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Case Report

Treatment of a persistent sciatic artery aneurysm by endovascular exclusion method through internal iliac artery closure

İnatçı siyatik arter anevrizmasının endovasküler dışlama yöntemi ile internal iliyak arter kapatılarak tedavisi

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ABSTRACT

A persistent sciatic artery (PSA) is an extremely rare congenital vascular malformation. A 64-year-old female patient was admitted with recurrent distal embolism of the right lower limb with symptoms of ischemic vascular complications. Computed tomography and angiography revealed an entirely occluded distal PSA and an accompanying aneurysm. The PSA aneurysm was successfully treated using two self-expanding stent grafts to occlude the origin of the internal iliac artery. After arterial reconstruction, ischemic symptoms showed complete recovery.

Keywords: Aneurysm/surgery; arteries/abnormalities; endovascular techniques; femoral artery/endovascular procedures.

ÖZ

İnatçı siyatik arter (PSA) çok nadir görülen bir doğuştan vasküler malformasyondur. Altmış dört yaşında kadın hasta iskemik vasküler komplikasyon semptomları ile birlikte sağ ayakta tekrarlayan distal emboli ile başvurdu. Bilgisayarlı tomografi ve anjiyografide distal PSA'nın tamamen tıkalı olduğu ve eşlik eden bir anevrizma olduğu izlendi. İnatçı siyatik arter anevrizması, internal iliyak arter girişini kapatmak için kendiliğinden genişleyen iki stent greft kullanılarak başarılı bir şekilde tedavi edildi. Arteriyel onarım sonrasında iskemik semptomlarda tam iyileşme izlendi.

Anahtar sözcükler: Anevrizma/cerrahi; arterler/anormallikler; endovasküler teknikler; femoral arter/endovasküler işlemler.

A persistent sciatic artery (PSA) is a rare congenital vascular malformation.[1,2] The embryonal axial or sciatic artery in the fetus persists in the adulthood and connects the internal iliac arteries (IIAs) to the popliteotibial arteries.[3] Hypoplasia of the femoral artery (FA) results in the PSA, providing the dominant inflow to the lower extremity. A PSA is prone to development of atherosclerotic stenosis as well as aneurysm degeneration, and is often associated with a higher rate of thromboembolic complications to the lower extremity.[4,5]

Herein, we present a female patient with complaints of right leg pain, cyanosis, and numbness and diagnosed with an entirely occluded distal PSA and accompanying aneurysm which was successfully treated using two self-expanding stent grafts.

CASE REPORT

A 64-year-old female patient was admitted to our emergency room with a 12-h history of a sudden-onset and severe pain in her right leg along

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with pallor and hypothermia. Physical examination revealed a non-palpable and diminished right femoral, popliteal, pedal, and posterior tibial arterial pulses. All of the left leg pulses were palpable and normal. Doppler ultrasonography revealed complete occlusion of the superficial femoral and popliteal arteries. The patient was sent to the operating room emergently, and femoral thromboembolectomy was performed with extraction of well-formed thrombi from the femoral (superficial and deep) and popliteal arteries. A satisfactory blood flow was achieved, and hand-held Doppler ultrasonography (Doppler Ultrasonic, Maquet, Cardiopulmonary Medikal Teknik San. Tic. Ltd. Şti., Antalya, Turkey) revealed biphasic pedal pulses after the thromboembolectomy procedure. The superficial FA was narrower than expected for this patient. In the postoperative period, anticoagulation therapy with low-molecular-weight heparin (2×100 U/kg, bid) was maintained. A written informed consent was obtained from the patient.

The following day, the patient complained of recurrent right leg pain, and developed cyanosis of the extremity. A full embolic work-up was performed including electrocardiogram, transthoracic echocardiography, and computed tomography (CT) angiography. Cardiac and other systemic examination findings were within the normal ranges. Upon further questioning, the patient described a history of right hip trauma in her childhood and a swelling on her right hip, since she was seven years old. On physical examination, a pulsatile mass was detected on her right hip (Figure 1a). Duplex ultrasonography revealed a large mass of 100.7×43.8 mm originating from a vascular structure (Figure 1b). Three-dimensional reconstruction of the CT images which were obtained three years ago demonstrated a complete PSA with incompletely developed femoral arteries, and an aneurysm sized 66 mm in diameter at the level of the posterior aspect of the major trochanter of the femur (Figure 1c). The CT demonstrated a right-sided

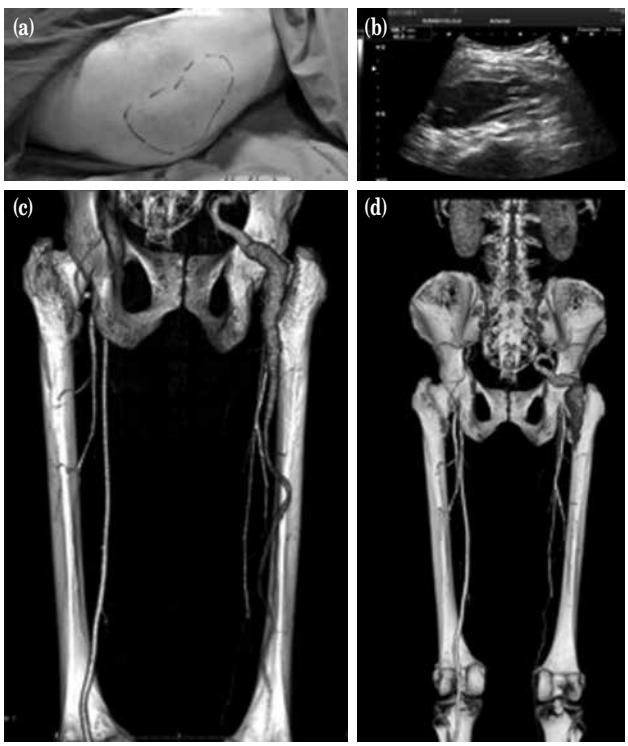

Figure 1. Physical examination, superficial and Doppler ultrasonography and computed tomography showed a persistent sciatic artery. **(a)** The pulsatile mass on the patient' right hip (dotted line). **(b)** Doppler ultrasonography revealed a large mass of 100.7×43.8 mm originating from a vascular structure. **(c)** Three-dimensional reconstruction of the computed tomography images obtained three years ago demonstrated a complete persistent sciatic artery with incompletely developed femoral arteries and an aneurysm 66 mm in diameter at the posterior aspect of the major trochanter of the femur (posterior view). **(d)** Three-dimensional reconstruction of the computed tomography images demonstrated a complete occlusion of the distal segment of the persistent sciatic artery.

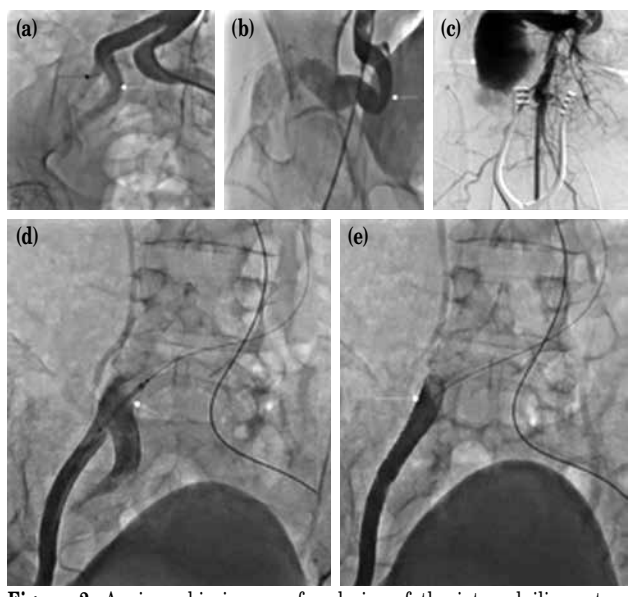

Figure 2. Angiographic images of occlusion of the internal iliac artery resulting from aneurysmatic persistent sciatic artery. **(a)** Angiogram showed an enlarged IIA (white arrows) and external iliac artery (black arrow). **(b, c)** Persistent sciatic artery with a tortuous and aneurysmatic pattern extending from the origin of the internal iliac artery. **(d, e)** Endovascular stenting of the internal iliac artery, and completion angiograms showing good patency of the metal stent following the procedure.

complete PSA with a narrower superficial femoral artery (SFA), thromboembolic total occlusion of the one third distal SFA, and a 66-mm aneurysm at the posterior aspect of the major trochanter of the femur with normal contralateral iliac and femoral arteries (Figure 1d). In addition, complete occlusion of the distal segment of the PSA was detected (Figure 1d). Angiography demonstrated the presence of the PSA and associated aneurysm (Figure 2a-c). Endovascular occlusion of the IIA was performed successfully with two overlapping self-expanding graft stents (27×10 mm and 27×9 mm stents; Bentley Innomed GmbH, Hechingen, Germany), as shown in Figures 2d, e. Completion arteriogram demonstrated complete occlusion of the IIA and aneurysm without any endoleak (Figure 2e). Repeated thromboembolectomy procedure was performed following the stent implantation with a hybrid procedure. A hand-held Doppler examination revealed biphasic Doppler pedal pulses. The ischemic complaints of the foot disappeared. The pulsating mass disappeared, the distal limb pulses returned to normal after 24 h, and the ankle-brachial index was measured as 0.9. The patient's postoperative recovery period was uneventful, and she discharged from hospital five days after the procedure. She remained asymptomatic during a six-month follow-up period.

DISCUSSION

During embryogenesis, the sciatic artery, a branch of the umbilical artery, provides the blood supply to the lower extremity buds. In the third month of the embryonic development, the sciatic artery regresses completely, and SFA continues to provide all blood supply to the lower extremity. While the majority of the sciatic artery is regressed, some parts are involved in the formation of other arteries such as the inferior and superior gluteal, peroneal, and popliteal arteries. If there is a failure in the development of the femoral vascular system, the axial artery may persist as the sciatic artery.[6] Based on the main blood flow of the lower extremity and development of FA and PSA, Pillet et al.[7] classified PSA into four categories while Gauffre et al.[8] added a fifth type.

Accordingly, PSA can be classified as follows:

Type 1: Complete PSA and normal FA

Type 2: Complete PSA and incomplete FA (2a: SFA is available, but cannot reach the popliteal artery, 2b: All parts of the SFA are rudimentary).

Type 3: Incomplete PSA (it is developed only in the proximal region of lower extremities and FA is normally developed).

Type 4: Incomplete PSA is only developed in the distal region of lower extremities and FA is normally developed.

Type 5: The PSA originates from the median sacral artery (5a: SFA is normally developed, 5b: SFA is rudimentary).

The classification is particularly important in guiding medical or surgical therapies. Our case corresponds to a PSA Type 2a, and only treatment of the PSA and related aneurysm may be sufficient to regain a distal blood flow, and SFA may not necessitate any further surgical treatment such as a bypass.

The PSA originates from IIA, passes through the greater sciatic foramen, and stays adjacent to the sciatic nerve (often coursing within the sciatic nerve sheath in certain patients).[3] Persistent sciatic artery is located inferior to the gluteus maximus muscle, travels along the edge of the adductor magnus muscle, and reaches the popliteal fossa where it joins the popliteal artery.[3] The vascular structure of a PSA is hypoplasic, and destined to degenerate during development. The anatomical location and inherent properties of the vascular tissue in a PSA make it highly susceptible to tortuous and ectatic vascular development. Furthermore, the PSA is subject to repetitive trauma, often resulting in aneurysmal degeneration due to its location.[3,6] Indeed, a PSA associated with aneurysms has been reported in 15 to 40% of the cases in the literature.[3,4] The patient in the current report described a right hip trauma which resulted in pulsatile swelling in a large bruised area, when she was seven years old. Thus, an acute injury may also have contributed to the aneurysm formation in our patient.

Aneurysmal degeneration of a PSA has been shown to be a potential source of recurrent thromboembolism in the lower extremity.[2,4] As in the present case, despite a completely occluded PSA, clots formed in the aneurysmal segment resulted in embolism to the FA branches. We believe that the most likely mechanism for thromboembolism spilling retrograde into the femoral artery is a mechanical compression of the superficially located large PSA aneurysm by

surrounding muscles or direct external pressure. The aneurysm, laden with a large thrombus burden, may have functioned as a pipette bulb, milking the thrombus into the femoral artery when externally compressed.

Steps need to be taken to prevent recurrent FA embolism in these patients. In patients with adequate blood flow to the distal lower extremity, the aneurysmal segment can be treated with surgical or endovascular techniques, as in our case.[2] If the blood flow to the lower extremity is impaired, treatment is usually completed with a femoral-distal bypass.[6] In particular, in patients with unilateral PSA, the IIA may be occluded by a stent graft placement at the origin at the common iliac separation, if necessary.[5] In the present case, lower extremity ischemic symptoms showed improvement after the procedure, and no additional complications were seen.

In conclusion, decision regarding the treatment of PSA depends on symptoms, anatomy, occlusive vascular disease, concomitant aneurysm, and type of PSA. The treatment of choice can be determined based on the status of each patient. The common goals of all the therapies are to subvert the problem of the lower extremity ischemia, prevent embolic events, and rupture of aneurysm.

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