Leiomyosarcoma Of The Femoral Vein In A Marathon Runner

Wilmer D. Bradley, M.D., Karl B. Fields, M.D., and Martha J. Delaney, M.A.

Physical fitness is associated with good health. A recent study by Blair, et al.\(^1\) found that a good fitness level prospectively predicted lower mortality for cancer and heart disease. Physicians, like laypersons, often assume good health in those who exercise regularly. Although physicians expect to find sports injuries in physically active individuals, they can overlook signs or symptoms that indicate systemic disease. Runners, for example, often develop injuries of the hips and lower extremities. Clement, et al.\(^2\) found that 96 percent of running injuries involved these areas and that most injuries occurred from overuse. Most lower extremity overuse injuries should cause characteristic physical findings and improve with appropriate treatment. A runner who develops diffuse swelling of a lower extremity warrants examination for systemic illness unless a specific sports injury can be identified.

**Case Report**

A 58-year-old male marathon runner who had been running 5 to 10 miles each day began to have left leg swelling and tightness after exercise. There was no associated pain, and he continued his running schedule and weight-training regimen for 2 weeks before consulting his primary care physician. At that time, the patient recalled no changes in training to suggest overuse, no traumatic injury, and no recent illness or generalized systemic symptoms. His physician tentatively diagnosed “muscle strain” and aspirin therapy was started. During the next 2 weeks, his leg tightness and swelling gradually increased, occurred without running, and eventually involved the thigh and entire left lower leg.

One month after onset of symptoms he came to us for a sports injury evaluation. His physical examination showed painless left lower extremity edema without other pertinent physical findings. Vascular occlusion was suspected, and a venogram was ordered. This test demonstrated a prominent filling defect in the proximal portion of the left superficial femoral vein. The deep system in the leg and thigh showed patency but markedly delayed filling. Because the findings were consistent with thrombosis, the patient was admitted to the hospital for intravenous heparin therapy and further evaluation. By the second hospital day swelling had decreased, and a soft tissue mass was palpable in the left femoral region.

The occurrence of venous thrombosis and a palpable mass in an active patient without clear risk factors suggested the possibility of malignancy. For this reason a computerized axial tomography (CT) scan of the upper thigh and abdomen was obtained. A soft tissue mass consistent with tumor or lymphadenopathy could be seen compressing the vascular structures in the left femoral triangle. Subsequent needle aspiration biopsy of this mass showed spindle cell sarcoma, also known as leiomyosarcoma.

Oncological consultants found on magnetic resonance imaging no further metastases. The patient subsequently underwent two courses of chemotherapy with intravenous doxorubicin hydrochloride and intraarterial cisplatin. In addition, he received local radiation therapy for tumor debulking in preparation for a limb salvage procedure. At the time of surgery, the tumor was noted to have originated from the left femoral vein, and the vein was sacrificed during a wide local excision. Evaluation of the tumor by the pathology laboratory confirmed leiomyosarcoma.

After an uneventful postoperative course, he returned home. He continued to have persistent left leg swelling, lessened by the use of a Jobst stocking. After 4 months he resumed full-time work and exercised by walking 3 miles each day. Unfortunately at his 1-year follow-up, his chest...
radiograph showed multiple lung nodules consistent with metastatic disease.

Discussion
Leiomyosarcoma, a highly malignant tumor, arises from smooth muscle. Sarcomas themselves rarely occur, comprising less than 1 percent of all cancers, for an incidence of less that 2/100,000.3 The range of incidence of leiomyosarcoma among histological types of soft tissue sarcomas is 2.4 to 11.3 percent.3 Leiomyosarcoma usually arises from visceral organ smooth muscle, with very rare occurrences from vascular smooth muscle or sweat glands. Of the known cases of leiomyosarcoma arising from blood vessel walls, 75 percent involved the inferior vena cava.4

In our review of the literature, we found fewer than 200 reported cases of leiomyosarcoma arising in the extremities.5-9 Those originating from the femoral vein were decidedly uncommon; Stitz and Boulter10 documented only 10 reports up to 1976. Only a few reports have appeared since then.11 Typically the patient experiences lower extremity edema, mass, and occasionally pain. Pain rarely occurs unless a palpable mass is present. Leiomyosarcoma of the distal inferior vena cava can similarly present with lower extremity edema, but typically the findings occur bilaterally.

Prognosis for vascular leiomyosarcoma generally is poor, with a 5-year survival of 32 percent to 47 percent.12 As with most neoplasms, early detection improves survival. Even though rare, vascular tumor must be considered as a cause for unusual lower extremity swelling. More common causes of generalized extremity swelling include deep venous thrombosis, mechanical causes (compression from braces, wraps), arteriovenous fistula, varicose veins, and lymphedema.13,14 Compartment syndrome, strain of the medial head of the gastrocnemius muscle, tendon ruptures, and perostitis can be added to the differential diagnosis in an athletic patient.15 Athletic injuries typically cause more specific physical findings, and the history helps confirm the diagnosis. A remote reason for extremity swelling is reflex sympathetic dystrophy, which occurs in a previously injured limb.16 Historical features, again, raise the question of this condition as a diagnostic consideration.

Deep venous thrombosis commonly causes extremity swelling but rarely occurs in a well-conditioned athlete. Usually deep venous thrombosis requires other risk factors, such as a coagulation disorder, trauma, cigarette abuse, or use of birth control pills.14 Tumors can precipitate thrombosis through a procoagulant effect or by mechanically obstructing vessels. Leiomyosarcoma directly occludes vascular channels in a fashion that mimics thrombosis.17 Disruption of flow, even without total venous obstruction, can lead to secondary thrombus formation.

This patient's overall good health, activity level, and lack of associated risk factors made him an unlikely candidate for deep venous thrombosis. The absence of a clear cause led to further diagnostic work-up and consideration of rare conditions. In retrospect he described symptoms that were similar to the other leiomyosarcoma cases involving the femoral vein. The gradual onset of his swelling fits the observations of other patients with slow-growing tumors who had symptoms lasting up to several years.12,18-20 In this case computed axial tomography scan led to direct needle biopsy of the mass and rapid confirmation of a tissue diagnosis. Magnetic resonance imaging also can be a useful adjunct in further delineating soft tissue invasion and endovascular extension.21

Summary
Leiomyosarcoma of the femoral vein is a rare tumor. Physicians involved in the care of athletic patients must not be cavalier in evaluating overuse injuries and should endeavor to make a specific diagnosis. If atypical findings, such as generalized extremity swelling, are present, the physician must consider systemic illness including malignancy in apparently healthy, physically active individuals.

References


