

# Spontaneous retroperitoneal hematoma associated with iliac vein rupture

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**Objective:** Spontaneous retroperitoneal hematoma (SRH) associated with iliac vein rupture is a rare but life-threatening emergency with high operative mortality. This study summarizes our experience in providing diagnostic and therapeutic management for this rare clinical entity.

**Methods:** Between May 2002 and May 2009, nine patients were admitted to our hospital for SRH and acute deep venous thrombosis (DVT). Medical data for demographics, clinical presentation, auxiliary examinations, treatment modalities, outcomes, and follow-up were retrospectively analyzed.

**Results:** Nine patients (8 women, 1 man) were enrolled in this study. All were aged >45 years (range, 46-70 years). The common clinical manifestations were sudden onset of left lower abdominal or lumbar pain, swelling of the left lower extremity, anemia, and hypotension. Most patients were diagnosed by duplex ultrasound imaging and computed tomography scan. Three patients were treated conservatively, and six underwent surgical or combined treatments, comprising 2 repairs of iliac vein, 1 iliac vein ligation and Palma-Dale bypass graft, 1 pelvic vein ligation, 1 removal of hematoma, and 1 repair of iliac vein, thrombectomy, and endovascular stent placement. The iliac vein ruptured in five patients. May-Thurner syndrome was found in three patients. One patient died after surgery (operative mortality, 16.7%). Postoperative morbidity was 50%. Mean volume of perioperative blood transfusion was  $900 \pm 640$  mL (range, 0-2000 mL). Mean lengths of stay were  $2.7 \pm 1.4$  days (range, 2-5 days) in the intensive care unit and  $16.9 \pm 2.4$  days (range, 14-21 days) in the hospital. Eight patients were postoperatively treated with 6 months of warfarin. Mean follow-up was  $30.5 \pm 15.0$  months (range, 6-50 months). The occurrence rate of chronic venous insufficiency was 87.5% during follow-up.

**Conclusions:** SRH with concomitant DVT, especially in women aged >45, should be considered in patients with sudden lower abdominal or lumbar pain, leg swelling, anemia, and shock. Spontaneous iliac vein rupture and the presence of May-Thurner syndrome should be considered in these patients. Surgical interventions were associated with high mortality and morbidity. In our experience, conservative therapy was safer than open surgical procedures. (J Vasc Surg 2010;52:1278-82.)

Spontaneous retroperitoneal hematoma (SRH) is a rare entity. Renal tumor rupture is regarded as a relatively common cause.<sup>1</sup> Spontaneous iliac vein rupture is an uncommon cause of SRH. To date, 35 patients with spontaneous iliac vein rupture have been reported,<sup>2-4</sup> of which almost 79% had concomitant deep venous thrombosis (DVT) or thrombophlebitis<sup>2</sup> and approximately 28.6% had May-Thurner syndrome.<sup>3,5-13</sup> In this study, we investigated SRH associated with iliac vein rupture. Owing to the inability to institute anticoagulation and initial uncertainty in diagnosis, management of these patients presents significant challenges. Most reported patients have been treated surgically, with a survival rate of 71%.<sup>2</sup> In this report, we describe our diagnostic and therapeutic experience in nine patients. Almost all of the previous reports studied one or two patients,<sup>14</sup> and we believe this is the largest single-center case series to date.

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## METHODS

**Patients.** A retrospective analysis was done on nine patients with SRH who were treated in our hospital from May 2002 to May 2009. These patients were not receiving anticoagulation at presentation. Patients with SRH that resulted from iliac vein rupture were included. Although no surgical or imaging evidence could confirm the diagnosis of iliac vein rupture in four patients with SRH, their clinical manifestations were highly suggestive of iliac vein rupture. Clinical characteristics in these patients were similar to those of spontaneous iliac vein rupture; therefore, they were also included.

The study excluded patients with retroperitoneal hematoma secondary to trauma, operations, coagulation disorders, arterial aneurysms, cardiovascular intervention, severe portal hypertension, or tumors. Also excluded were patients with retroperitoneal hematoma secondary to anti-thrombotic therapy. Patients with a single SRH were not admitted to our department. Medical data for patient demographics, clinical presentation, auxiliary examinations, management, clinical outcomes, and follow-up were retrospectively analyzed.

**Diagnosis.** Acute DVT was diagnosed by patient history, clinical symptoms and signs, and imaging results. Imaging examinations included color duplex ultrasound (CDU) scans, venography, and computed tomography

(CT) scans. Diagnosis of SRH was made by patient history, clinical presentation, laboratory tests, and imaging examinations. CDU or CT imaging was used. Hemorrhage was monitored according to patients' symptoms, vital signs, and results of blood routine tests.

### Therapeutic management

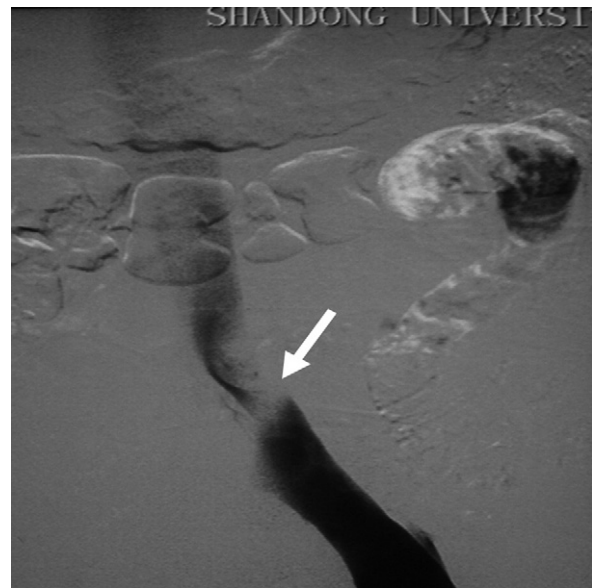
**Main principle.** Hemodynamic stability was the key judgement. If hemodynamic variables were stable after resuscitation, the patient was treated conservatively. If there were evidences of further hemorrhage or blue phlegmasia with impending venous gangrene, a surgical procedure was compelled.

**Conservative treatment.** All patients received prompt resuscitation once hypovolemic shock or retroperitoneal hemorrhage was identified. Colloidal solution, crystalloid solution, and blood products were used. Patients were confined to bed rest, and swollen limbs were elevated. Vital signs were observed by electrocardiographic monitoring for several days.

**Anticoagulation therapy.** Anticoagulation was usually given 24 hours after hemodynamic stability. In surgical patients, anticoagulation therapy was usually started 24 hours after surgery. In nonsurgical patients, anticoagulation was performed 24 to 48 hours after admission. Subcutaneous administration of low-molecular-weight heparin was used initially and substituted later by warfarin.

**Filter placement.** Indications for vena cava filter placement contain a contraindication to anticoagulation and compelled surgery. In surgical patients, filters were placed during the operation after the bleeding was controlled. In nonsurgical patients, filters were implanted after SRH and acute DVT were confirmed. Retrievable inferior vena cava filters (Aegisy Filter; Xianjian Technology, Shenzhen, China) were used to prevent pulmonary embolism. Retrievable filters were deployed at the second and third lumbar level from the right groin under fluoroscopic guidance and removed  $\leq 2$  weeks.

**Management of ruptured veins.** Emergency explorative laparotomy was performed under general anesthesia through an abdominal incision. After the retroperitoneal cavity was opened, hematoma was evacuated. The abdominal aorta was exposed for possible cross-clamping, although it was not blocked in the study. The location of bleeding was usually searched for from top to bottom. Retroperitoneal visceral organs were also explored one by one. When iliac vein rupture was highly suspected, the pelvic cavity was explored first. After the bleeding location was found, the tear was immediately oppressed. Proximal and distal ends of ruptured vein were exposed, and vascular clamps were used to isolate the tear later. If it was difficult to stop bleeding, the ruptured vein could be ligated. Palma-Dale bypass grafting procedure (crossover saphenous vein bypass grafting) and arteriovenous fistula formation could be performed after the left common iliac vein was ligated. Temporary arteriovenous fistulas were repaired 3 months after initial surgery.



**Fig 1.** Intraoperative venogram shows the left common iliac vein (*arrow*) is compressed by the right common iliac artery.

Fresh thrombus found to be occluding the lumen of the common or external iliac vein was removed with a Fogarty arterial embolectomy catheter through the venous laceration after bleeding was controlled. Then the ruptured vein was repaired. A drainage tube was placed and the wound was closed.

**Management of May-Thurner syndrome.** May-Thurner syndrome that was found intraoperatively was managed according to the patient's condition. If the patient was hemodynamically stable, endovascular stent implantation or surgical procedures were performed. Another skin incision was made in the left groin region. The distal thrombus of the lower limb was removed with compression by an Esmarch bandage. A Fogarty catheter was used to remove proximal thrombus. A 9F sheath was inserted through the venotomy site. Venography demonstrated left iliac vein compression with abundant collateral pathways (Fig 1 and Fig 2, A). A 14- × 60-mm stent (Bard Luminexx 3 Vascular Stent, CR Bard Inc, Murray Hill, NJ) was placed. Completion angiography showed luminal patency (Fig 2, B). The venotomy site was sutured and the wound was closed.

**Follow-up.** Patients were continuously anticoagulated with warfarin for 6 months after hospital discharge, with a target international normalized ratio of 2.0 to 3.0. The patient who underwent endovascular stent implantation was maintained on lifelong aspirin for stent patency. Patients were followed-up by CDU imaging<sup>15</sup> at 1, 3, and 6 months and then annually to evaluate patency and valvular function in the left lower limbs. Recurrent edema of the left lower extremity was usually managed by conservative treatment, including elevation, an elastic stocking, and venoactive drugs.



**Fig 2.** A, The venogram before treatment reveals the proximal portion of the left common iliac vein was obstructed. Abundant collateral pathways were formed by pericyclic plexus to the right iliac vein and via lumbar plexus to inferior vena cava. B, The completion venogram shows restoration of venous patency and significant reduction of collateral vessels.

## RESULTS

### Patient demographics and clinical manifestations.

Nine patients were enrolled in our study. All patients but one were women (88.9%). All patients were >45 years old, with a mean age of  $55.1 \pm 8.8$  years (range, 46-70 years). Two patients had recently received hormone replacement therapy. One patient suffered from chronic glomerulonephritis. One patient had been diagnosed with asthma for

several years. One patient had a history of varicosity in the left lower extremity. One patient complained of edema of the left lower limb after standing for a long time. Mean time from onset of symptoms to presentation to the hospital was  $34.6 \pm 41.6$  hours (range, 2-140 hours). An activity preceding the onset of the first symptom, which may increase venous pressure, was noted in each patient, including defecation, bending, and walking. All patients complained of left lumbar or lower abdominal pain, or both, and had left lower limb symptoms of swelling and pain. Signs of anemia were identified in all patients, including paleness of skin and mucosa, tachycardia, and shortness of breath. Hypotension (blood pressure  $\leq 90/60$  mm Hg) was detected in seven patients (77.8%). Sudden onset of syncope occurred in three patients (33.3%).

**Auxiliary examinations and diagnosis.** Laboratory tests on admission revealed mean hemoglobin was  $7.2 \pm 1.6$  g/dL (range, 5.0-9.8 g/dL) and mean hematocrit was  $22.9\% \pm 3.3\%$  (range, 18.6%-29.2%). Spontaneous iliac vein rupture was suspected in three patients after angiography, but no bleeding site was found during venography. Acute DVT of the left lower extremity was confirmed in all patients by CDU imaging ( $n = 5$ ), venography ( $n = 3$ ), or CT ( $n = 1$ ). Iliofemoral DVT was found in six patients, and extensive DVT from the iliofemoral vein to the venous plexus of the calf muscle was noted in three patients. Retroperitoneal hematoma was diagnosed in seven patients by CT ( $n = 7$ ) or CDU imaging ( $n = 2$ ), and all were located on the left side. Two patients (22%) were misdiagnosed. One patient was thought to have a rupture of an abdominal aortic aneurysm because the abdomen swelled significantly shortly after venography was started. She was immediately taken to the operating room. The other patient was originally thought to have a retroperitoneal tumor from the CT images.

**Methods and results of treatment.** Five patients underwent open surgical procedures. Three patients were treated conservatively. One patient underwent combined surgical and endovascular stent treatment. Rupture of the left iliac vein occurred in five patients (55.6%), comprising three common, one external, and one pelvic vein, which was a branch of the left internal iliac vein. The bleeding location was not discovered in one patient, but we presumed thrombus might have obstructed the bleeding site. An anterior longitudinal tear was identified in four patients, and a transverse tear was discovered in the pelvic vein. The length of the venous tears ranged from 2 to 4 cm. There was obvious bleeding at the site of rupture in four patients. Bleeding was not evident in one patient, and a thrombus in the venous lumen was found and removed.

The iliac vein was repaired in three patients. Iliac vein ligation and Palma-Dale bypass graft procedure were performed in one patient. Pelvic vein ligation was performed in one patient. May-Thurner syndrome was identified in three patients (33.3%). It was found unexpectedly during the exploration, and one patient underwent endovascular stent placement after thrombectomy. Only two patients underwent placement of an inferior vena cava filter because of



unavailability of expertise at the early stage and economics factor. The mean volume of perioperative blood transfusion was  $900 \pm 640$  mL (range, 0-2000 mL).

One patient died suddenly 24 hours after the operation, for an overall mortality of 11.1% and an operative mortality of 16.7%. An autopsy was not performed. Postoperative morbidity was 50%. Limb swelling increased after surgery in two patients, and thrombophlebitis occurred in one patient. These patients were treated conservatively. The other five patients recovered uneventfully. The mean intensive care unit length of stay was  $2.7 \pm 1.4$  days (range, 2-5 days). The eight patients who survived were discharged with a mean hospital length of stay of  $16.9 \pm 2.4$  days (range, 14-21 days).

**Follow-up.** The patients who survived to discharge were alive with a mean follow-up of  $30.5 \pm 15.0$  months (range, 6-50 months). Seven patients (87.5%) complained of recurrent swelling of the left lower limb. Chronic venous insufficiency was demonstrated in these patients by CDU imaging. Relief of leg swelling could usually be achieved by conservative treatment. No discomfort was complained of in the last patient during 6 months of follow-up. Follow-up CDU imaging showed the stent and deep veins of the left lower limb were patent. The patient has been undergoing close follow-up.

## DISCUSSION

SRH associated with iliac vein rupture is a rare but life-threatening emergency that usually occurs in middle-aged or elderly women.<sup>5</sup> Most patients in our study were women aged >45 years. Activities that can increase pelvic venous pressure, such as bending, defecation, and climbing, were noted in current and previous studies.<sup>16</sup> Common clinical characteristics included sudden onset of left lower abdominal or lumbar pain, swelling of the left lower limb, anemia, and shock.

SRH was often concomitant with DVT and May-Thurner syndrome in patients with iliac vein rupture. Previous reports indicate almost 79% of patients had DVT or thrombophlebitis, and approximately 28.6% were associated with May-Thurner syndrome; in our study, this was 100% and 33.3%, respectively. Why did SRH and DVT always occur together in these patients? From patients' clinical manifestations, it is difficult to distinguish which occurred first. Several researchers have speculated that DVT may be a cause of iliac vein rupture,<sup>17,18</sup> but the authors considered that DVT may be a complication of iliac vein rupture.<sup>19</sup> DVT may result from a combination of hypovolemia, local trauma with loss of endothelial integrity, compression by the hematoma, and May-Thurner syndrome.

The cause of spontaneous iliac vein rupture remains unclear. Activities that can increase venous pressure abruptly were often noted in these patients,<sup>16</sup> so a sudden increase in venous pressure was regarded as a potential cause.<sup>5</sup> The age-related and female preponderance showed loss of estrogen effect and complications of pregnancy may be related to this condition<sup>17,19</sup> because they would de-

crease distensibility of veins.<sup>20</sup> Given left-side preponderance and coincident presence, May-Thurner syndrome may be a factor in this entity.<sup>8</sup> Venous hypertension produced by proximal obstruction can trigger inflammatory cascades and decrease contractility.<sup>21,22</sup> The mechanism of iliac vein rupture may be a sudden rise in venous pressure in weakened vein due to loss of estrogen, complications of pregnancy, and May-Thurner syndrome.<sup>7</sup>

Accurate preoperative diagnosis of SRH was usually difficult. Hypovolemic shock often occurred at presentation, and extensive tests were impossible. In the past, numerous patients were not correctly diagnosed before surgery.<sup>5,16,23</sup> Most patients have been diagnosed by surgery, and only a few were diagnosed by venography.<sup>12,19</sup> May-Thurner syndrome was diagnosed preoperatively in one patient.<sup>3</sup> In this report, two patients were misdiagnosed, one as rupture of abdominal aortic aneurysm and the other as a retroperitoneal tumor.

Several treatment methods have been reported, such as conservative therapy, open surgical procedures, endovascular interventions, and combined surgical and endovascular treatment. Most patients underwent open surgical procedures.<sup>5</sup> One patient was treated with conservative therapy,<sup>19</sup> and several received endovascular interventions.<sup>8,12</sup> One patient received combined surgical and endovascular stent treatment.<sup>3</sup>

Three (33.3%) of our nine patients were managed conservatively, and the others were treated by surgical (n = 5) or combined methods (n = 1). Active hemorrhage was the main indication for surgical intervention. Hemodynamic status was the key judgement. The observing indexes included patient's symptoms, vital signs, hemoglobin concentration, hematocrit, and urine volume, among others. If vital signs were not stable and the level of hemoglobin was decreasing continuously after resuscitation, a surgical procedure would be compelled. Endovascular stent graft placement may be performed when the bleeding site is identified by venography.<sup>12</sup>

Surgical intervention was associated with high mortality and morbidity in these patients. Operative mortality after surgery was 23.8% in the Cho et al<sup>19</sup> report, and survival after surgery was only 71% in the report by Tannous et al.<sup>2</sup> In the current study, the operative mortality rate was 16.7%, and postoperative morbidity reached 50%. Three patients were treated conservatively and recovered uneventfully. In our experience, conservative treatment was safer than open surgical procedures.

The long-term follow-up results of this rare entity are still unknown. The mean follow-up time in our study was  $30.5 \pm 15.0$  months. Chronic venous insufficiency was frequent during follow-up. The only asymptomatic patient in our study underwent combined surgical and endovascular treatment. Zieber et al<sup>12</sup> reported favorable outcomes using endovascular repair with stent grafts. In 2007, Kin et al<sup>3</sup> described a patient managed by surgery and placement of an endovascular stent, which was patent during an 8-month follow-up. Endovascular treatment may be helpful to improve follow-up results.

In view of the high incidence of DVT, anticoagulation is recommended after the bleeding has stopped. All patients in our study were anticoagulated after hemodynamic variables were stable. All of the patients with SRH and concomitant DVT had indications for vena cava filter placement. A retrievable inferior vena cava filter is recommended in this condition.

## CONCLUSION

In middle-aged or elderly women sustaining left-sided SRH and acute DVT, the possibility of spontaneous rupture of the iliac vein should be taken into account. The main clinical features include sudden lower abdominal or lumbar pain, swelling and pain of the left lower limb, anemia, and shock. Anticoagulation therapy is recommended 24 hours after bleeding has stopped. Open surgical procedures were associated with high mortality and morbidity. Conservative therapy seemed to be safer than open surgical procedures. Endovascular intervention may help to improve follow-up outcomes.

## AUTHOR CONTRIBUTIONS

Conception and design: JJ, XD, SH

Analysis and interpretation: JJ, XD, GZ, QS, ZW, SH

Data collection: JJ, XD, GZ, QS, ZW, SH

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Critical revision of the article: JJ, XD, GZ, QS, ZW, SH

Final approval of the article: JJ, XD, GZ, QS, ZW, SH

Statistical analysis: JJ, XD, SH

Obtained funding: Not applicable

Overall responsibility: SH

JJ and XD participated equally and share first authorship.

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