Vascular Malformations, Rare Causes of Sciatic Neuropathy: A Case Series

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BACKGROUND: Sciatica is typically a clear-cut symptom complex commonly related to an impingement at the spinal nerve level. Etiologies of sciatic neuropathy outside the neural foramina are uncommon.

OBJECTIVE: To describe 4 patients presenting with radiating leg pain due to sciatic nerve involvement, all with a vascular etiology.

METHODS: Four patients presenting with neuropathic pain were retrospectively reviewed. Preoperative 3 Tesla magnetic resonance imaging was used to identify these lesions, which most commonly showed diffuse T2 changes with nerve enhancement upon administration of contrast.

RESULTS: Exploration revealed vascular lesions. All patients went on to external and limited internal neurolysis of the involved sciatic nerve segment. Intraoperative histological study confirmed the presence of a venous angioma, an arteriovenous malformation, a venous malformation associated with Klippel-Trenaunay syndrome, and a capillary hemangioma. Follow-up demonstrated stable neurological examinations with reduction in pain at 1 year or greater.

CONCLUSION: In patients with sciatic distribution symptoms and signs, after initial negative spine imaging, high-resolution imaging of the sciatic nerve itself should be undertaken to address rarer causes such as vascular abnormalities. In these cases, exploration and fascicular biopsy provided a diagnosis; external and limited internal neurolysis improved pain.

KEY WORDS: Neuropathy, Peripheral nerve, Sciatic, Vascular malformation

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Although sciatica is most often radicular in origin, new radiating pain despite negative lumbar imaging studies warrants consideration of such nondiscogenic origins as plexopathy or neuropathy. Infrequent causes include benign or malignant tumors (eg, primary or secondary tumors), infections, mechanical entrapments, and vascular causes.[1,2] Of these, vascular causes, either isolated, posttraumatic, or syndromic, are the least common.[3-5]

Vascular lesions include both hemangiomas and vascular malformations. About 60% of vascular lesions occur in the head and neck region, followed by the trunk and extremities.[6] Their classifications are based upon the publications of Finn et al[7] and Mulliken and Glowacki.[8] Knowledge of the type of vessel affected, be it venous or arterial, and its categorization into "high or low flow," permits further lesional subdivision. Capillary hemangiomas are true tumors characterized by active endothelial proliferation, whereas telangiectasias, cavernous, venous, and arteriovenous lesions are considered malformative in nature.[7,8] Hemangioma affecting the peripheral nerve has been only rarely reported, most commonly affecting the median nerve.[3,6,7,9-18] Less frequent is involvement of the peroneal or tibial nerves, which present with pain as the initial symptom.[3,9,11,15,17] Opposed to the low-flow hemangiomas, arteriovenous malformations (AVMs) are high-flow lesions, characterized by aggressive growth and tissue destruction. The majority of AVMs are asymptomatic and affect the brain, lung, and the lower extremities. Lower-extremity AVMs most

ABBREVIATIONS: **AVM**, arteriovenous malformation; **EMG**, electromyography; **KTS**, Klippel-Trenaunay syndrome

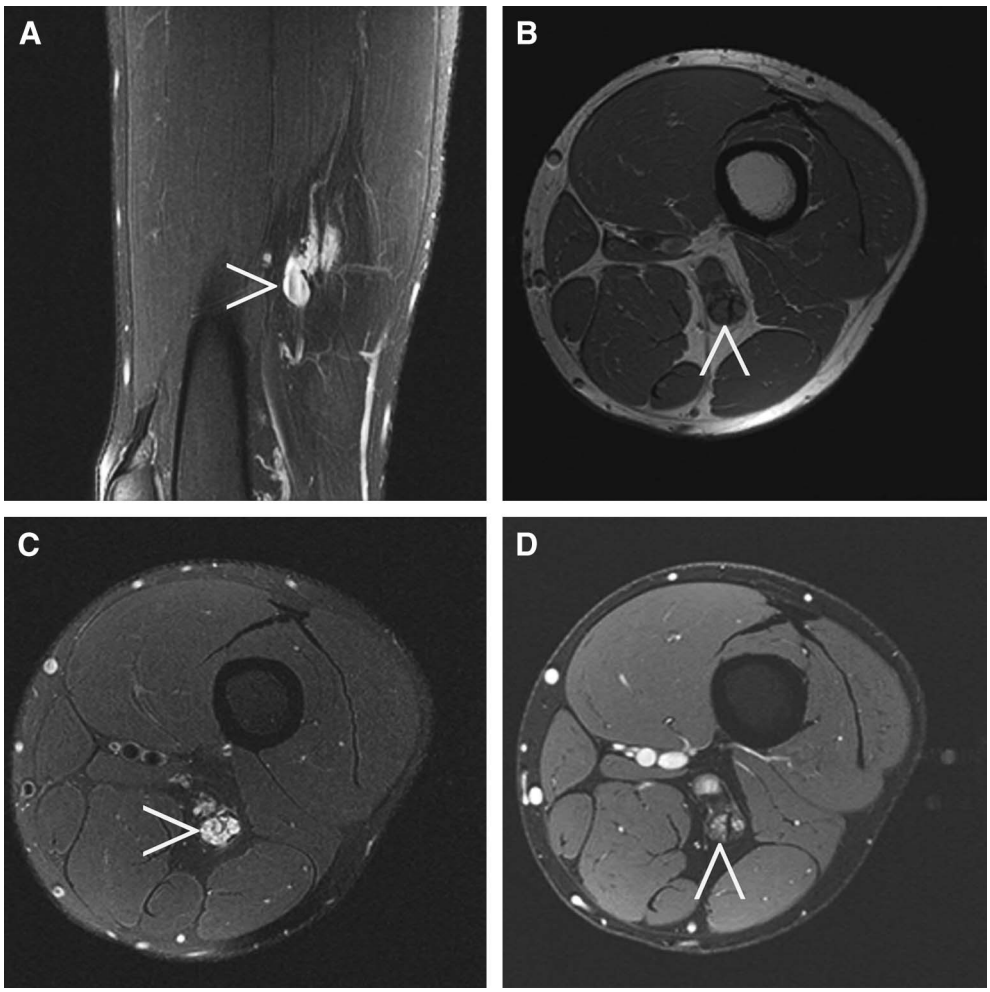

FIGURE 1. *MRI appearance of venous and cavernous angioma of the sciatic bifurcation (case 1).* **A**, *sagittal T1 weighted image with gadolinium of the distal thigh, arrowhead shows venous dilatation and vascular enhancement surrounding the sciatic bifurcation. Axial T1 (***B***), T2 (***C***), and T1 (with gadolinium) (***D***) weighted images in the distal thigh demonstrate the abnormality at the level of the distal sciatic nerve involving both the tibial and peroneal divisions (arrows). There is abnormal T2 signal and gadolinium enhancement present consistent with a vascular lesion.*

commonly manifest with dermatological signs including discoloration and swelling; fortunately, peripheral neuropathy is relatively infrequent.[3,9,11,15,17] To date, review of the literature by us and others reveals only 3 cases of vascular lesions causing sciatica.[3,19-22] Moreover, although multiple-organ involvement has been reported in Klippel-Trenaunay syndrome (KTS), the peripheral nervous system is rarely affected. Capillary, venous, and/or lymphatic vasculature is involved, but microscopic AVMs underlie the clinical features of KTS. To date, sciatica in KTS has rarely been reported. Nonetheless, peripheral neuropathy is a manifestation of the disorder.[8,23] Finally, although these vascular lesions represent a heterogeneous group of pathologies, it is likely that they share a common mechanism for their pain generation. It is unknown whether the effects of these vascular lesions upon nerve are due to compression, diversion of blood flow from the nerve (ischemia), or venous hypertension.

The present series reports 4 cases of sciatic neuropathy of vascular etiology collected over a 4-year period, including one example each of venous and capillary angioma, AVM, and hemangiomatosis, the latter in the setting of KTS. Each patient had a preoperative neurological evaluation, high-resolution magnetic resonance imaging (MRI) at 3 Tesla, and electrophysiological testing, operative exploration and external and limited internal neurolysis, fascicular biopsy (frozen and permanent sections performed to exclude vascular tumor), and postoperative follow-up assessment of at least 1 year.

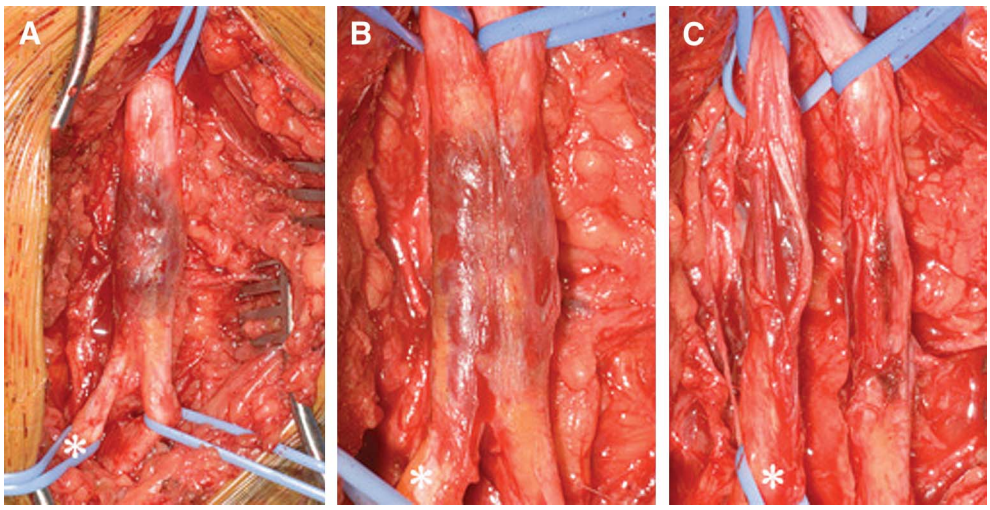

FIGURE 2. *Intraoperative appearance of venous and cavernous angioma of the sciatic bifurcation (case 1).* **A**, *intraoperative appearance of the sciatic bifurcation; top is proximal and the bottom is distal in all photos. The asterisk indicates the peroneal nerve adjacent to the tibial nerve. Note the blue discoloration of sciatic nerve just prior to its bifurcation.* **B**, *intraoperative photo after the proximal divisions of the sciatic nerve were separated in preparation for neurolysis of the blue lesion.* **C**, *despite internal neurolysis, vessels are seen to interdigitate between the nerves.*

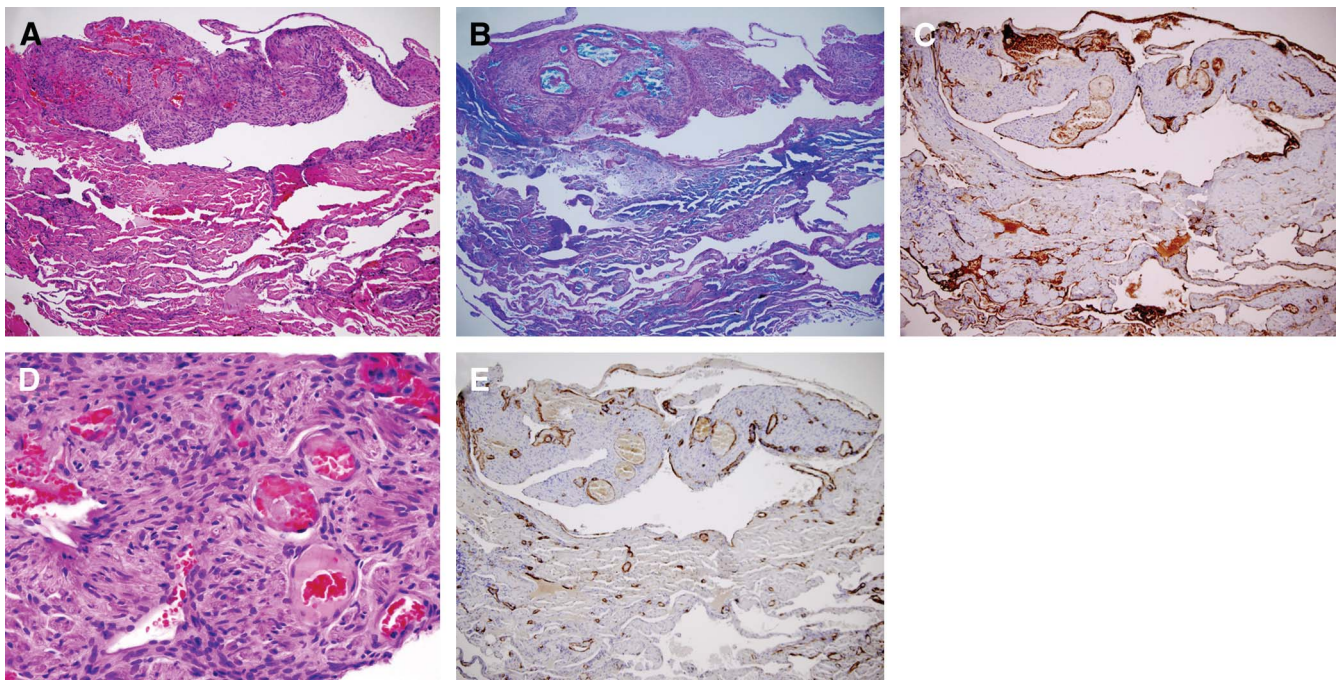

FIGURE 3. *Histology of combined venous and cavernous angioma of the sciatic bifurcation (case 1). The lesion consists of a cellular venous element (*A*, top) and numerous slit-like cavernous spaces (*A*, center). These 2 elements are best seen on Masson's trichrome stain (*B*). Although all vascular spaces are endothelial lined (*C*, Factor 8 immunostain), smooth muscle cells are most numerous in the upper element (*D*, hematoxylin and eosin stain;* E, *smooth muscle actin immunostain).*

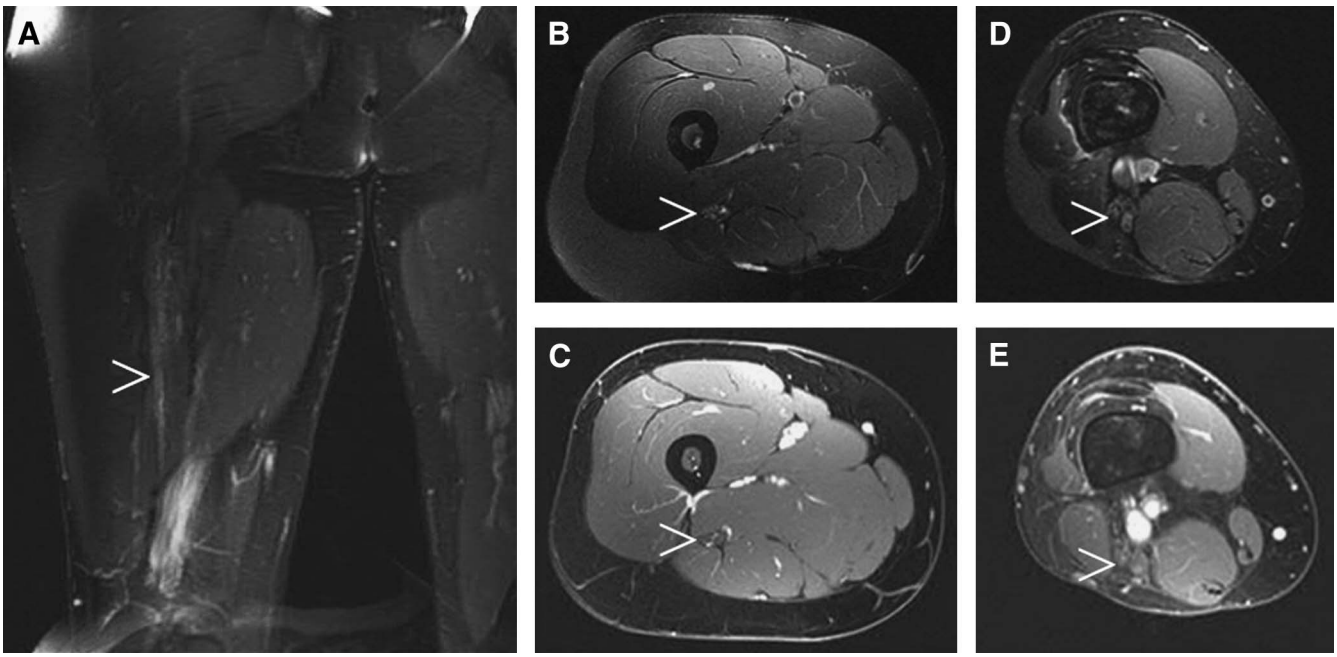

FIGURE 4. *MRI appearance of AVM of the sciatic bifurcation (case 2).* **A**, *coronal T2;* **B**, *axial T2 with fat suppression;* **C**, *T1 with fat suppression with gadolinium;* **D**, *T2 with fat suppression; and* **E**, *T1 weighted image (with fat suppression after intravenous gadolinium administration) at the distal thigh. The arrowheads show the sciatic nerve at both levels with diffuse fascicular infiltration by the vascular abnormality and abnormal enhancement. AVM, arteriovenous malformation.*

CASE REPORTS

Case 1: Venous and Cavernous Angioma

A 23-year-old man presented with left hamstring and knee pain, which had had its onset 11 years before. The quality of the pain was sharp, burning, localized, waxing and waning, and was aggravated by palpation or direct pressure, as in sitting. Furthermore, he reported left-calf pain, which had begun 1 year before and was worsened by stretching of the hamstrings. The past medical history included an anterior cruciate ligament tear in 2002, which had been arthrosopically repaired in 2005. The physical examination was unremarkable aside from nonradiating pain upon percussion of the distal thigh. His sensory and motor examinations were unremarkable. A MRI scan of the left knee was unremarkable, but imaging of the distal sciatic nerve in the distal thigh showed an enhancing lesion resembling a nerve sheath tumor (Figure 1). High-resolution MRI examination of the left lower thigh and upper calf, both with and without gadolinium administration, demonstrated focal fusiform enlargement of the sciatic nerve over a 3.5-cm segment. It featured increased T2 signal and was accompanied by mild diffuse enhancement involving the distal segments of the sciatic nerve and most proximal portions of the tibial and peroneal nerves (Figure 1). Electromyography (EMG) showed no evidence of sciatic neuropathy or lumbosacral radiculopathy.

External and limited internal neurolysis of the sciatic nerve and disconnection of the sciatic nerve varix were performed (Figure 2). The intraoperative features of the lesion were impressive, consisting of a blue discoloration between nerve fascicles. Pathological examination indicated the presence of a mixed venous and cavernous angioma within epineurium (Figure 3). The patient noted worsening of his pain postoperatively for 1 month; however, at 6 month follow-up his pain had completely resolved. Full sensory and motor examination demonstrated no deficit and there was no pain to palpation over the surgical site. An unsolicited letter at 2 years postoperatively reported no pain in the left leg. MRI done at that time revealed a smaller vascular portion and decreased T2 hyperintensity in the sciatic nerve. There were no denervation changes in the muscles.

Case 2: AVM

A 33-year-old woman presented with persistent right-sided, radiating leg pain. It began 4 years previously during pregnancy, affected mainly the right calf, and worsened during menses. In the course of the disease, pain became more debilitating and constant. Its quality was that of deep throbbing associated with allodynia over the calf when pain was intense. The pain was associated with mild swelling and discoloration of the distal leg. Physical examination revealed tenderness to percussion in the right lower limb, the maximal point being the popliteal fossa and distal third of the thigh, and radiating paresthesias in a sciatic distribution from the lower buttock toward the foot. Sensorimotor functions were normal. An ankle jerk was present. Slight discoloration was noted on the medial aspect of the distal leg. An MRI scan demonstrated enlargement and T2 hyperintensity of the distal sciatic nerve including both the tibial and peroneal components with prominent vessels interdigitating between the nerve fascicles (Figure 4). The abnormality was most prominent in the distal thigh but extended

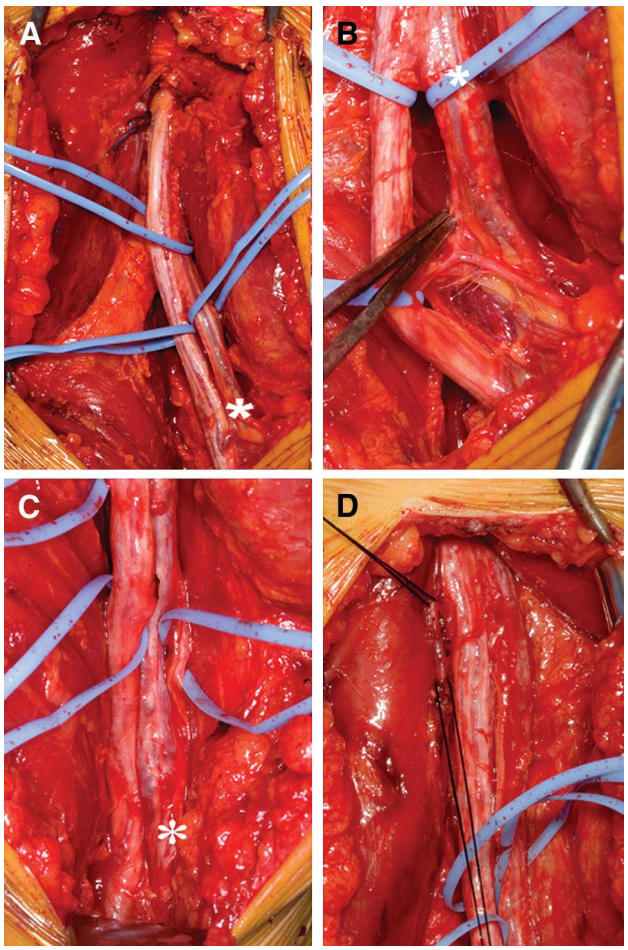

FIGURE 5. *Intraoperative appearance of AVM of the sciatic bifurcation (case 2).* **A**, *intraoperative appearance of the sciatic bifurcation at the popliteal fossa; proximal is top and distal is bottom in all photos. The asterisk indicates the peroneal nerve adjacent to the tibial nerve. Note vascular structures running parallel to nerve fascicles. Blue in color, they likely represent draining veins.* **B**, *arterial structure within the perineurium and related to the lesion.* **C**, *operative field after the proximal divisions of the sciatic nerve have been separated.* **D**, *silk stitches indicate a large, extraneural draining vein. AVM, arteriovenous malformation.*

proximally as well as distally toward the popliteal fossa. An EMG showed no abnormality.

Surgical exploration with external neurolysis as well as limited internal neurolysis of the sciatic nerve and its terminal branches was undertaken (Figure 5). Prominent vascularity suggestive of a vascular malformation was apparent at the sciatic bifurcation (Figure 5). Engorged, purple veins as well as small arterial feeders were noted, in particular, affecting the peroneal and, to a lesser degree, the tibial nerve fascicles (Figure 5). One prominent draining vein adjacent to peroneal fascicles was resected (Figure 5). Histological examination revealed the lesion to be an AVM (Figure 6). At 6 month follow-up and 1 year follow-up, the patient's pain had improved. However, her pain still occurs intermittently associated with her menstrual cycle. She continues to use over-the-counter non-steroidal anti-inflammatory agents for pain control on an as-needed basis. Her motor and sensory examinations were normal at 6 month follow-up.

Case 3: Venous Malformation in Klippel-Trenaunay Syndrome

A 36-year-old man presented with recurrent, short-duration, stabbing pain in the sciatic distribution, affecting the left lower extremity. Lasting a few minutes to an hour, it extended from the buttock along the thigh, popliteal fossa, and leg to the sole of his foot. At age 10, a diagnosis of KTS was made. At age 17 and 23, he underwent surgeries to remove vascular malformations of the left leg to decompress the sciatic nerve and its terminal branches. The first surgery provided symptomatic relief, but upon recurrence of symptoms, a second unsuccessful attempt was made. Over 12 years, the pain gradually worsened. On physical examination, his left lower extremity was found to be significantly shorter than the right. In addition, flexion contracture in the knee was noted as was an equinovarus deformity of the foot. Also of note were abnormal vasculature of plantar aspect of the fourth toe, gigantism of the fourth and fifth toes with local overlap, diffuse atrophy of the muscles of the left leg, mild weakness of hip flexors, gluteus maximus muscle, and foot intrinsic muscles, and moderate to severe weakness of hamstrings. Absence of the deep tendon reflex was noted in the left ankle. Aside from decreased perception of light touch along the lateral side of the calf and foot, sensation was unremarkable. Tenderness to percussion was worse in the popliteal fossa and the region of the ischial tuberosity region than in the buttock and distally. An MRI scan of the pelvis and lower extremity showed vascular enhancement within and around the sciatic nerve, in particular, at the level of the ischium and sciatic bifurcation (Figure 7). The nerve itself was mildly enlarged and showed T2 hyperintensity at this level. The results of an EMG were consistent with chronic changes of sciatic neuropathy.

The patient underwent surgical exploration and external neurolysis of the sciatic nerve in the entire thigh and popliteal fossa. Internal neurolysis was performed at points of maximal tenderness noted preoperatively. At the time of surgery, vascular involvement was extensive (Figure 8). Histological examination was consistent with a venous malformation (Figure 9). Following surgical exploration, the patient reported significant decrease of pain, in particular, in the buttocks. Given the chronic nature of KTS, it is difficult to assess the effect of the most recent surgery. Although the patient experienced marked improvement of symptoms immediately after surgery and at 6 months follow-up, neuropathic pain persisted and required a reduced dose of gabapentin and oxycodone at 1 year. His neurological examination was unchanged compared with preoperatively at 6 months follow-up.

Case 4: Capillary Hemangioma

An 18-year-old woman presented with paresthesias and pain in the left lower extremity. Symptoms had been noted for 2 months before presentation and were intermittent at onset but constant later in the course. She denied radiation of pain, numbness, or focal weakness. Maximal pain was noted over the medial aspect of the knee and in the thigh.

The past medical history was remarkable for primary biliary atresia, which had been treated by liver transplantation during infancy and continued immunosuppression throughout much of her life. At the time of presentation, the patient was awaiting a second liver transplant. Physical examination revealed nonradiating pain upon palpation and

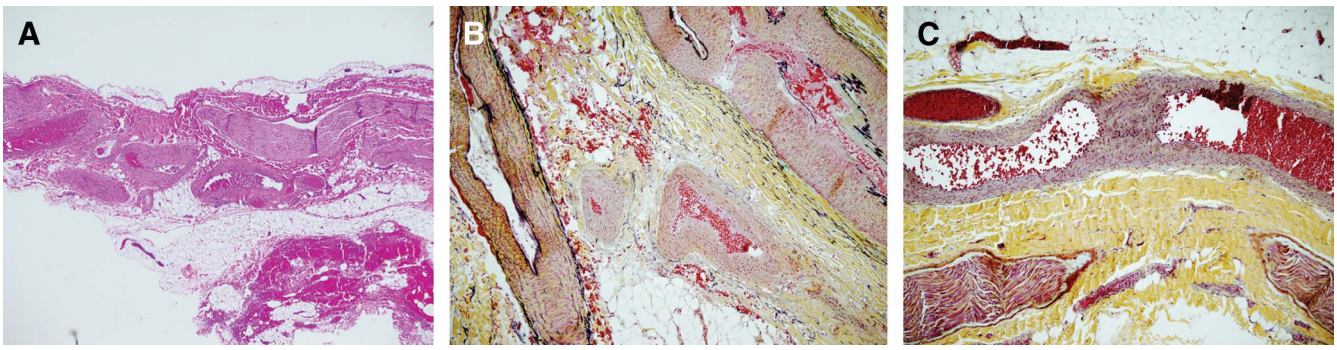

FIGURE 6. *Histology of AVM of the sciatic bifurcation (case 2). Arteriovenous malformation, note the abundance of vessels within the epineurium (A) (hematoxylin and eosin). The Movat preparations show sizable arteries and smaller veins (B) as well as a large vein accompanying a nerve fascicle (C). AVM, arteriovenous malformation.*

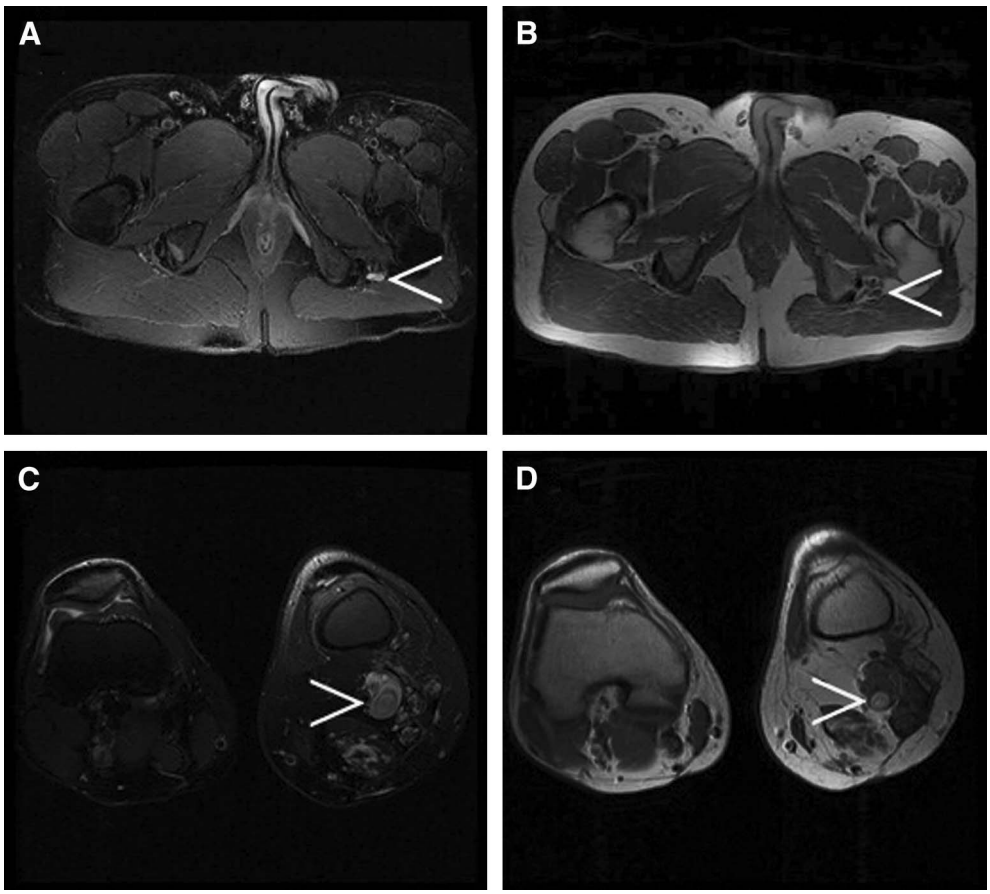

FIGURE 7. *MRI appearance of venous malformation of the sciatic nerve and bifurcation in KTS (case 3). A, axial T2 with fat suppression and (B) axial T1 with fat suppression after intravenous gadolinium administration in MR images of the lower pelvis. C, axial T2 with fat suppression and (D) axial T1 weighted images after intravenous gadolinium at the distal thigh. The arrowheads in each instance show the sciatic nerve with diffuse fascicular infiltration by the vascular abnormality and abnormal enhancement. KTS, Klippel-Trenaunay syndrome.*

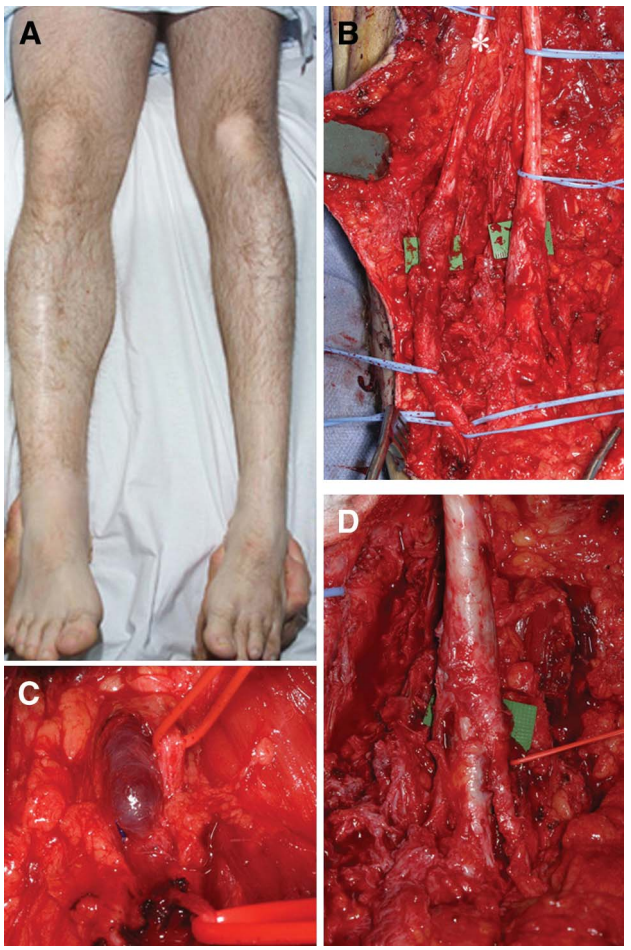

FIGURE 8. *Intraoperative appearance of a venous malformation of the sciatic bifurcation in KTS (case 3).* **A**, *atrophic appearance of left lower limb.* **B**, *intraoperative appearance of the sciatic bifurcation, the proximal is top and distal is bottom in all intraoperative photos. Asterisk indicates the peroneal nerve adjacent to the tibial nerve. Note the tibial and peroneal nerves appear abnormal at the popliteal fossa. Green backgrounds centered at bluish discoloration in the peroneal (*) and tibial nerves consistent with the venous malformation.* **C**, *vein running in the perineurium related to the lesion in the tibial nerve.* **D**, *high magnification view of the tibial nerve, green background and red tape is around the vein noted within the popliteal fossa. KTS, Klippel-Trenaunay syndrome.*

percussion of the left thigh. Weakness of the gastrocnemius, posterior tibialis, and flexor digitorum muscles were noted, as was hypersensitivity of the plantar aspect of the foot. The sensory examination was unremarkable, and reflexes were symmetric. MRI scan identified a lesion in her left sciatic nerve predominantly affecting the tibial portion of uncertain etiology (Figure 10) but with some features suggesting slow flow within a venous vascular structure, especially at the level of the sciatic bifurcation. EMG was normal without evidence for sciatic or tibial neuropathy.

Surgical exploration was undertaken. At the time of surgery, beneath the gluteal sling and approximately 10 cm below the greater trochanter, an enlarged, pink sciatic nerve featured hypervascularity of the tibial division (Figure 11). Limited internal neurolysis was performed. Pathological evaluation demonstrated a capillary hemangioma (Figure 12). At 6 months follow-up, her pain was relieved and her motor function remained stable. Phone follow-up at 18 months revealed that the patient had significant decrement in her pain. She still described occasional pain events which were not comparable with the preoperative pain.

DISCUSSION

Although the vast majority of sciatica is due to degenerative causes and is radicular in nature, various other causes are to be considered when spine imaging is negative. Sciatica has been reported, although rarely, to be related to vascular lesions along the course of the sciatic nerve from the pelvis to its bifurcation.[3,21,24] Other vascular lesions discussed as causes of sciatica are anatomic anomalies such as a sciatic artery or vein, gluteal varicoses, venous thrombosis, hemangiomatosis in KTS, and venous or capillary hemangioma.[1,3,5,25-30] Vascular causes of peripheral neuropathy are extremely uncommon and require a focused workup. The present series included 4 patients with pain in a sciatic nerve distribution accumulated over a period of 4 years. All had presented with symptoms suggesting spinal etiologies, but investigations of the spine were unremarkable in every instance. A thorough physical examination, in particular, the reproduction of neuropathic pain by percussion along the course of the sciatic nerve as well as high-resolution 3-Tesla MRI of peripheral nerve were conducted to identify the origin of the pain. Features suggesting lesions of the nerve on MRI scans included diffuse T2 and T1 changes along with abnormal enhancement with gadolinium administration. Flow-related enhancement was present on T2 weighted fast-spin echo images in and around the affected nerves consistent with the dilated blood vessels of vascular malformations in all cases. Electromyographic studies performed in all patients revealed no abnormality aside from chronic changes in the one patient with KTS (who had previous operations on the sciatic nerve).

In this small series, external neurolysis and limited microvascular dissection of affected nerve fascicles led to satisfactory relief of symptoms. Although only 1 of the 4 patients is currently pain free, the remaining 3 are improved in their opinion compared with their preoperative status. This type of surgical approach and the outcomes achieved are consistent with the few cases reported in literature (Table).[3,15,19,20] The most extensive data, including treatment success and long-term follow-up, is that regarding hemangioma of the median nerve mimicking carpal tunnel syndrome which seems to respond well to decompression and internal neurolysis.[16] Unfortunately, rare reports of AVM associated with the sciatic nerve differ in terms of treatment, involving mainly endovascular embolization, and lack sufficient follow-up.[7,24] Nonetheless, resection of one AVM was highly successful.[21] Di Iorio et al[13] reported on vascular lesions

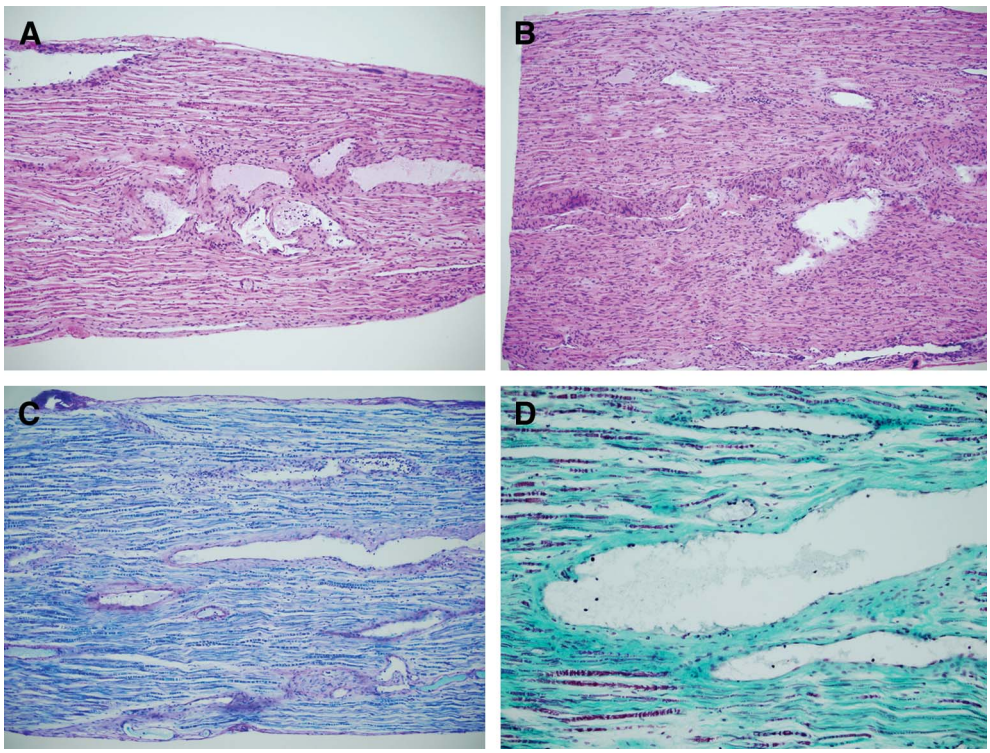

FIGURE 9. *Histology of a venous malformation of the sciatic bifurcation in KTS (case 3). The biopsy, encompassed by epineurial tissue, shows numerous thin-walled but media-containing veins within the endoneurium (**A** and **B**, hematoxylin and eosin). The veins within the endoneurium are further highlighted on Luxol-fast blue (**C**) and Masson trichrome stain (**D**). KTS, Klippel-Trenaunay syndrome.*

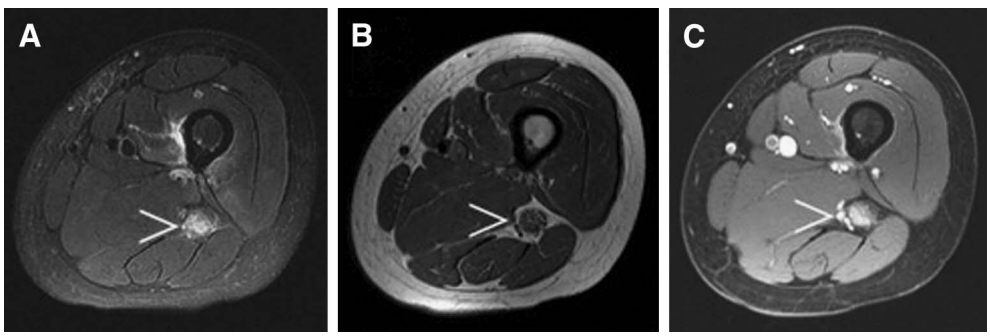

FIGURE 10. *MRI appearance of a capillary hemangioma of the sciatic nerve (case 4). **A**, axial T2 weighted image with fat suppression. **B**, axial T1 weighted image. **C**, axial T1 with fat suppression after gadolinium of the distal thigh. The arrowhead in each image shows the tibial division with fascicular infiltration by the vascular abnormality with abnormal enhancement. Note sparing of the peroneal nerve.*

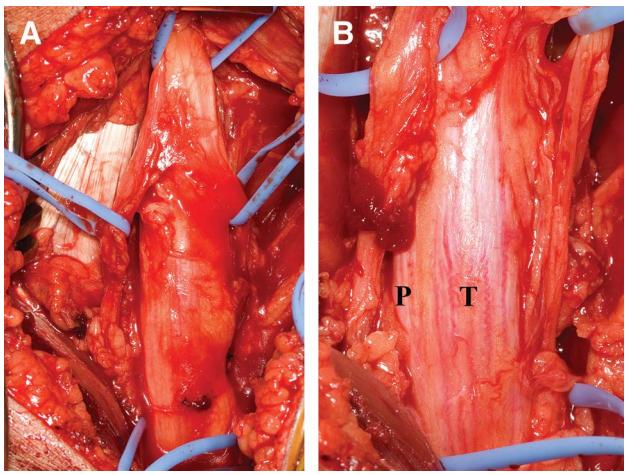

FIGURE 11. *Intraoperative appearance of a capillary hemangioma of the tibial division of the sciatic nerve (case 4).* **A**, *isolation of the sciatic nerve proximal to the bifurcation shows no obvious abnormality.* **B**, *upon opening the epineurium, fine pink streaking corresponding to the capillary hemangioma is well seen restricted to the tibial division (T). Limited internal neurolysis was then performed. Peroneal division (P).*

associated with peripheral nerves in the setting of KTS; however, the follow-up data in this publication are insufficient. Detailed follow-up data are needed in all cases to determine whether external and limited internal neurolysis results in long-term relief without further loss of neurological function and satisfactory pain control.

The collective experience with vascular lesions as a cause of sciatica is limited to a small number of case reports. Collection of these rare cases through literature report at this time appears to be the only viable option in understanding the pathophysiology, natural history, and treatment of these lesions.

Disclosure

The authors have no personal financial or institutional interest in any of the drugs, materials, or devices described in this article.

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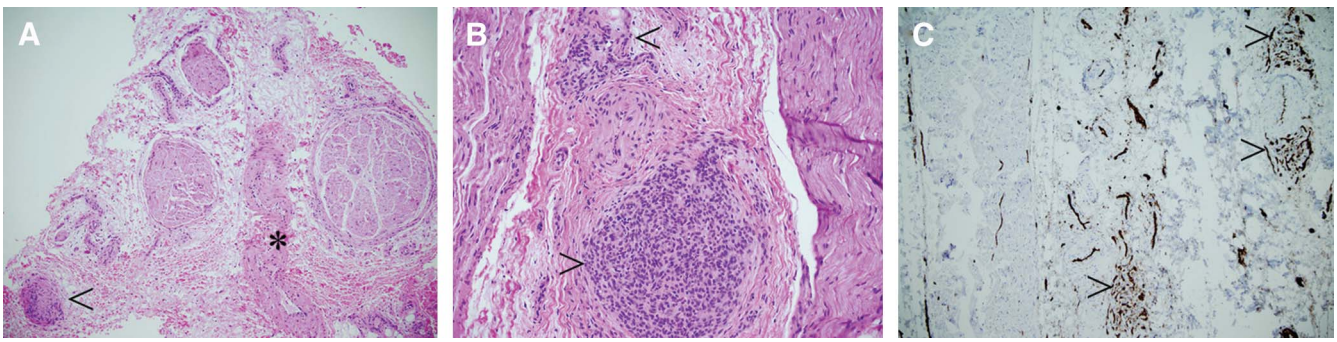

FIGURE 12. *Histology of a capillary hemangioma of the tibial nerve (case 4).* **A** *and* **B**, *capillary hemangioma, the fascicles are associated with a sizable vein (***A**, *asterisk (*) and tufts of densely compacted capillaries (***A** *and* **B**, *arrowheads, hematoxylin and eosin). Note their position between two nerve fascicles (***B**, *arrowheads). A CD31 stain highlights several of these capillary complexes (***C**, *arrowheads)*

TABLE. Literature Reported Cases of Vascular Lesions and Sciatica[a]

Patient	Age, y	Sex	Symptoms	Signs	Pathology	Treatment	Outcome
A	10	M	R leg p, weakness	R leg weakness, percussion tenderness at site of the nerve lesion	Hemangioma	Resection of nerve segment, leg amputation	Pain resolved, loss of leg
B	5	F	L leg p	L leg weakness	Hemangioma	Internal neurolysis	Pain resolved
C	53	M	R leg p, tenesmus and rectal p	None	AVM	Internal neurolysis	Pain resolved
1	23	M	L leg p	None	Hemangioma	Internal neurolysis	Pain resolved
2	33	F	R leg p	Swelling, discoloration	AVM	Internal neurolysis	Pain improved
3	36	M	L leg p, weakness	Atrophy, weakness	Venous malformation	Internal neurolysis	Pain improved
4	18	F	L leg p	None	Hemangioma	Internal neurolysis	Pain improved

[a]A, Stewart and Bettin[24]; B, Purcel and Gurdjian[22]; C, Vos et al[12]; R, right; L, left; p, pain; AVM, arteriovenous malformation.

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COMMENTS

The patient with spinal imaging negative ''sciatica,'' particularly if neuropathic pain is present, is challenging. Most surgeons have found that sciatic nerve imaging is usually not productive. Gompel et al, however, have demonstrated otherwise. They identified 4 cases of vascular malformations of the sciatic nerve. Preconceived notions in this regard, therefore, may not always be appropriate. The character of the pain and neurological examination may provide clues regarding such a diagnosis. Obviously, however, with such clues, magnetic resonance imaging (MRI) must be ordered. Further sciatic nerve imaging may be in order in selected cases. Regardless, the authors are to be congratulated for their observations and clinical astuteness. They have unquestionably heightened our awareness.

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Dr Gompel and his colleagues present 4 cases in which a patient is found to have a vascular malformation causing radiating leg pain from sciatic nerve involvement. Each case includes a thorough preoperative evaluation including imaging, a complete discussion of the external and limited internal neurolysis performed to treat the malformation, and at least 6 months of follow-up data. The surgical approach of limited internal neurolysis was shown to improve pain in venous angioma, arteriovenous malformation, Klippel-Trenaunay syndrome, and capillary hemangioma.

This type of report is of great value in the literature for this rare condition and will be an aid in determining the treatment of sciatica caused by various vascular malformations. The authors present the cases in a complete and thoughtful manner that will be increasingly helpful as more similar cases are identified and reported.

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This is an interesting and well-written case series in which the authors describe 4 patients with sciatica caused by vascular lesions. These cases serve as an important reminder to clinicians that thorough clinical and imaging evaluations are needed in cases of sciatica that are not readily explained by lumbar MRI studies. These patients presented with some clinical features that were not typical of sciatica from the usual discogenic origin, for example, percussion tenderness over the lesion in the thigh, progression of symptoms to constant pain even in recumbency, discoloration of the leg with swelling, exacerbation of pain during menses, etc. The latter cyclical feature in case 2 was reminiscent of endometriosis, another uncommon cause of nondiscogenic sciatica. [1] We have also treated several vascular lesions presenting as nerve sheath tumors in the brachial plexus,[2] and these cases have taught us to be wary of possible severe hemorrhage when approaching these lesions surgically. Preoperative angiography and embolization may be essential in some of the high-flow lesions.

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