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Cystic Adventitial Disease of the Tibial Vein Arising From the Subtalar Joint: A Case Report

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ABSTRACT

Soft tissue ganglion cysts are a well-known cause of tibial nerve compression in the tarsal tunnel. We describe a patient who presented with tibial nerve symptoms and was found to have an adventitial cyst of the tibial vein arising from the subtalar joint, with the joint connection confirmed both on imaging and at surgery. Surgical decompression of the cyst with transection of the vascular pedicle arising from the subtalar joint improved her symptoms at 6 months, and postoperative magnetic resonance imaging showed resolution of the cyst. Cystic adventitial disease is a rare, poorly understood condition in which a cyst is identified in the adventitia of a vessel, usually an artery. Only 3 cases of adventitial cysts have been reported in the foot and ankle region, 2 in the lesser and 1 in the greater saphenous vein. None of the previous cases have been recognized to be joint connected. This case provides additional evidence for an articular origin for adventitial cysts and helps guide management strategies for these joint-connected cysts.

An intraneural ganglion cyst arises from a pathologic joint when synovial fluid escapes the joint and uses an articular branch of the nerve as a conduit. The cyst is found within the epineurium (1–9). When synovial fluid escapes into the tissue surrounding the joint (rather than entering a nerve conduit), an extraneural, soft tissue ganglion cyst forms instead. A third, less appreciated type of ganglion cyst exists as well, known as an adventitial ganglion cyst. This type of ganglion cyst occurs in the adventitia of a blood vessel, most commonly an artery, but occasionally a vein.

Ganglion cysts are the most frequent cause of tarsal tunnel syndrome (10). Tibial nerve symptoms result from compression of the tibial nerve by the space-occupying ganglion cyst in the confines of the tarsal tunnel, formed by the flexor retinaculum posterior and inferior to the medial malleolus of the ankle (11,12). These cysts can compress the tibial nerve proper or the medial or lateral plantar nerves. All 3 types of ganglion cysts have been reported in the foot and ankle region, including only 3 cases of adventitial cysts (12–15). In the previously reported cases of foot and ankle adventitial cysts, a joint connection was not recognized. We have previously proposed that adventitial cysts arise by the same mechanism as intraneural ganglion cysts, but use an articular/capsular vessel as the conduit rather than an articular nerve branch (16). The adventitia in vessels is analogous to the epineurium of nerves.

We discuss an unusual presentation and type of a ganglion cyst. A patient who had tibial nerve symptoms was found to have a cyst near the tibial nerve in the tarsal tunnel. At surgery, an adventitial cyst of the tibial vein was identified with a connection to the subtalar joint. Retrospective review of the magnetic resonance imaging (MRI) studies confirmed the adventitial cyst of the tibial vein and identified the articular vessel that served as the conduit from the subtalar joint. This case adds to the evidence supporting the unified articular theory and supports the joint origin of cystic adventitial disease (7,16).

Case Report

Clinical Presentation

A female, aged 35 years, presented for the evaluation of a painful mass in her medial right ankle. She reported a 2-year history of fullness in the area, with progressive tenderness, as well as tingling and burning paresthesias radiating to her forefoot. She had subjective numbness in the lateral 2 toes, as well as hypersensitivity and dysesthesias.

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Examination revealed a fluctuant mass in the right medial ankle at the level of the tarsal tunnel and tenderness to percussion with radiating paresthesias into the lateral 2 toes. Motor examination was normal, with full range of motion of the ankle and subtalar joints, and she had a normal gait.

Radiographs of the foot revealed mild soft tissue swelling about the midfoot and hindfoot, without any osseous changes. MRI of the foot showed a septated ganglion cyst adjacent to the tibial nerve, arising from the subtalar joint (Fig. 1).

Treatment

Initial management consisted of ultrasound-guided aspiration of the cyst and injection of 1.5 cc betamethasone (6 mg) and 0.5% ropivacaine. This procedure provided mild, temporary relief and decreased the size of the cyst. Owing to continued symptoms over the ensuing 2 months after the aspiration and injection, operative excision of the ganglion cyst was undertaken.

A tarsal tunnel decompression was performed to allow access to the tibial neurovascular bundle and the ganglion cyst. The medial and lateral plantar nerves were noted to be edematous. Once the nerves were neurolyzed, the main cyst became quite apparent. At the level of the medial malleolus, during mobilization of the vascular bundle, the cyst was identified within the more anteriorly located tibial vein (Fig. 2). The cyst wall was opened sharply and the contents were expressed to decompress the adventitial sheath. The tibial artery and veins were traced distally and ultimately the subtalar origin of the multilobulated cyst was identified. The wide-based stalk was sharply dissected off of the joint capsule and a capsular window was made. Pathology was consistent with an adventitial cyst (Fig. 3).

Postoperative reinterpretation of the MRI by a musculoskeletal radiologist (K.K.A) experienced with adventitial and intraneural ganglion cysts

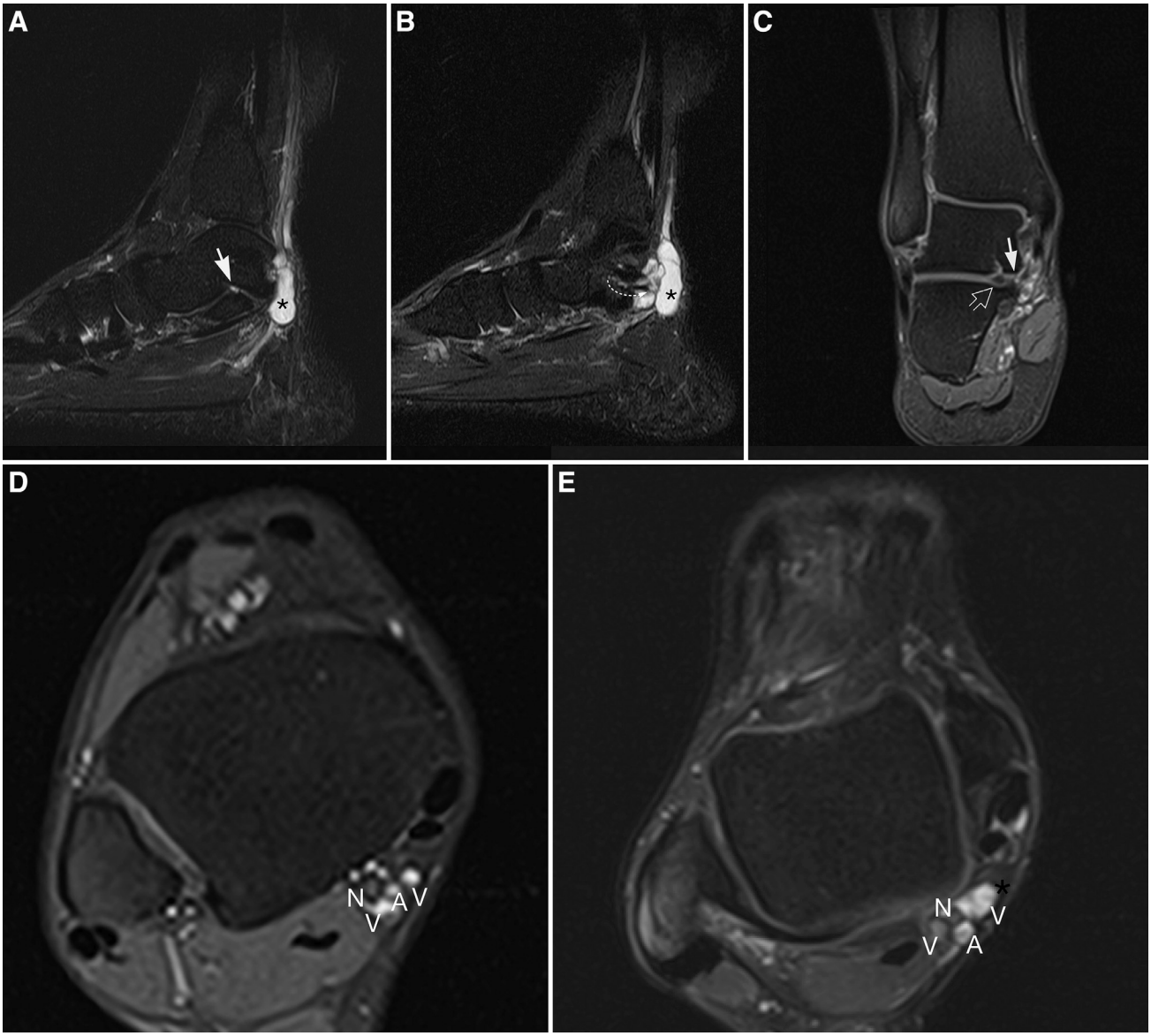

Fig. 1. Magnetic resonance imaging (MRI) features. (*A*, B) Sequential sagittal short tau inversion recovery MRIs of the ankle showing the cyst from the subtalar joint (arrow) extending (dashed arrow) to the primary tubular adventitial cyst (*). (*C*) Coronal T1-weighted MRI with fat suppression after contrast showing a tiny venous articular branch (arrow) at the subtalar joint with nonenhancing cyst (open arrow). (*D*) Axial T1-weighted MRI post contrast with fat saturation shows the normal relationships of the 2 tibial veins (V), and the tibial artery (A) and nerve (N) slightly distal to the tibiotalar joint. (*E*) Axial T2-weighted MRI with fat saturation shows an adventitial cyst associated with the more anteriorly positioned tibial vein (V) at the level of the tibiotalar joint.

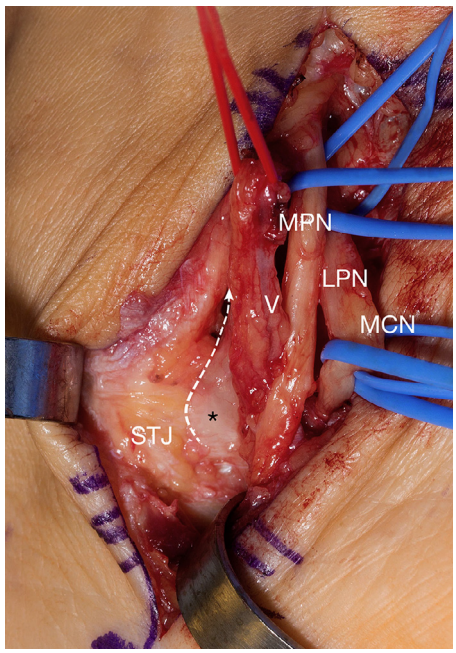

Fig. 2. Operative findings. After cyst decompression, the connection of the cyst is traced to the subtalar joint (STJ). Cyst (*) is seen extending to the vascular bundle (dashed arrows, red vasoloop) containing 2 tibial veins and the tibial artery. The more posteriorly located vein without cyst (V) is seen. Medial (MPN) and lateral plantar (LPN) and medial calcaneal nerves (MCN) are protected (blue vasoloops).

confirmed a tubular adventitial cyst within a tibial vein and the articular vessel to the subtalar joint serving as the origin of the cyst was identified (Fig. 1). The tibial nerve and the medial and lateral plantar nerves showed mild T2 hyperintensity; there was no evidence of muscle denervation.

At 10 months postoperatively, the patient had improved symptoms and resolution of her pain. Postoperative MRI at 3 months was free of recurrent cyst.

Discussion

We present a case of an adventitial cyst of the tibial vein in a patient with symptoms consistent with tarsal tunnel syndrome in which a connection to the subtalar joint via an articular vein was identified both radiologically and at surgery. Cystic adventitial disease is a rare, poorly understood condition in which cyst is identified in the adventitia of a blood vessel. A recent systematic review of 724 patients (729 cysts) revealed the rarity of veins being affected compared with arteries (1:12). In the foot and ankle region, only 3 cases of cystic adventitial disease have been reported, all affecting veins (2 involving the lesser and 1 involving the greater saphenous vein). In all 3 cases, the cyst was located adjacent to the ankle joint, but none had a joint connection identified (13–15). In the case presented by Lie et al (13), the cyst was noted to extend to the calcaneofibular ligament.

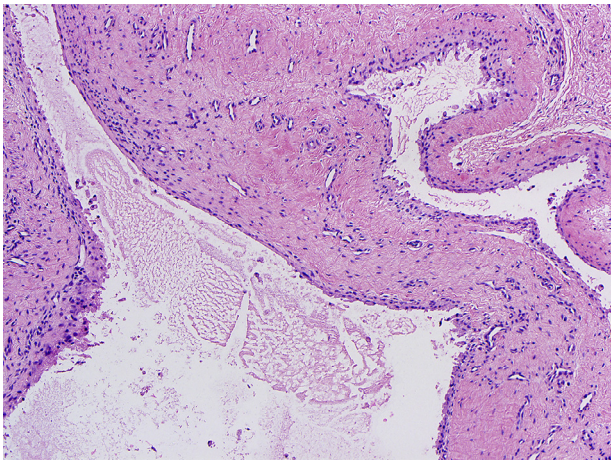

Fig. 3. Histopathology of the adventitial cyst (magnification × 10; hematoxylin and eosin stain).

Our patient presented with nerve symptoms, presumably from a mass effect on the tibial nerve within the confined tarsal tunnel. Similarly, one of the cases of cystic adventitial disease in the greater saphenous vein presented with symptoms owing to compression of the saphenous nerve (15). One reported case of ulnar artery cystic adventitial disease also presented with nerve symptoms, likely owing to compression of the ulnar nerve in Guyon's canal (17). In these cases, including ours, the nerve-related symptoms improved after cyst surgery.

Mounting evidence supports the unified articular (synovial) theory for intraneural and extraneural ganglion cysts, but we believe that adventitial cysts also represent a part of the spectrum of joint-derived ganglion cysts (7,12,16,18). In the case presented herein, the articular vein that served as the conduit for the adventitial cyst was identified both on MRI and at surgery. We believe that in all cases of cystic adventitial disease, if one looks for the joint connection, it will be present. Similar to other ganglion cysts, whether soft tissue (i.e., extraneural) or intraneural, these cysts arise from capsular defects. Synovial fluid can then escape through the capsular defect and dissect from the joint along a capsular vein into the adventitia of the capsular vein or artery (19,20). Although the best treatment is not known, based on this concept, we believe the same principles of treatment used for intraneural ganglion cysts can and should be used for symptomatic adventitial cysts. These principles include careful dissection of the neurovascular structures, decompression and resection of the cyst, and debridement at the affected joint of origin. Addressing the joint connection (pedicle) is the most important step to prevent recurrence. As we have shown, the blood vessel including the cystic lesion does not need to be resected to effect symptom relief or to minimize recurrence risk. Although an interesting case simply for its rarity, the importance of the presented case lies in the clearly demonstrated joint connection and the successful treatment using concepts learned from intraneural ganglion cysts, which together provide additional support for the unified articular theory.

References

1. Desy NM, Wang H, Elshiekh MA, Tanaka S, Choi TW, Howe BM, Spinner RJ. Intraneural ganglion cysts: a systematic review and reinterpretation of the world's literature. J Neurosurg 2016;125:615–630.
2. Hebert-Blouin MN, Tubbs RS, Carmichael SW, Spinner RJ. Hilton's law revisited. Clin Anat 2014;27:548555.
3. Lipinski LJ, Rock MG, Spinner RJ. Peroneal intraneural ganglion cysts at the fibular neck: the layered "U" surgical approach to the articular branch and superior tibiofibular joint. Acta Neurochir (Wien) 2015;157:837–840.
4. Spinner RJ, Amrami KK, Wolanskyj AP, Desy NM, Wang H, Benarroch EE, Skinner JA, Rock MG, Scheithauer BW. Dynamic phases of peroneal and tibial intraneural ganglia formation: a new dimension added to the unifying articular theory. J Neurosurg 2007;107:296–307.
5. Spinner RJ, Atkinson JL, Scheithauer BW, Rock MG, Birch R, Kim TA, Kliot M, Kline DG, Tiel RL. Peroneal intraneural ganglia: the importance of the articular branch. Clinical series.. J Neurosurg 2003;99:319–329.
6. Spinner RJ, Atkinson JL, Tiel RL. Peroneal intraneural ganglia: the importance of the articular branch. A unifying theory. J Neurosurg 2003;99:330–343.
7. Spinner RJ, Desy NM, Amrami KK. The unifying articular (synovial) origin for intraneural ganglion cysts: moving beyond a theory. J Hand Surg 2016;41:e223–e224.
8. Wilson TJ, Amrami KK, Spinner RJ. Intraneural ganglion cysts: a predictable method to their madness. Ann Phys Rehabil Med 2017;60:414–415;.
9. Wilson TJ, Hebert-Blouin MN, Murthy NS, Amrami KK, Spinner RJ. Recognition of peroneal intraneural ganglia in an historical cohort with "negative" MRIs. Acta Neurochir (Wien) 2017;159:925–930.
10. Takakura Y, Kitada C, Sugimoto K, Tanaka Y, Tamai S. Tarsal tunnel syndrome. Causes and results of operative treatment. J Bone Joint Surg Br 1991;73:125–128.

11. Antoniadis G, Scheglmann K. Posterior tarsal tunnel syndrome: diagnosis and treatment. Dtsch Arztebl Int 2008;105:776–781.
12. Kawakatsu M, Ishiko T, Sumiya M. Tarsal tunnel syndrome due to three different types of ganglion during a 12-year period: a case report. J Foot Ankle Surg 2017;56:379–384.
13. Lie JT, Jensen PL, Smith RE. Adventitial cystic disease of the lesser saphenous vein. Arch Pathol Lab Med 1991;115:946–948.
14. Mentha C. [Mucoid degeneration of the veins]. Presse Med 1963;71:2205–2206.
15. Yoshii S, Ikeda K, Murakami H. Cystic, myxomatous adventitial degeneration of a saphenous vein. J Vasc Surg 1998;27:780–782.
16. Desy NM, Spinner RJ. The etiology and management of cystic adventitial disease. J Vasc Surg 2014;60:235–245.
17. Pivec C, Sillat T, Moritz T, Riegler G, Nanobachvili J, Bodner G. A rare case of Guyon's canal syndrome caused by cystic adventitia degeneration: high-resolution ultrasound findings. Ultraschall Med 2017;38:556–557.
18. Zuckerman SL, Spinner RJ. Understanding the dynamics and compartments in joint-related ganglion cysts. J Foot Ankle Surg 2017;56:415–416.
19. Spinner RJ, Desy NM, Agarwal G, Pawlina W, Kalra M, Amrami KK. Evidence to support that adventitial cysts, analogous to intraneural ganglion cysts, are also joint-connected. Clin Anat 2013;26:267–281.
20. Wilson TJ, Hebert-Blouin MN, Murthy NS, Garcia JJ, Amrami KK, Spinner RJ. The nearly invisible intraneural cyst: a new and emerging part of the spectrum. Neurosurg Focus 2017;42:E10.