CASE REPORT

Angiosarcoma at the femoral artery puncture site: A diagnostic dilemma

Femoral arter ponksiyon alanında anjiyosarkom: Tanısal çıkmaz

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Summary– The femoral artery is the most common vascular access site used for angiographic interventions. Various complications, such as hematoma, bleeding, dissection, arteriovenous fistula, and pseudoaneurysm have been described following an iatrogenic puncture. However, angiosarcoma formation at the access site is very uncommon and poses a diagnostic dilemma, due to the resemblance to an organized hematoma. A 75-year-old patient who had previously undergone coronary angiography developed angiosarcoma with vascular complications due to local invasion at the puncture site. Although the tumor was completely excised and flow was re-established, the patient died 17 months later as a result of multiple metastases and associated complications. The presence of a persistent mass with vascular complaints should raise suspicion for this rare and aggressive type of tumor.

Özet– Femoral arter anjiyografi işlemleri sırasında en sık kullanılan vasküler giriş yolu özelliğini taşımaktadır. İşlem sonrası hematom, kanama, diseksiyon, arteriyovenöz fistül ve psödoanevrizma gibi çeşitlli komplikasyonlar tanımlanmıştır. Giriş bölgesinde anjiyosarkom oluşumu ise oldukça nadir olup, organize hematom ile benzerliğinden dolayı doğru ve zamanında tanısı zordur. Yetmiş beş yaşında bir hastada koroner anjiyografi sonrası anjiyosarkom gelişmiş ve de lokal yayılıma bağlı olarak girişim bölgesinde vasküler komplikasyonlara yol açmıştır. Kitlenin tamamen çıkarılmasına ve kan akımının tekar sağlanmasına rağmen hasta 17 ay sonra çoklu metastaz ve de komplikasyonları sonucunda kaybedilmiştir. Girişim bölgesinde kalıcı bir kitlenin varlığı nadir görülen bu tümörün varlığından şüphe ettirmelidir.

Angiosarcoma is an aggressive metastatic tumor. Familial syndromes, lymphangiomas, hemangiomas, hemochromatosis, and lesions, such as port wine stains, can play an important role in angiosarcoma. Early diagnosis is important with respect to mortality and morbidity concerns.

Several publications have appeared in recent years documenting the diagnosis, treatment, and prognosis of angiosarcoma. This study is a rare clinical presentation of angiosarcoma that was identified after an angiographic intervention. The aim of this case report was to note that this aggressive tumor should be considered in the presence of a persistent mass with vascular complaints.

CASE REPORT

A 75-year-old patient who had undergone coronary angiography at another institution 6 months earlier presented at the outpatient clinic. He had undergone triple coronary bypass surgery 10 years prior. The latest coronary angiography revealed severe native coronary lesions with functioning grafts. No further coronary intervention was deemed necessary. The early postprocedural period was uneventful except for a mild hematoma at the puncture site and he was discharged the next day.

Abbreviation:

DVT *Deep vein thrombosis*

The patient was later admitted to the cardiology clinic with pain in the right groin. There was tenderness and a lump at the puncture site. These findings were consistent with the presence of a hematoma. Another visit was scheduled and he was sent home, but the patient was lost to follow-up.

Approximately 6 months later, he presented at the cardiovascular surgery outpatient clinic with complaints of typical intermittent claudication and edema

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in the right leg. There was a relatively weak pulse in his right lower extremity and edema without any sign of acute DVT (deep vein thrombosis). Duplex sonography revealed total occlusion of the femoral artery secondary to a mass in the groin area. Venous flow was interrupted at the femoral level, but the lumen below the groin level was free of thrombosis. Magnetic resonance imaging scans confirmed the presence of a 55x49-mm mass with heterogenic density (Fig. 1). Digital subtraction angiography was performed and the right femoral artery was found to be totally occluded at the level of the initial puncture site (Fig. 2). Distal flow was visible through the collaterals.

The patient was scheduled for surgery in order to re-establish blood flow in the extremity and removal of the mass. Following a standard right groin incision, a solid mass encircling both the femoral vein and artery was exposed. Initial intraoperative inspection of the mass was suggestive of a neoplasm rather than an organized hematoma. The lesion was densely adhered to the vessels and it was excised en-masse with the vessels (Figs. 3, 4). Venous and arterial flow was subsequently re-established with 8-mm expanded polytetrafluoroethylene graft interposition (Fig. 5).

A histopathological examination of the specimen was positive for cluster of differentiation 31 and Factor VIII but negative for human herpesvirus 8, calretinin, cytokeratin 7, keratin 20, thyroid transcription factor 1, and prostate-specific antigen. These findings confirmed the diagnosis of a high grade angiosarcoma and the patient was referred to the oncology department. During the course of his treatment he suffered from ipsilateral DVT accompanied by a recurrent mass located in the neighboring muscle tissue. Multiple lung metastases were later detected. The patient was lost 17 months after his initial diagnosis.

DISCUSSION

The femoral artery is the most common vascular access site used for interventional procedures. Its caliber makes it less prone to spasm and permits the use of larger catheters. Various complications, however, have been described following an iatrogenic femoral artery puncture. Hematoma and minor bleeding can

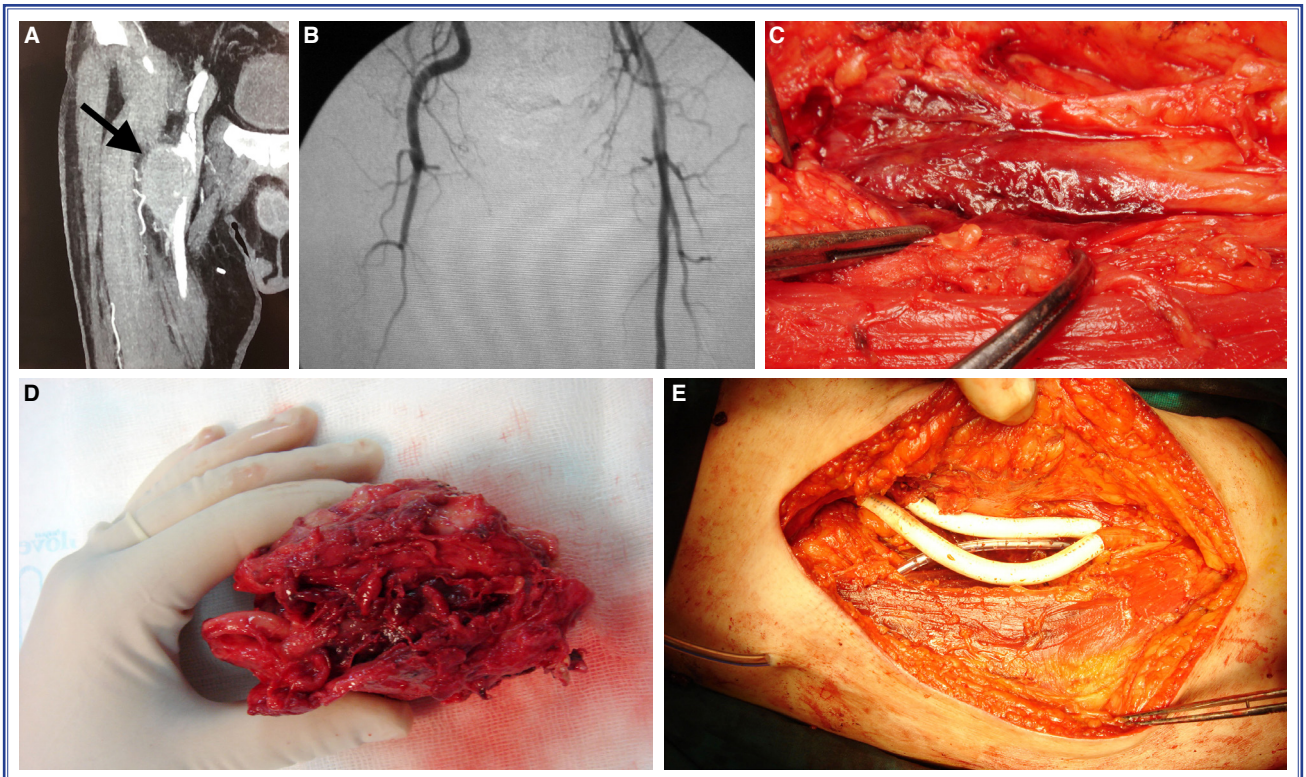

Figure 1. **(A)** Magnetic resonance imaging scan of the mass. **(B)** Digital subtraction angiography DA image showing interrupted flow at right superficial femoral artery. **(C)** Intraoperative view of the tumor. **(D)** Totally excised tumor with portions of femoral vein and artery visible inside the mass. **(E)** Re-established venous and arterial flow with expanded polytetrafluoroethylene graft interposition.

be managed conservatively, while major complications, such as dissection, pseudoaneurysm, and arteriovenous fistula might require additional interventions.

The absence of distal flow accompanied by claudicatio intermittens after a vascular intervention suggested the presence of a local dissection or distal emboli secondary to a pseudo aneurysm.[1] However, a tumor is very seldom suspected in such circumstances, due to the rare occurrence of vascular tumors, even among sarcomas.[2–4] These tumors are classified according to their immunohistological origin as either endothelial or mesenchymal, and according to their morphology as intimal or mural.[5–7]

Angiosarcoma stems from the intimal layer and has an endothelial differentiation, which can be seen in atypical endothelial cells.[8] It is an aggressive and metastatic tumor.[9] Although the etiology is unknown, various factors have been implicated, including foreign graft materials, chronic edema, radiotherapy, and carcinogens, such as polyvinyl chloride and thorotrast.[1,10,11] Angiosarcoma can also be associated with familial syndromes, such as neurofibromatosis type-1, Maffucci syndrome, von Recklinghausen syndrome, hemochromatosis, and Klippel–Trenaunay syndrome.[12]

Once detected, options for treatment in advanced cases are limited to radical surgery, which can even include amputation of the involved extremity.[1,4,10] In less advanced cases, vascular patency should be restored with graft interposition after en-bloc resection of the tumor. In accordance with general guidelines of tumor surgery, at least a 2-cm disease-free margin should be secured after resection. A sole endarterectomy has also been proposed as an alternative to complete resection, but the risk of recurrence is higher.[2–6]

Naturally, early diagnosis is crucial in malignancies and especially for such an aggressive type of tumor. In our case, the diagnosis was delayed due to misinterpretation of the mass as a persistent organized hematoma. Though it is extremely rare, angiosarcoma should be included in the differential diagnosis in cases with a persistent mass at the puncture site.

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