

Two-stage combined treatment of leiomyosarcoma of iliac vein using robotic surgery

Stanislav Valerevich Berelavichus, PhD,^a Andrey Evgenevich Zotikov, PhD,^b Andrey Germanovich Kriger, PhD,^a Vladimir Igorevich Panteleev, MD,^a and Ayrat Radikovich Kaldarov, MD,^a *Moscow, Russian Federation*

ABSTRACT

Vascular leiomyosarcoma is a rare tumor originating from the vascular smooth muscle cells. Leiomyosarcomas of the external iliac vein are extremely rare. Only single observations of this tumor are reported. We describe a 48-year-old woman who underwent a two-step combined treatment: robot-assisted removal of the extraperitoneal tumor and removal of the lower part of the tumor from the open femoral approach. There have been no signs of tumor recurrence during the 34-month follow-up period. (*J Vasc Surg Cases and Innovative Techniques* 2019;5:586-8.)

Keywords: Vascular leiomyosarcoma; Venous leiomyosarcoma; Iliac veins; External iliac vein; Robotic surgery

Vascular leiomyosarcoma, an uncommon retroperitoneal mesodermal tumor that originates from the vascular smooth muscle cells, constitutes about 5% of all adult sarcomas.¹ Only a few observations of external iliac vein leiomyosarcomas are described in the world literature. We present a case of two-step combined treatment of this tumor using robotic technologies. The patient's consent was obtained for publication.

CASE REPORT

The patient, a 48-year-old woman, was admitted to our clinic with right-sided lower back pain, pain in the right ilioinguinal area, and edema of the right leg that she suffered for the past few months. Outpatient examination revealed a retroperitoneal tumor, with thrombosis of the external iliac vein from the bifurcation of the common iliac to the femoral vein. Evaluation revealed enlargement of the right lower limb (the diameter at the thigh was 52 cm vs 50 cm of the left leg), without apparent edema. The tumor was not palpable. Peripheral pulsation was preserved and adequate on both lower extremities. The functionality of the right leg was not reduced. Her past medical history was unremarkable.

Computed tomography showed the extraperitoneal mass (60 × 45 mm) in the right inguinal-iliac region, which looked

like lymphosarcoma, and thrombosis of the external iliac vein (Fig 1). Duplex ultrasound revealed an extraperitoneal tumor, with occlusive thrombosis of the external iliac vein and partial thrombosis of the common femoral vein.

The patient received enoxaparin sodium at a dosage of 0.8 mL 12 hours before surgery and within 10 days after surgery.

A two-step procedure was performed. The first step was robot-assisted removal of the extraperitoneal tumor. Intraoperatively, in the area of the right iliac vessels, a circular mass of 6 cm was defined, covered with tumor-free peritoneum. The right external iliac artery was mobilized using scissors and bipolar grasper at a distance of 6 to 8 cm proximal to the upper pole of the mass and taken on a garrot. The external iliac vein was in a collapsed state; the blood flow in it could not be tracked. It was then isolated to the level of its origin. During mobilization of the tumor, involvement of the external iliac vein was revealed in its posterior wall. Blood flow was absent in the proximal segment of the vein. Taking these two facts into account, we made the decision to transect the vein. It was clipped and transected at a distance of 5 cm from the bifurcation. Mobilization of the tumor's posterior wall to its lower pole was performed. The external iliac artery was completely separated from the tumor's capsule. As the external iliac vein along the entire length was inside the tumor to its lower pole, its distal segment was isolated circularly 3 cm above the Poupart ligament. The vein in this area had signs of intravascular thrombosis. The distal external iliac vein directly above the ostium of the femoral canal was sutured and ligated. The internal iliac vein was clipped; the distal vein was transected 1 cm from the lower pole of the tumor. The vessel was full of thrombi. The complex was fully mobilized en masse and removed (Fig 2).

The second step of surgical treatment was performed—phlebectomy of the great saphenous vein and resection of the common femoral vein. The femoral vein was separated starting from the ostium (diameter, 8-9 mm); the vein was soft at the level of the great saphenous vein inflow. It was immediately crossed above the inflow, ligated, and sutured. Mobilization of the femoral vein was continued in the distal direction. A blood

From the Department of Abdominal Surgery,^a and Department of Vascular Surgery,^b A.V. Vishnevsky Centre of Surgery.

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Correspondence: Ayrat Radikovich Kaldarov, MD, Department of Abdominal Surgery, A.V. Vishnevsky Centre of Surgery, 27, B. Serpuhovskaja, Moscow 117997, Russian Federation (e-mail: ayratikus@gmail.com; kaldarov@ixv.ru).

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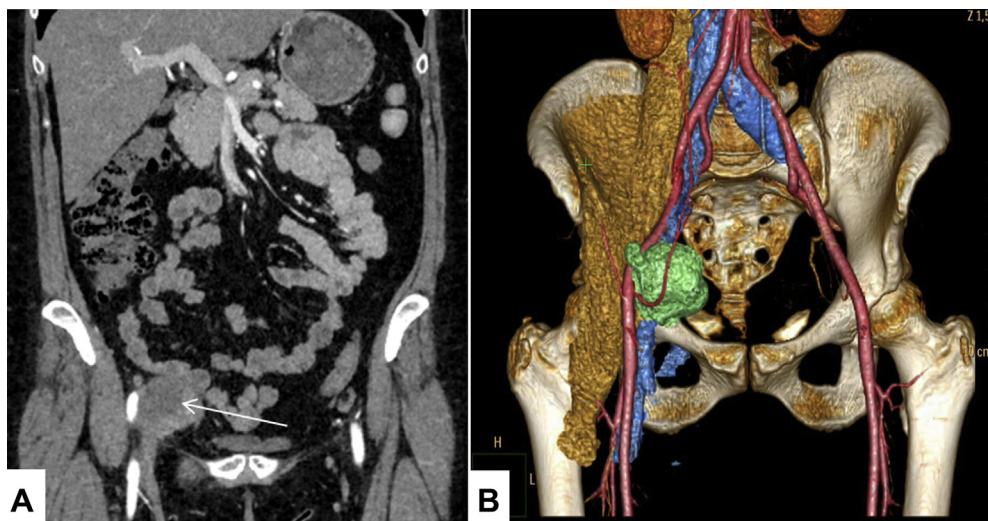


Fig 1. Contrast-enhanced computed tomography of the abdominal cavity and small pelvis. **A**, Arterial phase, frontal scan, pelvic leiomyosarcoma (arrow). The iliac artery passes along the tumor contour; the iliac vein is involved in the tumor. **B**, Three-dimensional reconstruction.

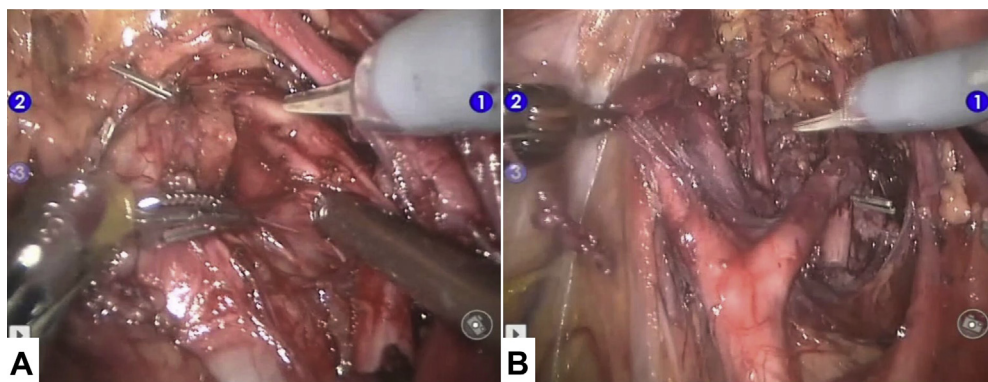


Fig 2. Intraoperative photographs of the robot-assisted step. **A**, Mobilization of the neurovascular bundle. **B**, Appearance of the operative field after tumor removal. 1, The right robotic hand with the scissors; 2, the left robotic hand with the bipolar.

clot that occluded the vessel was determined by palpation 2 cm lower. A significant fibrous process was noted around the vein. The inguinal ligament was dissected. Mobilization of the common femoral vein was continued after its transition to the external iliac vein, which was separated over a length of 3 cm to the clip that was placed earlier. The specimen was removed (Fig 2). The frozen section showed negative margins.

Histologic examination showed that it was a grade 3 leiomyosarcoma (5.7 × 4.3 × 3.5 cm) originating from the right external iliac vein. In the vein's wall were extensive foci of fibrosis, inflammatory infiltrates, accumulations of hemosiderophages, and focal growths of tumor (Fig 3).

Postoperative recovery was uneventful. The patient was discharged from our clinic on day 14 after surgery. There were no signs of edema of the lower limb. Chemotherapy was not performed. At follow-up, neither computed tomography nor magnetic resonance imaging showed any signs of tumor recurrence. Long-term follow-up reached 34 months at the time of the writing of this article.

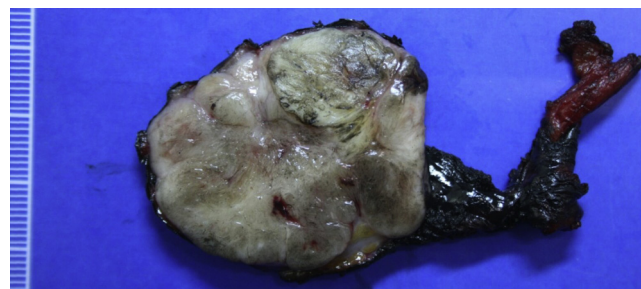


Fig 3. The specimen. Leiomyosarcoma arises from the iliac vein; blood thrombus in the lumen of the vein.

DISCUSSION

Leiomyosarcoma of the major veins is a rare extraperitoneal tumor that originates from vascular smooth muscle cells.^{2,3} In the world literature, about 400 observations of patients with leiomyosarcoma of the inferior vena cava

have been published.² The earliest reference dates to 1871, when Perl⁴ described sarcoma of the inferior vena cava that was found at autopsy; the first surgical treatment was performed in 1928 by Melchior.⁵

The tumor most frequently arises from the inferior vena cava; rarely, it develops from the great saphenous, femoral, or internal jugular vein. Leiomyosarcoma of the iliac vein is rare.³ There are single observations of such patients described.

By growth pattern, leiomyosarcomas are divided into intraluminal, extraluminal (the most common variant, representing 62%), and mixed forms.⁶ Depending on the growth pattern, size, location, and degree of circulatory disorder, clinical symptoms can be variable.^{6,7} Edema of the limb is observed in one-third of patients. Most often, patients are suffering from pain of various intensity. The tumor has a slow growth rate, and therefore early diagnosis is often difficult. Metastases to the liver and lungs are found in up to 50% of cases.¹

In our case, the tumor had mixed growth, so there were both vascular and somatic manifestations of disease. The main methods of diagnosis of leiomyosarcoma are radiologic: computed tomography and magnetic resonance imaging with intravenous administration of contrast material. Using these methods, we not only can define the location and size of the mass but also can estimate the extent of the intended surgical intervention.⁶

Presently, there are no guidelines for treatment of leiomyosarcoma of the major veins. Radical surgical resection with negative margins remains the main option. In the described case, we tried to perform a completely robotic removal of the pelvic tumor with preservation of the neurovascular bundle with minimal tissue trauma. During separation of the tumor, the iliac artery, its branches, and the femoral nerve were successfully isolated. However, after final mobilization of the complex and transection of the iliac vein above and below the tumor, we found it impossible to perform a radical resection with the transabdominal robotic approach. Therefore, the second step of the surgical treatment, phlebectomy, was performed through the femoral approach. As a result, an adequate volume of tumor resection within disease-free tissue was achieved.

In the world literature, we did not find any such cases of robotic surgical treatment of leiomyosarcomas or

descriptions of any combined method for removing the mass from two approaches. This case is also rare because of the "difficult" location of the tumor in the limited anatomic space. The robotic technique has advantages when precise movements of vessel and nerve mobilization are necessary in limited visibility and with limited movement of the surgeon's hands.

The treatment success is demonstrated by the disease-free survival, which is 34 months, without recurrence of the disease and without the use of chemotherapy or radiation therapy.

CONCLUSIONS

Leiomyosarcoma of the external iliac vein is an extremely rare tumor. Tumor at this location is mostly reported by single clinical observations. For the first time, a combined method of treatment was used: the first step, robot-assisted removal of the tumor; and the second step, removal of the vessel's lower part from the femoral approach with a partial dissection of the Poupart ligament. The use of robotic technology can improve the outcome of treatment of patients with this pathologic process.

REFERENCES

1. Roland CL, Boland GM, Demicco EG, Lusby K, Ingram D, May CD, et al. Clinical observations and molecular variables of primary vascular leiomyosarcoma. *JAMA Surg* 2016;151:347-54.
2. Schettini AV, Coubeau L, Mazzeo F, Lerut J. Leiomyosarcoma of the inferior vena cava level II including both renal veins: surgical approach. *Acta Chir Belg* 2015;115:96-8.
3. Sahu R, Aggarwal R, Kumar P, Mittal S, Sood V, Sarin J. Primary external iliac vein leiomyosarcoma. *Ann Vasc Surg* 2019;57:274.e5-9.
4. Perl L. Ein Fall der Sarkom der Vena cava inferior. *Virchows Arch Pathol Anat* 1871;53:378.
5. Melchior E. Sarkom der Vena cava inferior. *Dtsch Z Chir* 1928;213:135-40.
6. Marko J, Wolfman DJ. Retroperitoneal leiomyosarcoma from the radiologic pathology archives. *Radiographics* 2018;38:1403-20.
7. Yamamoto A, Nakata W, Yamamichi G, Tsujimura G, Tsujimoto Y, Nin M, et al. A rare case of vascular leiomyosarcoma originating from a branch vessel of the external iliac vein. *Case Rep Med* 2018;2018:5160591.

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