SHORT REPORT

Epithelioid Hemangioendothelioma on Left Femoral Artery After Multiple Femoral Artery Interventions

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Epithelioid hemangioendothelioma (EHE) is an extremely rare tumor of vascular origin with intermediate malignancy potential. It usually originates from veins and very rarely from arteries. This case report presents a patient who underwent femoral mass resection after multiple femoral artery interventions for coronary heart disease with a probable preoperative diagnosis of pseudoaneurysm of the femoral artery. He had complete resection of the mass with corresponding segment of femoral artery because of atypical gross appearance. The histopathology revealed an intravascular EHE with moderate mitotic index and mild cellular abnormalities.

Keywords: Vascular tumor; Hemangioendothelioma.

Introduction

Iatrogenic arterial injuries may result from any invasive diagnostic or therapeutic procedure but the relative occurrence and severity of these arterial injuries with those of penetrating and blunt vascular trauma is unknown. Local complications after femoral arterial catheterization, such as hematomas, pseudoaneurysms, arteriovenous fistulas, and arterial occlusions, are becoming more common with the growing numbers of complex invasive procedures in increasingly older patients.

Vascular tumors consist of a heterogeneous group varying from benign to a highly malignant subgroup and one of the rare form of this group is EHE, which was accepted to be an intermediate or borderline type and first described in 1923. Its usually seen as a solitary mass of soft tissues of middle-aged patients without sex predilection. More than half of these tumors originate from a large vein of an extremity (iliac or femoral vein). In this case report, we discuss one of the extremely rare vascular malignancy, challenging the standard technical approach to this well known complication of arterial interventions.

Case Report

A 77-year-old man was referred because of a pulsatile mass on left femoral artery. The patient’s past medical history revealed smoking and recent multiple femoral artery interventions for coronary heart disease (coronary angiography, PTCA and stent replacement). The mass was associated with pain in the inguinal area and mild claudication. On physical examination, he was found to have a blood pressure of 140/85 mmHg and a heart rate of 90 bpm. The lungs were clear to auscultation, and cardiac examination was normal except a mild systolic murmur across the precordium with radiation to the neck. Abdominal examination was normal without any palpable mass. A pulsatile mass was found at the femoral artery puncture site in the left groin. Peripheral pulses on left lower extremity were diminished compared to the right. The ECG showed rhythm of the pacemaker inserted 1 year ago and echocardiogram showed a globally decreased left ventricular function with an ejection fraction of 35%. Significant laboratory findings included erithrocyte
sedimentation rate of 42 mm/h and hemoglobin of 11.2. The patient was referred for peripheral arterial angiography and Doppler ultrasound investigation, and both reported a pseudoaneurysm of the common femoral artery (Fig. 1). We began the operation with the preoperative diagnosis of femoral artery pseudoaneurysm. During surgery, vertical incision was made over the pulsation, to expose distal and proximal parts of the artery for standard pseudoaneurysm repair. After dissecting the fatty subcutaneous tissue we reached the mass which was smooth outside with big lobulations inside, yellowish-gray and homogeneous, and measured $6 \times 5 \times 4$ cm$^3$. Because of this uncommon appearance we sent a specimen for frozen section, which showed thrombosis with mild cellular atypia. With this indefinite and unclear result, we performed a wide dissection with the surrounding tissue and removed 1.5 cm of the femoral artery including the possible pedicle like connection with the mass. We repaired the femoral artery by end-to-end anastomosis. Routine postoperative vascular follow-up of the patient was uneventful. However, tissue pathology revealed one of the uncommon vascular tumor, epitheloid hemangioendothelioma (Fig. 2). The cells were variably spindle shaped and epitheloid with moderate amounts of eosinophilic cytoplasm. There were moderate mitotic figures. No tumor necrosis was noted.

**Discussion**

Femoral artery pseudoaneurysms (PSA) occur as a complication of procedures requiring femoral artery access such as cardiac or peripheral angiography and prolong hospitalization, consuming health-care resources and result in significant morbidity. Incidence of PSA ranges from 0.2 to 8% and appears to be increasing. Several authors suggest that therapeutic rather than diagnostic interventions predispose patients to developing PSA.4 Coronary angiography, angioplasty and coronary stenting are recognised risk factors for false aneurysm formation.5 With the increasing number of these procedures performed each year, adequate measures must be taken to ensure prevention, and this is especially important in patients receiving anti-thrombotic drugs, all of which are additional risk factors for its formation.6 Treatment options for this pathology evolved in the past decade are towards non-surgical management, which are ultrasound guided compression, thrombin injection and implantation of endovascular covered stents.7,8

Differential diagnosis of these masses at femoral region creates a confusing problem, especially if it has occurred after multiple femoral artery interventions, as it was in our case. One of the rare differential diagnosis of these masses are vascular malignancies which are extremely uncommon.9

Epithelioid hemangioendothelioma is an intermediately aggressive neoplasm of vascular origin and to date about 35 cases have been reported in the literature.10 Its usually venous in origin often found in the deep soft tissues of the extremities, but also can occur within different tissues or organ parenchyma.
These tumors have no gender predilection and occur in the 2nd to 9th decades of life. Local pain and swelling, that may be of weeks to years duration, are the most common clinical symptoms. Basic histological investigation shows cords of round to spindle shaped endothelial cells. In addition to this, cellular pleomorphism, increased mitotic findings and indefinite lesions can also be seen in these type of rare tumors. Ultrasonographic evaluation of femoral masses can diagnose pseudoaneurysms or arteriovenous malformations in most of the patients but in cases like this, its usually not enough for accurate diagnosis. MRI technique, on the other hand, may be helpful in imaging and demonstrating the exact pathology. In fact, all of these non-invasive methods of examination and angiography are not usually enough for exact preoperative diagnosis of these probable soft tissue tumors of vascular origin. Postoperative total body CT scans and MRI studies revealed no local and distant metastasis in our patient. In fact local recurrence is equal to 13%, and 30% of these tumors will metastasize; therefore, follow-up program is highly recommended to study regional lymph nodes and lungs, which are the most involved sites.

The experience with epithelioid hemangioendothelioma is limited, and long-term behavior of this neoplasm is uncertain. Adjuvant chemotherapy, radiation therapy, or both have no proven benefit and questions regarding treatment planning still exist, although most sources regard wide excision as the treatment of choice.

The radiological features of these kinds of masses must always be correlated with the clinical and histopathologic findings to arrive at an accurate final diagnosis because the imaging findings are non-specific. As EHE has been rarely identified, a thorough evaluation of the clinical, histopathologic, and radiologic variables should be strictly considered to properly assess and treat this rare entity.

References


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