A Very Rare Cause of Thrombotic Peripheral Occlusion

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Abstract

A 45-year-old healthy woman presented with claudication of the right leg. The resting ankle-brachial index (ABI) was reduced to 0.6, and a duplex scan revealed an occlusion of the right popliteal artery. Angiography presented a patent superficial femoral artery that ends above the knee joint. Laterally, there was delayed retrograde contrast filling of the popliteal artery. After exploring the internal iliac artery, we crossed a thrombotic occlusion of a persisting sciatic artery (PSA). Local thrombolysis with recombinant tissue plasminogen activator (1 mg/h) was initiated. The Angiography 18 hours later showed a reduction of thrombotic material and relevant stenosis in the proximal part of the vessel. Residual thrombus and the stenosis were covered by two stentgrafts (Gore Viabahn Endoprosthesis) that were stabilized by an interwoven stent (Supera). Final angiography displayed a patent sciatic artery and a three-vessel run off. Postinterventional ABI was normalized to 1.0. The magnetic resonance imaging 6 days after the intervention demonstrated a patent PSA again and a normal blood flow on the left leg. A PSA should be included in the differential diagnosis of lower limb ischemia or suspected aneurysm formation. We demonstrated the feasibility of an interventional approach with an excellent outcome in this case.

Keywords

- acute limb ischemia
- thrombotic occlusion
- persisting sciatic artery
- thrombolysis
- stentgraft
- interwoven stent

Acute limb ischemia (ALI) is a limb and life-threatening condition[1] that requires urgent evaluation and management. It is characterized by a sudden decrease in arterial perfusion of the limb, with the symptom duration of less than 2 weeks.[2,3]

Common causes of ALI are embolism, thrombosis of native arteries or reconstructions, peripheral arterial aneurysm, as well as spontaneous dissection, and traumatic arterial injury.[4] Also, uncommon causes like vasculitis, popliteal entrapment syndrome, adventitial cystic disease, paradoxical embolism, and foreign body embolization have to be kept in mind.

An extremely rare cause of ALI is a persisting sciatic artery (PSA). PSA is a seldom vascular anomaly and was first described in a post-mortem case in 1832.[5] It has been estimated to occur in 0.01 to 0.06% of the population.[6] PSA is associated with several complications like aneurysm formation, neuropathy due to compression of the sciatic nerve, or thromboembolism resulting in critical limb ischemia.[7]

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Table 1 Initial ankle-brachial index

	Right (mmHg/index)		Left (mmHg/index)	
	Rest	Post-exercise	Rest	Post-exercise
brachial artery	*150*	*165*	135	
posterior tibial artery	85/0.6	50/0.3	150/1.0	150/0.9
dorsalis pedis artery	85/0.6	40/0.2	150/1.0	150/0.9
Walking distance, localization of pain (S1: the onset of pain, S2: interruption of exercise (treadmill, 12% slope; 3.2 km/h speed)				
P1: 75 m the right calf, 95 m the right calf and right posterior				
P2: 181 m because of pain of the right calf				

We describe a case of a middle-aged lady presented with ALI due to a thrombotic occlusion of a PSA that necessitated interventional treatment.

Case Description

A 45-year-old healthy woman presented with acute onset of claudication of the right calf since 10 days. There were no specific cardiovascular risk factors overt. She had quit smoking 11 years ago.

The resting ankle-brachial index (ABI) on the right leg was reduced to 0.6, and the walking capacity on a treadmill (slope 12% and velocity 3.2 km/h) was impaired with 181 m. Post-exercise ABI was further diminished to 0.25 (►**Table 1**). A duplex scan was suggestive for occlusion of the right popliteal artery.

Therefore, we performed a digital subtraction angiography with a left retrograde approach. Iliac arteries seemed to be healthy. On the right side, we found a patent superficial femoral artery (SFA) that ended above the knee joint. Laterally, we detected a delayed retrograde contrast filling of the popliteal artery (►**Fig. 1**). Our hypothesis was that we were dealing with a vascular anomaly. After exploring the internal iliac artery, we were able to cross a thrombotic occlusion of a PSA that ended up in conjunction with the popliteal artery we have seen during the initial angiography. The initial sheath was replaced by a 5F 45 cm Fortress (Biotronik, Berlin, Germany) sheath. Local thrombolysis with a recombinant tissue plasminogen activator (rtPA) with 1 mg/h using a catheter with a lysis length of 50 cm (Cragg-McNamara 5F-Medtronic, Dublin, Ireland) was initiated. In addition, 500 IU Heparin/h were administered over the sideport of the sheath. The angiogram 18 hours later showed a clear reduction of thrombotic material. The underlying cause was a severely calcified relevant stenosis in the proximal part of the PSA. After upsizing the sheath to an 8F 45 cm system (Super Arrow-Flex, Teleflex Medical Europe Ltd., Athlone, Ireland), the lesion was predilated using a 6×150 cm balloon (Passeo-18, Biotronik, Berlin, Germany) in combination with a 0.014 inch wire (PT^2, BostonScientific, Marlborough, MA). Residual thrombus and the stenosis were covered by the implantation of two stentgrafts (Gore Viabahn Endoprosthesis: 8×100 and 8×50 mm, Gore Medical, Flagstaff, AZ) that were stabilized by a 7.5×100 mm interwoven stent (Supera-Abbott,

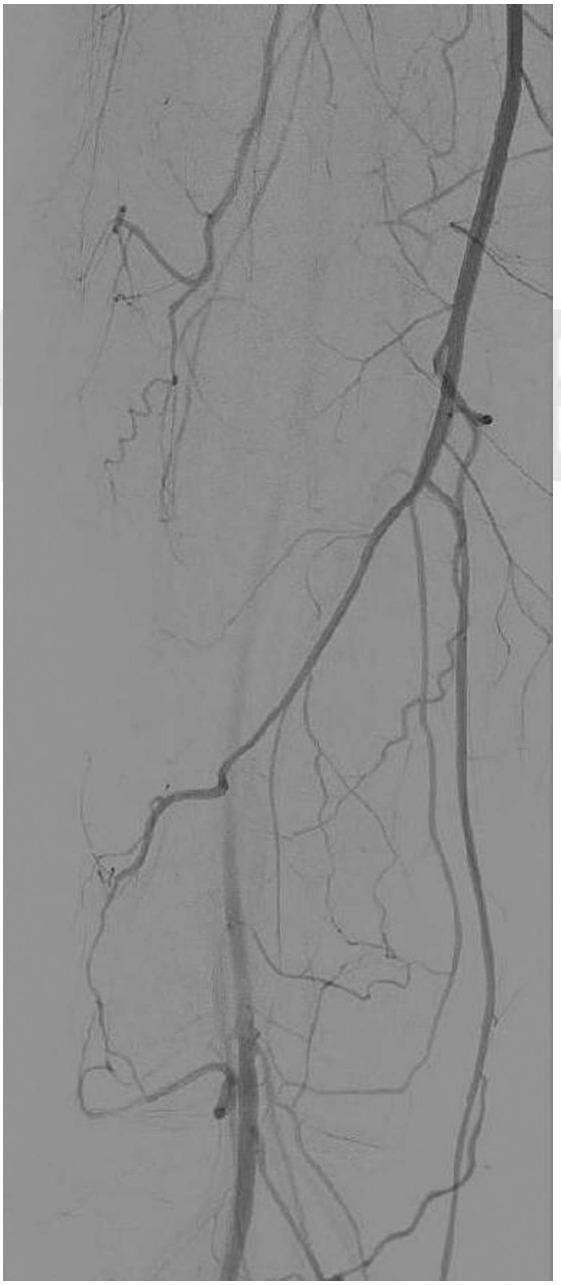

Fig. 1 Initial angiography demonstrated an incomplete superficial femoral artery (*right*) that ends above the knee joint and delayed contrast filling of the popliteal artery (*left*).

Fig. 2 MR angiography performed 6 days after the endovascular recanalization showing a patent PSA on the right side.

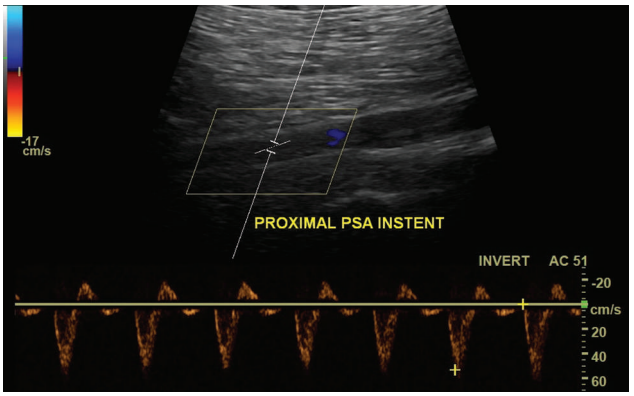

Fig. 3 Duplex ultrasound (with the patient in a prone position) 15 months after the index procedure displays a regular instant peak systolic velocity and triphasic wave form.

Chicago, IL). Final angiography displayed a patent PSA with a three-vessel run off. The puncture site was closed with the help of an 8F Angio-Seal (Terumo, Shibuya, Japan33). Post-interventional anticoagulation was started with 75 mg clopidogrel and 60 mg edoxaban. ABI was normalized to 1.0. A magnetic resonance imaging performed 6 days after the intervention showed again a patent PSA and a regular blood flow in the left leg (►**Fig. 2**).

An outpatient visit 14 weeks later demonstrated a regular resting ABI of 1.0. The treadmill exercise test could be performed without any complications (walking capacity >300 m), while the post-exercise ABI was slightly decreased to 0.8. Medication with clopidogrel and edoxaban was well tolerated. In consensus with the patient, we decided to stop the oral anticoagulant edoxaban. Visits 12 and 24 months later again demonstrated regular ABI, walking capacity (►**Table 2**), as well as duplex ultrasound (►**Fig. 3**). We advised her to attend our outpatient department annually.

Discussion

We presented a case of successful interventional treatment using catheter directed lysis and dedicated stents in an extremely rare case of ALI due to a thrombotic occluded PSA.

A PSA is very rare with around 200 cases reported so far.[8] As a branch of the umbilical artery, the sciatic artery supplies the lower limb with blood during embryonic development. Usually, sciatic artery disappears when the SFA has developed regularly during the embryonic course.[9] The reasons for an incomplete or absent development of the femoral artery are not clear. A bilateral PSA is present in approximately 30% of cases (pathology differentiates between five types, ►**Table 3**).[10] In our case with an incomplete SFA, we dealt with the IIa subtype. Patients with PSA are often asymptomatic, and PSA is found by chance during (physical) examination. A positive Cowies' sign—distal palpable pulse without palpable femoral (groin) pulse—is

Table 2 Ankle-brachial index 27 months after intervention

	Right (mmHg/Index)		Left_(mmHg/Index)	
	Rest	Post-exercise	Rest	Post-exercise
brachial artery	*140*		130	
posterior tibial artery	145/1.0	150/0.9	160/1.1	150/0.9
dorsalis pedis artery	155/1.1	160/0.9	155/1.1	145/0.9
Walking distance, localization of pain (S1: the onset of pain, S2: interruption of exercise (treadmill, 12% slope; 3.2 km/h speed)				
P1:				
P2: >300 m				

Table 3 Different types of a PSA

Type	1	2a	2b	3	4	5a	5b
PSA	Complete	Complete	Complete	Incomplete (persistent upper part)	Incomplete (persistent lower part)	originates from sacral median artery	
SFA	Normal	Incomplete	Absent	Normal	Normal	Developed	Absent

Abbreviations: PSA, persisting sciatic artery; SFA, superficial femoral artery.

considered pathognomonic for a PSA.[11] Symptomatic patients suffer from claudication, rest pain, necrosis, mass formation, and neurological problems.[7] Treatment options depend on symptomatology, classification, and the underlying pathology. A meta-analysis found that in 20% of cases, patients with a PSA were asymptomatic.[7] Asymptomatic patients should be followed-up clinically.[7,12] An aneurysm formation is the most common complication seen in 48%, followed by stenosis (7%) and occlusion (9%).[7] Surgical therapy options like ligation, bypass, and embolectomy have been described. In around 8% of the cases, amputation due to ischemia and distal complications was necessary.[7] An endovascular first strategy, including catheter-directed lysis, implantation of stents, and/or stentgrafts or coiling, was also mentioned in the already published case reports.[8] The reason for the occlusion in our case was most likely an appositional thrombus on the underlying high-grade stenosis of the PSA.

Recent guidelines state that an interventional approach using intra-arterial catheter directed thrombolysis can be performed with equivalent results in comparison to surgery[4] in patients with ALI due to thrombotic occlusion. As long as our patient had neither relative nor absolute contraindications for thrombolysis,[13] we decided to perform lysis using rtPA.

The second component of our lysis regime is heparin. By default, we administer 500 IE heparin/h over the sideport of the sheath to prevent pericatheter thrombosis and maintain its patency. There is a huge heterogeneity concerning the application and dosage (from no heparin to heparin with a target activated partial thromboplastin time of 1.5 to 2 times baseline).[14] Due to the lack of comparative randomized controlled trials, the guidelines do not recommend continuous systemic therapeutic heparinization.[4]

In cases with ALI and suspected thrombotic occlusion, a retrograde contralateral access should be preferred over an antegrade approach. It offers a stable position of the long crossover sheath and so reduces the risk of bleeding and dislodgement.[15] In addition, in our case, we would have missed the correct diagnosis with a femoral antegrade approach.

Although ALI in the described case was apparently caused by thrombosis of the diseased native vessel, we decided to set the patient on antiplatelet therapy with clopidogrel 75 mg and the direct oral anticoagulant edoxaban 60 mg. Recent guidelines gave a weak IIb recommendation for long-term anticoagulation after thrombectomy or endovascular treatment of a prosthetic bypass graft occlusion.[4] Due to the complex intervention with the implantation of two layers of stent materials in a highly flexible vessel region (proximal part in projection to the hip joint) we suspected that the PSA is prone to early reocclusion. On the opposite, we expected the bleeding risk to be very low in the 45-year-old otherwise healthy lady. After 14 weeks, edoxaban was stopped without bleeding complications.

Conclusion

In conclusion, it is important to think of this rare congenital vascular anomaly since this vessel is prone to complications such as acute lower limb ischemia and/or aneurysm formation. As described, an interventional approach is safe and feasible.

Conflict of Interest
None declared

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