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CASE REPORT

Monomelic Ischemic Neuropathy of the Tibial and Peroneal Nerve After Onyx Embolization of Vasa Nervorum Supplying a Surgically Excluded Popliteal Artery Aneurysm

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Abstract Ischemic neuropathy is an exceedingly rare complication after peripheral artery embolization. We report a case of ischemic damage to the tibial and peroneal nerve after embolization of the vasa nervorum that served as feeding collaterals to a surgically excluded popliteal artery aneurysm.

Keywords Embolization · Vasa nervorum · Popliteal artery aneurysm · Nerve ischemia · Ischemic neuropathy

Introduction

Ischemic neuropathy describes any hypoxic injury to peripheral nerves that is caused by insufficient arterial blood supply [1]. Causes of monomelic neuropathies include atherosclerotic peripheral arterial disease, thromboembolic events, iatrogenic arteriovenous fistula with subsequent distal ischemia, or compartment syndrome. Several reports have described ischemic neuropathy as a complication after endovascular embolization, for example of the facial nerve [2–5], the sciatic nerve [6, 7], the lumbosacral plexus, or even lumbar spinal cord [8, 9].

Main trunk tibial nerve neuropathies (with or without additional peroneal nerve involvement) are rare. In fact, a study from Drees et al. from the Cleveland Clinic [10] only found 52 affected patients within a time span of 25 years. The most common cause was trauma (50%), but the second most common cause was ischemia (19%), mostly due to embolic events or large artery occlusions. Few additional case reports describe ischemic neuropathy of the tibial and/or peroneal nerve after aorto-bi-femoral bypass [11] and coronary angiography [12], likely due to peri-interventional emboli. Direct iatrogenic embolization of vasa nervorum of the tibial nerve with glue has been described as a complication of endovascular treatment for recurrent hemarthrosis [13].

Case Report

A 54-year-old man was referred to our interventional radiology clinic for embolization of a popliteal type II endoleak equivalent. He had received surgical exclusion of a popliteal artery aneurysm on both sides via femoropopliteal-III-bypass with great saphenous vein grafts 9 years ago. According to the surgical reports, he had originally complained of progressive sensory disturbance and tingling of his toes (beginning at the fifth toe and spreading to the first toe) on both sides after walking 200–300 m, with full recovery over 45 min when standing still. Additionally, his calf muscles would initially painfully harden but get better soon after short ambulation (walk-through phenomenon). MRI of the lumbar spine had not shown spinal canal stenosis or herniated disk. Further

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work-up had revealed bilateral partially thrombosed popliteal artery aneurysms (28 mm and 25 mm). The sensory complaints and calf claudication had been interpreted as intermitting distal arterio-arterial embolization, which is why the popliteal artery aneurysms were treated with bypass surgery.

However, postoperatively the symptoms had not changed, and throughout all postoperative outpatient visits over the following 9 years the patient had kept complaining about the same tingling sensations and initial cramping of his calf muscles when walking. At his last pre-interventional visit, he had reported on newly occurring popliteal pain and swelling on the left side for the past week. Ultrasound and CT angiography both displayed a growing popliteal aneurysm on the left (40 mm) with perfusion of the aneurysm sac via a branch of the deep femoral artery (Fig. 1A), analogous to a type II endoleak. The finding was discussed with the patient and by our interdisciplinary vascular board which recommended endovascular embolization of the feeding vessel to prevent aneurysm rupture.

Catheterization and angiography of the deep femoral artery confirmed the CT finding (Fig. 2A); therefore, superselective catheterization of the aforementioned branch that fed the aneurysm sac was performed. During contrast injection (Iodixanol 300 mg/L, injection rate 3 mL/s, total volume 12 mL), the patient suddenly screamed from pain in his lower leg accompanied by numbness of the foot, both of which, however, quickly subsided. Embolization of the branch was performed with Onyx 34L (Medtronic plc, Dublin, Ireland), closing the afferent branch up until the popliteal aneurysm (Fig. 2B).

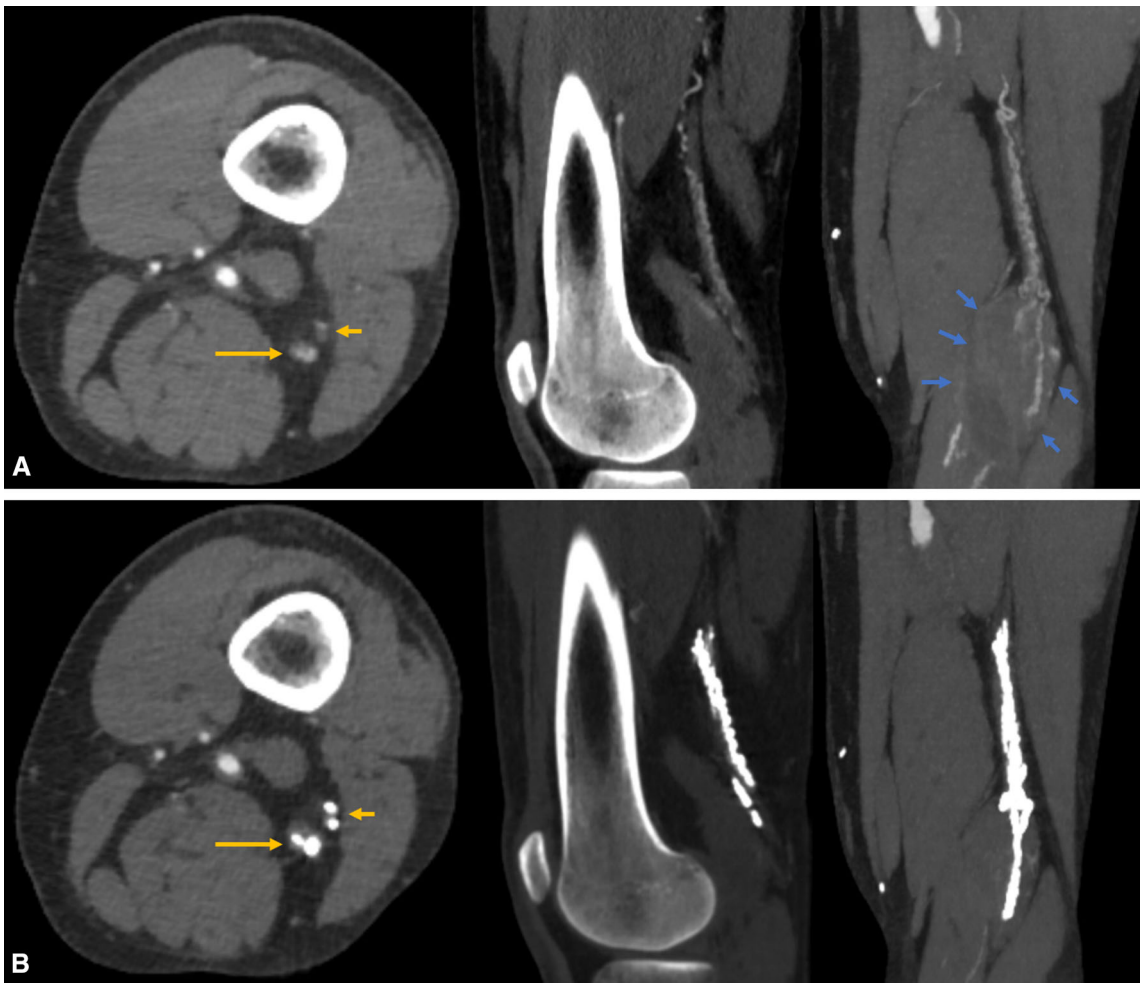

Fig. 1 **A** Pre-interventional CT angiography of the left leg at the level of the femoropopliteal junction in axial plane, sagittal plane, and coronal plane (from left to right). Yellow arrows on the axial plane point to the tibial nerve (long arrow) and common peroneal nerve (short arrow). The hyperdense/white dots inside the nerve structure correspond to the vasa nervorum that are supplied by a branch of the deep femoral artery. The blue arrows on the coronal plane outline the enhancing aneurysm sac that is perfused by the vasa nervorum. **B** Post-interventional CT angiography at the same planes. The vasa nervorum are now filled with hyperdense liquid embolic agent (Onyx 34L). The aneurysm sac is not perfused anymore

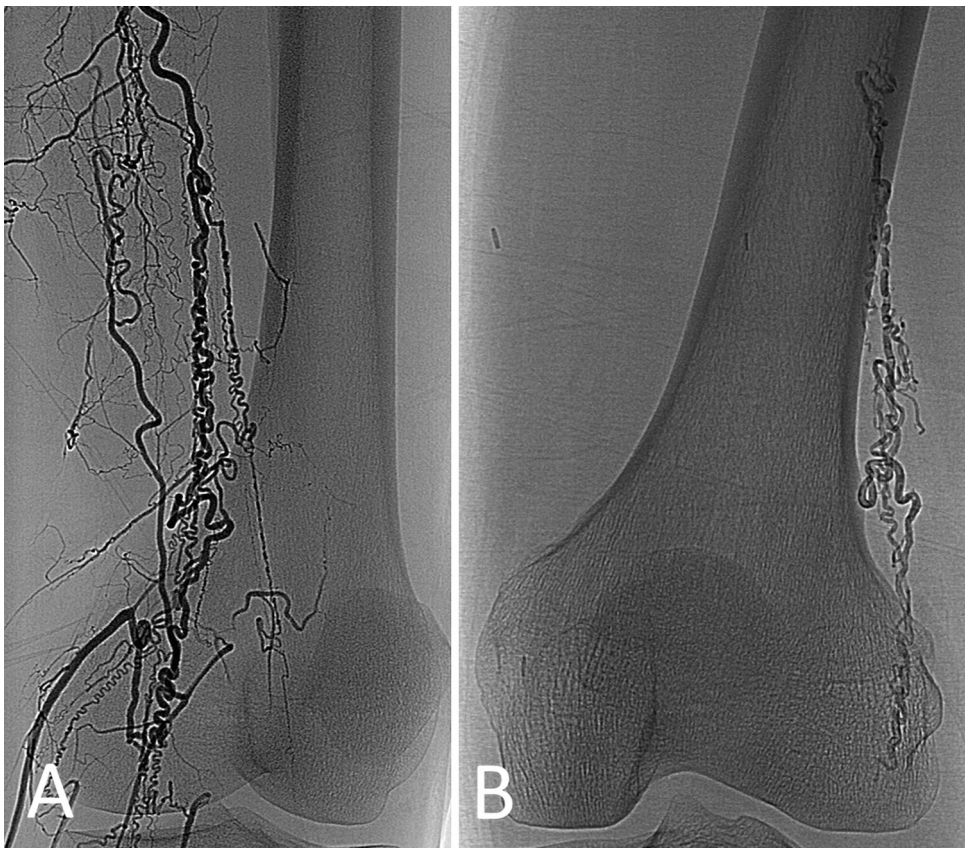

Fig. 2 **A** Oblique digital angiography after selective catheterization of the deep femoral artery shows a tortuous hypertrophied artery (in the middle of the picture). The vessel morphology is typical for vasa vasorum or vasa nervorum. Note the vast collateralization between the vas nervorum and the sural arteries supplying the calf muscles, which presumably lead to the "steal-phenomenon" when exercising. **B** X-ray after superselective embolization of the vas nervorum with Onyx 34L shows complete filling of the branch up to the aneurysm level. This corresponds to the pictures shown in Fig. 1B

During embolization, the excruciating pain occurred again, this time without subsiding. Despite strong pain medication including opioids, the patient continued to complain of severe pain as well as persisting numbness and weakness of his left foot.

Immediate neurologic consultation confirmed severe hypoesthesia and hypoalgesia of the left foot sole and dorsum, and to a lesser degree of the whole lower leg, together with weakness (strength 2/5) of the foot and the first toe in plantar flexion, dorsiflexion, inversion, and eversion. Achilles tendon reflex was absent. Electroneurography concluded bilateral chronic injuries to the tibial and peroneal nerves but with additional signs of acute nerve damage at the popliteal level on the left side. The results from electromyography were correlating with complete left-sided denervation of the innervated muscles. CT confirmed the Onyx 34L (Medtronic plc, Dublin, Ireland) distribution around the tibial and common peroneal nerves (Fig. 1B). Conservative management including physiotherapy and prescription of pregabalin was initiated. Two years after the intervention, the patient was able to partially recover muscle strength to 4/5 and was able to independently walk with the aid of crutches. Four years after the intervention, he was able to wean off the pregabalin completely and only needed the crutches for long walking distances.

Discussion

Our case highlights several important aspects of the development and clinical presentation of peripheral ischemic neuropathy with pictorial demonstration of the vasa nervorum as possible collateral feeders of aneurysms and consequences of inadvertent embolization. The tibial and common peroneal nerves are branches of the sciatic nerve with blood supply coming from several sources. At knee level, arterial supply arises from muscular, cutaneous, and geniculate branches of the popliteal artery [14] with extensive anastomoses between the many vessels in the lower limb vasculature. Since usually there is no single feeding artery, this leaves the nerves in a watershed area. This, however, allows for compensatory arterial collateralization when one or several supplying arterial branches occlude.

In our case, the patient initially presented with bilateral partially thrombosed popliteal artery aneurysms and neural claudication that manifested as exertion-dependent and transient sensory loss of his toes. This finding together with the lack of any spinal pathologies should make the physician wary of possible ischemic neuropathy at the level of the aneurysm. Although nerve palsies due to compressive popliteal pseudoaneurysms have been described [15, 16], symptoms in case of compression are neither exertion-

dependent nor intermittent. The neuropathy and painful calf muscle hardening in our case probably both arose from thrombotic-occluded small arterial branches that originated from the partially thrombosed aneurysm sac and which supplied the tibial and common peroneal nerve as well as the surrounding calf muscles. Due to the chronic nature of aneurysmatic disease, the vasa nervorum together with muscular arterial branches were able to eventually get most of their blood supplies from a branch of the deep femoral artery which subsequently hypertrophied (collateralization). When resting, both the nerves and the muscles are sufficiently perfused, and the patient remains asymptomatic. But when beginning to exercise, the muscles initially get into a state of relative ischemia, leading to calf pain (calf muscle claudication). While continuing to exercise, blood flow starts to divert from other areas including the vasa nervorum and toward the muscles, thus leading to the ischemic neural claudication, while muscle cramps simultaneously improved (walk-through phenomenon).

Because continued perfusion of aneurysms through feeding collaterals coming off vasa nervorum is an exceedingly rare phenomenon, this was only realized after embolization had already been performed. The best clue was the suddenly expressed pain already during superselective contrast injection which in a way would correspond to "provocative" testing. We hypothesize that this reaction was either caused by short-term ischemia or chemical irritation due to high local concentration of the contrast medium (similar to the amytal test). Knowledge of this rare complication helps to recognize the vasa nervorum on preinterventional CT angiography (Fig. 1) as well as clinical harbingers during the intervention. Alternative therapeutic options in such cases include direct sac puncture embolization or surgical excision of the aneurysm.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed Consent For this type of study, formal institutional review board consent is not required. Informed consent was obtained from all individual participants included in the study.

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