Spontaneous iliac vein rupture: An uncommon, but frequently lethal, event

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ABSTRACT
Spontaneous rupture of the iliac veins is a distinctly uncommon problem often misdiagnosed as an arterial rupture because of significant retroperitoneal bleeding. It often occurs with acute left-sided deep vein thrombosis and physical activities that exacerbate acute venous hypertension. A significant number of these patients will have anatomy associated with May-Thurner syndrome. Delayed imaging on computed tomography scanning might suggest a venous etiology for a retroperitoneal hematoma rather than arterial bleeding. We found 53 previously reported cases of iliac vein rupture Our report details two additional cases and the treatment options and outcomes. (J Vasc Surg Cases Innov Tech 2021;7:558-62.)

Keywords: Acute venous hypertension; Iliac vein rupture

Rupture of an iliac vein was first reported in 1961 by Hosse.

The presence of acute deep vein thrombosis (DVT) of the iliofemoral veins that results in acute venous hypertension frequently precedes iliac vein rupture.

Spontaneous rupture of the iliac veins will often be precipitated by physical activity. The vast majority of cases will involve the left iliofemoral venous system and occur primarily in women, with a significant number of patients having anatomy associated with May-Thurner syndrome.

Phlegmasia cerulea dolens from an extensive iliofemoral DVT has also been reported with iliac vein rupture, as have pulmonary emboli. We report two cases of spontaneous iliac vein rupture. We also reviewed treatment options. The patients provided written informed consent for the report of their case details and images.

CASE REPORT
Patient 1. A 76-year-old man had been admitted to an outside facility on November 27, 2020 after he was found unresponsive after exercising on a treadmill. On examination, he had a nontender lower abdominal mass and was hypotensive. An electrocardiogram suggested a myocardial infarction. His hemoglobin was 9.9 g/dL, and his troponin levels were elevated.

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He underwent cardiac catheterization from the right groin, which demonstrated severe stenosis of the right coronary artery.

After cardiac catheterization, his blood pressure had decreased into the 70s, and his hemoglobin had decreased to 6 g/dL. Thus, 4 U of blood were administered. A computed tomography (CT) angiogram of the abdomen subsequently demonstrated extensive inferior vena cava (IVC) thrombus extending up to level of the renal veins (Fig 1, A). An extensive left iliofemoral DVT was noted with occlusion of the left common iliac vein (Fig 1, B). In addition, a large retroperitoneal hematoma was found in the pelvis. Delayed images on the CT angiogram demonstrated contrast extravasation from the left iliac veins (Fig 2).

The patient was taken to the operating room. A suprarenal inferior vena cava filter (Optease Elite; Cordis, Hialeah, Fla) was inserted through the right common femoral vein because of the extensive thrombus within the IVC. Venography through the left common femoral vein confirmed occlusion of the common iliac vein and extravasation of contrast from the left external iliac vein with extensive thrombus (Fig 3, A). Three Amplatz plugs (12 mm, 14 mm, and 20 mm; Abbott Vascular, Abbott Park, Ill) and two 20 × 30 Azur coils (Terumo, Somerset, NJ) were placed in the left external iliac vein. Subsequently, minimal bleeding from the vein was observed (Fig 3, B). The point of bleeding was in the external iliac vein, which we judged to have a 14-mm diameter. The currently available covered stents can achieve this diameter only with significant overdilatation. We were also concerned that achieving apposition and control of the bleeding using covered stents would not have been possible in the occluded common iliac vein.

A Perclose device (Abbott Vascular) was placed in the left common femoral vein because of acute venous hypertension within the common femoral vein. Postoperatively, heparin was administered, which was then transitioned to apixaban (Eliquis; Bristol-Myers Squibb, New York, NY). His IVC filter was removed 3 weeks later. He experienced only mild left lower extremity edema during follow-up.

Patient 2. A 77-year-old man had undergone colon resection in 2017 to treat acute diverticulitis. Postoperatively, he developed an acute DVT of the left lower extremity and received anticoagulation therapy. The thrombus extended from the
common femoral vein into the femoral vein. He later developed an incisional hernia. In August 2020, he underwent a CT scan of the abdomen before undergoing repair of the incisional hernia. The CT scan demonstrated atretic left common and left external iliac veins, with collateral veins along the pelvic sidewall believed to be secondary to May-Thurner syndrome. He was asymptomatic, and it was thought that recanalization of the iliac veins was not feasible.

Approximately 3 years later, he underwent hernia repair without difficulty. However, he presented to the emergency room 2 days later with an acutely swollen left lower extremity and left lower quadrant pain. Venous duplex ultrasound imaging demonstrated extensive thrombus in the left common femoral vein, femoral vein, and popliteal and tibial veins. His hemoglobin was 6.6 g/dL. A CT scan with intravenous contrast demonstrated a 20 × 30 × 13-cm hematoma on the left side
of the pelvis with hyperdense material within the hematoma, suggestive of active bleeding. An arteriogram demonstrated no active bleeding. Because of the contraindications for anticoagulation, an Optease Elite IVC filter (Cordis) was placed through the right femoral vein.

He remained hemodynamically stable for 2 days. However, he subsequently became hypotensive, with a further decrease in hemoglobin. A vascular surgery consultation was requested. He received another 2 U of blood and was taken to the operating room. At exploration, a large retroperitoneal hematoma was encountered with brisk venous bleeding from a branch of the left external iliac vein. This was controlled by oversewing the vein. Enoxaparin (Lovenox, Sanofi-Aventis, Paris, France) was started, and he was subsequently transitioned to apixaban (Eliquis, Bristol-Myers Squibb). On December 8, 2020, he underwent removal of the IVC filter. He continued to take apixaban.

**DISCUSSION**

Spontaneous rupture of the iliac veins is uncommon. We found a total of 53 cases, which were mostly from isolated case reports, using a Google search. Jiang et al reported a series of nine patients, although for four of these patients, no imaging studies or operative findings to confirm iliac vein rupture were reported. Of these 53 cases, 14 patients died, for a mortality rate of 26.4%.

Diagnostic findings can be present that can suggest an iliac vein rupture. These include lower abdominal pain or a mass due to retroperitoneal bleeding, lower extremity pain and swelling secondary to an acute DVT, occurring predominantly on the left side, and a distinct predilection for occurring in women. Pulmonary emboli can also occur. Often, a precipitating event will have occurred that could exacerbate the acute venous hypertension, such as bending, coughing, vomiting, childbirth, and so forth.

The predominance of iliac vein ruptures will occur on the left side, suggesting an association with May-Thurner syndrome. Hosn et al reported that of the 48 cases with iliac vein rupture they reviewed, 85% of the patients were women, 79% had had DVT, and 94% had had the iliac vein rupture on the left side.

The etiology of spontaneous iliac vein rupture is likely multifactorial. Mechanical factors such as compression of the left iliac vein as seen with the anatomy present with May-Thurner syndrome is likely a contributing factor, because a significant number of patients will have underlying May-Thurner syndrome. Also, a preponderance of cases have occurred on the left side and in women. Other potential etiologic factors include vein wall inflammation from underlying thrombus, a sudden increase in intra-abdominal pressure (Valsalva maneuver), and, in women, hormonal factors such as the loss of the vessel protective effects of estrogen after menopause.

CT scanning with contrast is the most common imaging study used to evaluate these patients. Magnetic resonance imaging with gadolinium has also been reported. Venography could demonstrate extravasation of the contrast material, as was seen in one of our patients.

No uniformity of opinion has been reached regarding the treatment of iliac vein rupture because the presentation can range from a hemodynamically stable patient, for whom conservative treatment (eg, anticoagulation...
therapy and stenting of any underlying venous compression) is appropriate, to a patient presenting with fatal retroperitoneal hemorrhage. Some patients have undergone surgical exploration owing to the belief that the source of bleeding was either a gynecologic issue or a ruptured artery.\textsuperscript{10,11} In such patients, ligation or primary repair of the vein was usually performed.

If feasible, the use of covered stents represents an excellent option, although some patients could also require decompressive celiotomy if evidence of compartment syndrome is found. Any underlying May-Thurner syndrome should be treated. A retrievable IVC filter should be placed if the patient has a contraindication to anticoagulation therapy. Anticoagulation treatment should be

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**Fig 3. A,** Venogram demonstrating thrombus within the left external iliac vein (asterisk) with extravasation of contrast. **B,** Venogram after placement of Amplatzer plugs and coils.
instituted and continued for 6 months postoperatively. Patients with phlegmasia might require venous thrombectomy or Palma-Dale bypass. Hemodynamically stable patients can be treated conservatively.

The treatment procedures for the 53 reported cases we found are summarized in Table. Some of these patients had undergone combined treatment modalities such as conservative treatment of the retroperitoneal hematoma, placement of an IVC filter, and stenting of the iliac veins in the presence of May-Thurner syndrome.

No consensus has been reached for treatment other than to control the bleeding; treat any underlying May-Thurner syndrome, if present; place a temporary IVC filter (if anticoagulation therapy is contraindicated); perform venous thrombectomy or Palma-Dale bypass if phlegmasia has occurred; and prescribe anticoagulation therapy for ≥6 months. Hemodynamically stable patients can be treated conservatively.

CONCLUSIONS

Spontaneous iliac vein rupture is a very rare event associated with high mortality. Accordingly, a high index of suspicion is required for a correct and timely diagnosis. Diagnostic clues include contrast extravasation on delayed imaging on CT scans, the presence of a concomitant acute DVT, female sex, left-sided predominance, and a precipitating event that increased the intra-abdominal venous pressure. A variety of treatment options exist, with the specific treatment in large part determined by the hemodynamic status of the patient.

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