, Brennan MF. Completely resected recurrent soft tissue sarcoma: primary anatomic site governs outcome. *J Am Coll Surg* 2002;194:436-47.
7. Hornick JL. Subclassification of pleomorphic sarcomas: How and why should we care? *Ann Diagn Pathol* 2018;37:118-24.
8. Savina M, Cesne AL, Blay JY, Ray-Coquard I, Mir O, Toulmonde M, et al. Patterns of care and outcomes of patients with METAstatic soft tissue SARcoma in a real-life setting: the METASARC observational study. *BMC Med* 2017;15:78.
9. Grimer R, Judson I, Peake D, Seddon B. Guidelines for the management of soft tissue sarcomas. *Sarcoma* 2010;2010:506182.
10. Farnell MB, Aranha GV, Nimura Y, Michelassi F. The role of extended lymphadenectomy for adenocarcinoma of the head of the pancreas: strength of the evidence. *J Gastrointest Surg* 2008;12:651-6.
11. Bjorkman P, Kantonen I, Blomqvist C, Venermo M, Alback A. En bloc resection of visceral aorta and right kidney due to aortic sarcoma using temporary extracorporeal bypass grafting. *J Vasc Surg Cases Innov Tech* 2019;5:589-92.

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Supplementary Table. Previous report about prognosis of sarcoma

Series	Objects	Number of patients	Median follow-up (months)	5-year overall survival (%)	5-year disease-specific survival (%)
Schwarzbach et al ¹	Patients who underwent resection for peritoneal sarcoma involving major blood vessels	25	19.3	66.7	—
Poultides et al ⁵	Patients who underwent surgical resection for sarcoma	150	28	52	—
Stojadinovic et al ⁶	Patients having at least one local recurrence after complete resection	239	82		57.8-79.2
Savina et al ⁸	Patients with metastatic soft tissue sarcoma	2165	61	43.9	—