A Case in Which Sarcoma Was Diagnosed Two Years after Stent Placement to Treat Right Iliac Vein Stenosis that Accompanied Severe Unilateral Leg Edema

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Abstract

A 70-year-old man complained of right leg swelling due to right iliac vein stenosis. No mass was identified around the stenotic site, and the vessel wall had not become thickened. Self-expandable stents were positioned at the stenotic site. About two years later, chest CT revealed lung nodules. Pathology showed sarcoma. A mass that was considered to be the primary lesion was found around the stent in the right iliac vein. Although sarcoma of the iliac vein is very rare, it should be considered in the differential diagnosis of iliac vein stenosis, even if there are no suspicious findings from image studies.

Key words: sarcoma, iliac vein stenting, iliac vein stenosis, leg swelling

(Intern Med 51: 2165-2168, 2012)
(DOI: 10.2169/internalmedicine.51.6794)

Introduction

The iliac vein is known to be narrowed or occluded if thrombus has occurred in the vein (1) or the vein is externally compressed by, e.g., a pelvic tumor (2, 3) or the iliac artery (4-7). Although rare, primary venous tumors have also been reported as the cause of stenosis or occlusion of a vein (8-12). We encountered a case in which lung metastasis led to a diagnosis of a sarcoma originating from the iliac vein two years after stent placement to treat right iliac vein stenosis of unknown etiology, for which imaging studies provided no findings suggesting a tumor. The case suggests that although sarcoma of the iliac vein is very rare, it should be considered in the differential diagnosis for iliac vein stenosis, even if there are no suspicious findings in radiograms or echograms. We report a rare case of a tumor that slowly progressed after initial examinations by imaging techniques were not suspicious for a tumor.

Case Report

The patient was a 70-year-old man who complained of swelling of the whole right leg. His family history was non-contributory. He had been receiving treatment for diabetes and hypertension for more than 10 years. He became aware of swelling of his whole right leg from late February 2007; in early March 2007 he visited his primary-care physician who referred him to our hospital. His whole right leg had been markedly swollen at the time of our initial evaluation in early March. Vascular ultrasound and contrast-enhanced computed tomography (CT) showed narrowing of the right iliac vein. No mass was found around the stenotic segment, and the vessel wall had not been thickened (Fig. 1). Contrast venography performed on admission confirmed the iliac vein stenosis (Fig. 2). For revascularization, the right popliteal vein was punctured to insert a 6 Fr guiding sheath. A Dejavu floppy shaft guidewire (Cordis, Miami Lakes, FL, USA) was passed through the stenotic segment to perform intravascular ultrasound (IVUS), which found neither external compression nor thickening of the vein wall (Fig. 3). After predilation by a 5.0-40 mm Wanda balloon catheter (Boston Scientific, Natick, MA, USA), a 10-60 mm SMART stent (Cordis) was inserted into the stenotic site, and then postdilation was performed using a 8.0-40 mm Wanda catheter. Because there was a residual pressure gra-
Figure 1. (A&B) Images of pelvic computed tomography (CT) before the stent placement procedure. Contrast-enhanced CT showed the right iliac vein stenosis (white arrow). No mass was identified around the stenotic segment, and the vessel wall was not thickened.

Figure 2. An image of contrast venography before the stent placement procedure. White arrows show the right iliac vein stenosis. The patient was in prone position.

Figure 3. An image of IVUS before the stent placement procedure. IVUS image found neither external compression nor thickening of the vein wall.

Figure 4. An image of contrast venography after the stent placement procedure. White arrows show satisfactory dilation with self-expandable stents. The pressure gradient across the stents disappeared.

dient at the proximal side of the stent and IVUS confirmed that stenosis still remained at the segment, a 10-40 mm SMART® stent and a 10-40 mm E-Luminexx® (Bard, Murray Hill, NJ, USA) stent were additionally placed at the stenotic segment, with postdilation by a 8.0-40 mm Wanda® catheter. We ultimately needed three stents as we failed to position these stents owing to jumping up. Angiography confirmed satisfactory dilation (Fig. 4). The procedure was completed after performing IVUS, which also confirmed satisfactory dilation at the site. The pressure gradient across the stents disappeared. The leg edema disappeared immediately after the procedure, and the appearance of the lower limbs became symmetrical after a few days. The patient was discharged with prescription of oral warfarin.

At a routine examination in November 2008, nodules were detected in the right middle lung fields by chest radiograph. Chest CT images also indicated two nodular contrast areas in the middle lobe of the right lung (Fig. 5). Positron emission tomography (PET) performed in late November
found accumulation in the right inguinal region as well as two nodules in the middle lobe of the right lung and one nodule in the right upper lobe.

In mid-December 2008, resection of the middle lobe and partial resection of the upper lobe were performed. Pathology indicated spindle cell/pleomorphic sarcoma.

CT images in mid-January 2009 revealed a mass around the stent in the right iliac vein (Fig. 6). In early February, resection of the right inguinal tumor and reconstruction of the right iliac artery and vein were performed. Resected tumor size was 30×35×60 mm. Pathology indicated spindle cell/pleomorphic sarcoma. Sarcomas in the lung were multiple; multiple sarcomas which primarily developed in the lung are very rare. In addition, the size of sarcoma around his right iliac vein was much bigger than each pulmonary sarcoma. Therefore we diagnosed sarcoma around the right iliac vein was primary and pulmonary multiple sarcomas were metastasis.

The patient did well after the surgery. However, he had graft infection with methicillin-resistant Staphylococcus aureus in December 2009. Unfortunately, he died of sepsis in January 2010.

**Discussion**

In daily practice we occasionally encounter swelling of the leg resulting from stenosis or occlusion of a vein. Many such conditions are caused by thrombus in the vein (1) or external compression exerted by, e.g., a pelvic tumor (2, 3) or the iliac artery (4-7), and definite diagnosis for these cases can be relatively easily made by imaging studies. Although rare, a vein can also become narrowed or occluded because of a sarcoma originating from the vein (8-12). In these cases of venous sarcoma, tumors could be discovered as a mass around the vein (8-12); in such a case, diagnosis of the tumor would be easy. However, the present case does not match any of these cases; percutaneous ultrasound, CT, angiography, and IVUS could not detect a definite mass around the stenotic segment of the vein, and thickening of the vein wall was not observed. This made us fail to suspect the existence of a tumor. The reason why the invasive sarcoma caused leg swelling even though there was no obvious mass might be because the sarcoma invited fibrotic change in the vein and reduced elasticity of the vein.

Stent placement was considered to be effective in improv-
ing the symptom of marked swelling of the leg (13), and it did improve the symptom immediately and drastically. However, a sarcoma of the iliac vein was diagnosed about two years later, due to lung metastasis. We have confirmed that the images produced at the initial examinations had no findings showing thickening of vein wall or the existence of a mass. If there was such a finding neoplastic lesions would have been suspected, but in fact, the diagnosis of tumor was difficult for the present case, due to the lack of findings from percutaneous ultrasound, CT, or IVUS to show those conditions. The case suggests that when a stenotic lesion of a vessel is encountered, the possibility of a tumor originating from the vessel should always be considered, even if, as in the present case, evident abnormality such as thickening of vessel wall or existence of a neoplastic lesion could not be found.

To treat a tumor of vascular origin, revascularization is needed after thorough lymphadenectomy and dissection of the tumor (8-12). However, without an evident mass, as in the present case, it is difficult to perform biopsy for diagnosing a tumor. If a stenotic lesion lacks a definite diagnosis, it might be diagnostically valuable to consider a bypass surgery after resection of the lesioned vessel, rather than performing endovascular repair.

Some reports state that PET is helpful for the diagnosis (14). Also, in the present case, accumulation was observed by PET. It may be valuable to use PET to rule out the possibility of neoplastic lesion for stenosis of unknown etiology.

Conclusion

In the present case of iliac vein stenosis with the symptom of severe swelling of the lower limb, neoplastic lesions could not be suspected at first, because neither thickening of the vein wall nor a mass around the stenotic site was detected. Later, however, lung metastases were found, which led to the diagnosis of a sarcoma originating from the iliac vein. The case suggests that for a narrowed vessel, PET and/or diagnostic surgical treatment should also be considered, bearing in mind the possibility of a neoplastic lesion even if a mass cannot be found.

The authors state that they have no Conflict of Interest (COI).

References


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