External iliac venous aneurysm in a pregnant woman: A case report

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We report an external iliac venous aneurysm in a young pregnant woman who was diagnosed incidentally by ultrasound scanning. The aneurysm was successfully treated by tangential aneurysmectomy and lateral venorrhaphy. Primary iliac venous aneurysm is a rare vascular abnormality. The clinical significance of the disease is unknown. However, embolism, rupture, and thrombosis might occur as they can occur with popliteal venous aneurysm. In fact, three of four reported patients with iliac venous aneurysms had a thromboembolic event. For those reasons, prophylactic treatment is indicated. This is the first patient with an iliac venous aneurysm to be diagnosed without complication. (J Vasc Surg 2004;40: 174-8.)

Primary venous aneurysm is a rare vascular abnormality that can occur throughout the venous system. Although venous aneurysms occur most commonly in the neck and central thoracic veins, they also have been reported in the visceral veins and the extremities. However, there are few reports of iliac venous aneurysms. Complications such as embolism, rupture, and thrombosis have been reported in cases of aneurysms of popliteal vein or other areas.[1-3] For those reasons, prophylactic therapy is indicated. We report an extremely rare case of external iliac venous aneurysm treated successfully by tangential aneurysmectomy and lateral venorrhaphy.

CASE REPORT

A 20-year-old pregnant woman underwent an endovaginal ultrasonogram in the 7th week of gestation, which incidentally revealed a pelvic mass with blood flow. She was asymptomatic and had no history of pelvic trauma or inflammatory disease. The patient was referred to our hospital. Endovaginal duplex ultrasonogram showed aneurysmal formation of the left external iliac vein. Because of the risk of rupture and because thrombosis might increase during pregnancy, the patient elected to undergo a therapeutic abortion. Abdominal computed tomogram showed an external iliac venous aneurysm, 8 cm in diameter, that compressed the bladder (Fig 1, *A*). An ascending phlebogram showed a solitary, saccular aneurysm of the external iliac vein (Fig 1, *B*). Surgery was recommended.

At the operation, a venous aneurysm was exposed through a left lower quadrant retroperitoneal incision and a vertical left femoral incision (Fig 2, *A*). After entering the retroperitoneal space, the aneurysm sac was easily dissected free from all surrounding tissue except the bladder. After systemic injection of heparin 3000 U, the left iliac vein was clamped proximally and distally, and the aneurysm was opened. No mural thrombus was seen within the aneurysm, and the aneurysmal wall was thin and smooth (Fig 2, *B*). A vascular clamp was placed tangentially across the transitional area, and the aneurysm was resected flush with the vascular clamp. After unclamping, a lateral venorrhaphy was created with a continuous 5-0 polypropylene suture with four stay sutures. The length of the suture line was about 6 cm (Fig 3, *A*).

The patient received anticoagulation therapy postoperatively consisting of 5 days of heparin and 1 year of oral warfarin. Intermittent pneumatic compression was used for 1 day and elastic stockings for 2 weeks after surgery. The postoperative course was uneventful, without dependent edema of the legs. Pathologic examination showed a true aneurysm without inflammation.

The external iliac vein was patent without any dilatation on ascending phlebogram 1 week after operation (Fig 3, *B*). An enhanced computed tomography scan also showed no abnormal finding 4 months after surgery. The patient remained asymptomatic 16 months after operation.

DISCUSSION

Venous aneurysm is an unusual vascular malformation. Primary aneurysm, defined as an aneurysm without prior trauma or inflammatory disease and without arteriovenous fistula, is quite rare. These lesions occur most commonly in the jugular veins, central thoracic veins, visceral veins, and extremities. Gillespie et al[4] presented a retrospective review of 39 venous aneurysms in 30 patients. Of these, 77% were located in the lower extremities, 10% were in the upper extremities, and 13% involved the internal jugular vein. Fifty-seven percent of lower extremity aneurysms occurred in the deep venous system.

However, only four reports of a primary aneurysm of the iliac vein have been published in the English language literature (Table).[5-8] Most are a consequence of congenital or acquired arteriovenous fistulas.[9,10]

Symptoms of venous aneurysms depend on the location. Patients with aneurysms of the lower extremities generally complain of painful masses, and those with the jugular or axillary veins present with asymptomatic masses.[4]

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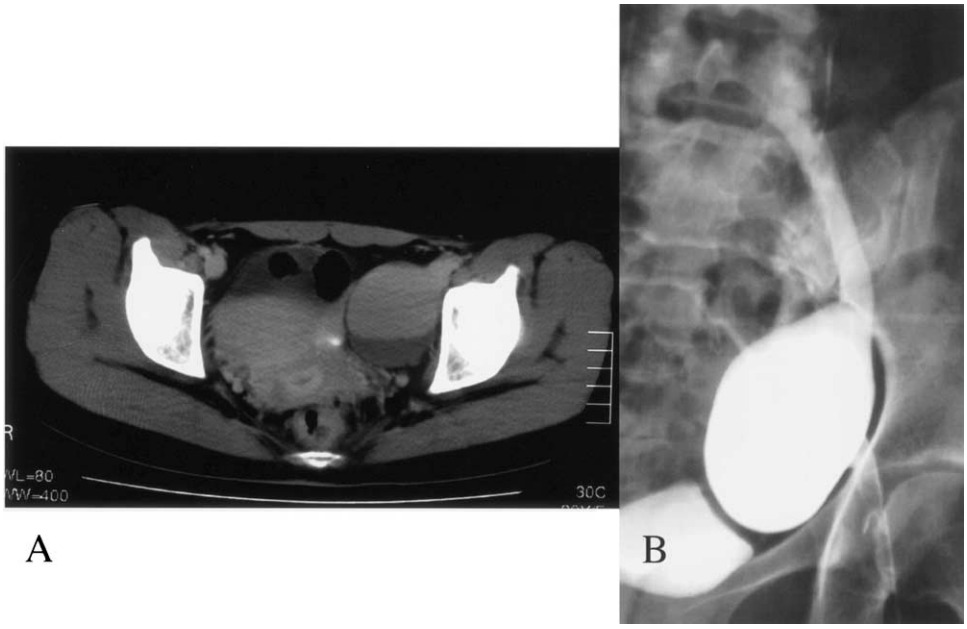

Fig 1. A, Computed tomogram showing an aneurysmal dilatation of the external iliac vein, 8 cm in diameter, which is compressing the bladder. **B,** Preoperative ascending venogram showing a solitary, saccular aneurysm.

One patient with iliac venous aneurysm developed acute onset of a painful swelling with bluish discoloration of the lower extremity caused by thrombosis.[5] The patient, a 69-year-old man, was an exercise enthusiast and had been jogging the previous evening. This case was associated with iliac compression syndrome. The authors reported that an obstruction to the venous return from the left leg, together with the increased blood flow during exercise, might have generated sufficient venous hypertension to stretch the vein wall, and an aneurysmal dilatation had been created over a period of time. The second patient reported an onset of exercise intolerance and episodic hemoptysis.[6] This patient, a 33-year-old man, was a healthy marathon runner and long-distance bicyclist. The complaints had appeared to be due to recurrent pulmonary embolisms caused by internal iliac venous aneurysm. The symptoms in the above two cases were due to thromboembolic events. The third patient developed pain and tenderness in the right lower quadrant of the abdomen. This patient, a 19-year-old man, underwent laparotomy for suspected appendicitis, which revealed the venous aneurysm.[7] On the second postoperative day, the aneurysm thrombosed, which was diagnosed by means of an edema that suddenly occurred, a venogram, a computed tomography scan, and a magnetic resonance imaging scan. The symptom was resolved after the second operation. The last case, a 21-year-old woman, was asymptomatic, and she underwent explorative laparoscopy because it was thought that an adnexal cyst was detected on ultrasound scan.[8] The lesion proved to be an aneurysmal dilatation of the left external iliac vein.

Recommended treatment of venous aneurysm depends on the anatomic location. Aneurysms of the jugular or axillary vein or upper extremities might be treated expectantly. Because of the high risk of thromboembolism, even with full anticoagulation, popliteal venous aneurysm must be treated surgically. Aldridge et al[1] reported 24 cases of popliteal venous aneurysms in which all patients had thromboembolic events. Seventeen of 24 cases (71%) had pulmonary embolism, and 8 of 12 patients (75%) who were on anticoagulation therapy experienced pulmonary embolism.

The clinical significance of iliac venous aneurysm is unknown. However, embolism, rupture, and thrombosis might occur in this location as well as in the popliteal fossa or others.[1-3] In fact, three of four reported patients with iliac venous aneurysms had a thromboembolic event. The remaining patient received 6 months of oral anticoagulation therapy after laparoscopy. In our case, we believed that because the uterus would enlarge during pregnancy, it might compress the aneurysm and increase the risk of complications.

Surgical procedures such as ligation, tangential excision with lateral venorrhaphy or autologous vein patch, and resection with reconstruction have all been advocated as definitive treatments.[1] Once a thromboembolic event or rupture occurs, ligation or resection is mandatory. When complications have not occurred, surgical decision making is more complicated. Resection with end-to-end anastomosis can be performed in selected cases. Simple ligation of the iliac vein predisposes the patient to post-thrombotic leg syndrome. For these reasons, venous reconstruction after resection of the aneurysm is needed. Reconstruction of the large veins has been described previously.[11] Spiral saphenous vein grafts or prosthetic grafts are used for the inferior

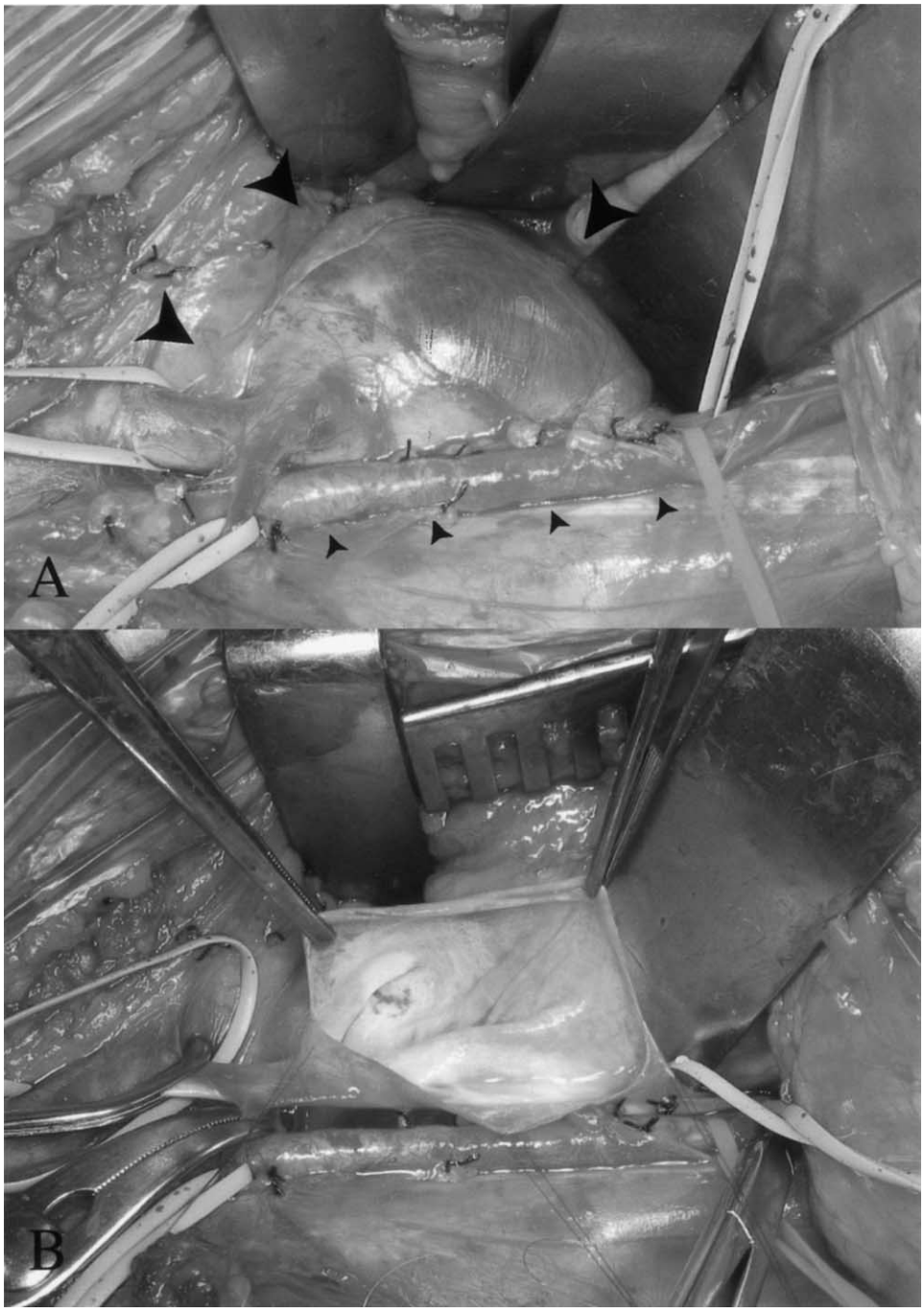

Fig 2. Intraoperative photographs. **A,** The venous aneurysm *(large arrows)*, exposed through a left lower quadrant retroperitoneal incision and a vertical left femoral incision, was located medial to the external iliac artery *(arrows)*. **B,** The aneurysm was opened after clamping of the iliac vein proximally and distally. No clot was seen within the aneurysm, and the aneurysmal wall was thin and smooth.

vena cava or iliac vein reconstruction. The authors reported that all femorocaval or long iliocaval grafts were reconstructed with a femoral arteriovenous fistula, and they said that the efficiency was still unknown. However, each procedure carried the risk of occlusion and required long-term anticoagulation therapy after surgery. Although one case did use a superficial femoral vein graft for venous reconstruction, this procedure is less desirable because it increases wound size and requires longer operation time. In the case reported here, the saccular aneurysm was treated

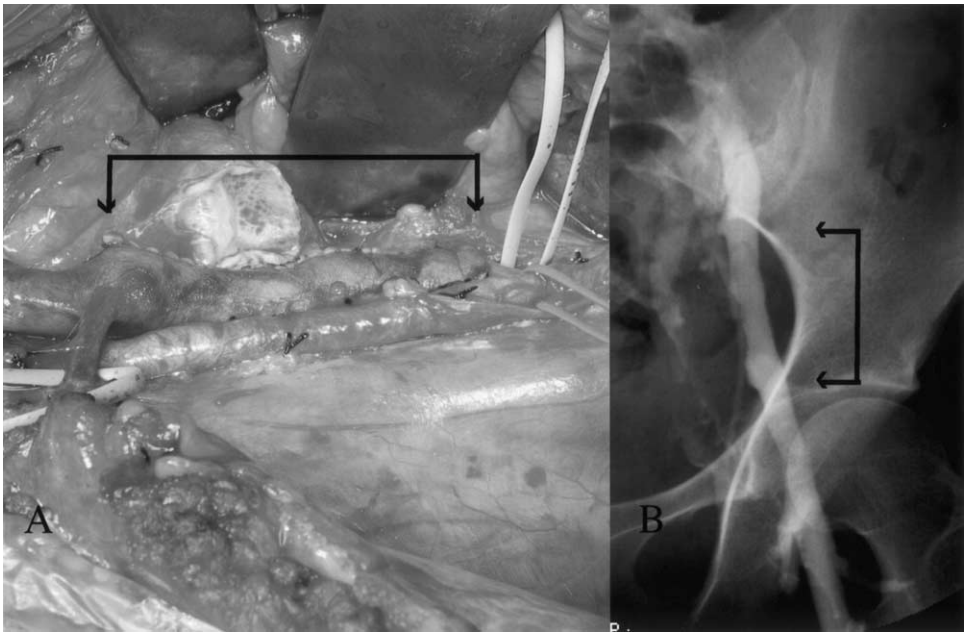

Fig 3. A, Intraoperative photograph. Lateral venorrhaphy was made by the continuous suture with four stay sutures. The length was about 6 cm. **B,** Postoperative ascending venogram showing the patent external iliac vein without dilatation. *Arrows* indicate the region of venoplasty.

Iliac venous aneurysms

Author/ year	*Sex*	*Age (y)*	*Symptom*	*Thromboembolic complication*	*Aneurysm location*	*Preoperative diagnosis*	*Operative procedure*	*Outcome*	*Follow-up time (mo)*
Hurwitz et al[5]/1989	M	69	Painful swelling of lower extremity	Thrombosed aneurysm	Left common external iliac vein	Venous aneurysm	Resection with reconstruction with prosthetic graft creating AV fistula	Asymptomatic	22
Postma et al[6]/1989	M	33	Exercise intolerance and episodic hemoptysis	Recurrent pulmonary embolism	Left internal iliac vein	Venous aneurysm	Simple ligation	Asymptomatic	12
Petrunic et al[7]/1997	M	19	Abdominal pain	Thrombosed aneurysm	Left common iliac vein	Appendicitis	Resection with lateral venorrhaphy	No complication	12
Fourneau et al[8]/ 1998	F	21	Asymptomatic	None	Left external iliac vein	Adnexal cyst	Resection with reconstruction with the SFV of the opposite side	Irritation of saphenous nerve by graft harvesting	18
Banno et al/2004	F	20	Asymptomatic	None	Left external iliac vein	Venous aneurysm	Resection with lateral venorrhaphy	Asymptomatic	16

AV, Arteriovenous; *SFV*, superficial femoral vein.

by lateral venorrhaphy, which is safe and offers excellent long-term patency.

The common histologic appearance of a venous aneurysm is a thickened, fibrotic intima with medial thinning caused by a decrease in the number of medial smooth muscle cells.[1,12] Pathologic examination in the present case showed that all three layers were well preserved without any inflammation or medial thinning.

This report describes a very rare case of external iliac venous aneurysm. This is the first case of uncomplicated iliac venous aneurysm successfully treated with aneurysmorrhaphy. Because of the risk of thromboembolic

events, aneurysms of large veins in the lower body should be treated prophylactically even when asymptomatic.

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