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A Case Report and Literature Review: Intraneural Ganglion Cyst Causing Tarsal Tunnel Syndrome

Alison Migonis, DPM[1], Raymond Murano, Jr., DPM[2,3], Isaac E. Stillman, MD[4,5], Matthew Iorio, MD[6,7], John M. Giurini, DPM[8,9]

[1] *Attending, Central Vermont Medical Center, Department of Orthopedics, Berlin, VT*
[2] *Chief Resident, Division of Podiatric Surgery, Beth Israel Deaconess Medical Center, Boston, MA*
[3] *Clinical Fellow in Surgery, Harvard Medical School, Boston, MA*
[4] *Attending, Department of Pathology, Beth Israel Deaconess Medical Center, Boston, MA*
[5] *Associate Professor in Pathology, Harvard Medical School, Boston, MA*
[6] *Co-Director, Extremity Microsurgical Reconstruction, University of Colorado, Anschutz Medical Campus, Aurora, CO*
[7] *Associate Professor in Surgery, University of Colorado, Aurora, CO*
[8] *Associate Professor in Surgery, Harvard Medical School, Boston, MA*
[9] *Chief, Division of Podiatric Surgery, Beth Israel Deaconess Medical Center, Boston, MA*

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ABSTRACT

Intraneural ganglion cysts are benign mucinous lesions that form within joints and enter adjacent nerves via an articular branch. Despite being morphologically characterized as benign, they can demonstrate considerable intrafascicular destruction and expansion, resulting in worsening compressive neuropathies or nerve injury. There have been several suggested theories of pathogenesis, but the most widely accepted articular (synovial) theory describes a capsular defect in a neighboring joint that allows joint fluid to egress and track along the epineurium of the innervating articular branch. In this case report, we describe an intraneural ganglionic cyst located in the tarsal tunnel with extensive involvement of the tibial nerve. We describe the symptoms, diagnosis, and treatment as well as review the current literature on intraneural ganglionic cysts.

Intraneural tumors can represent a considerable source of patient morbidity and pain, especially in the instance of nerve resection and persistent paresthesia or motor deficits. Although they are benign tumors, ganglion cysts can be prone to intraneural expansion and scarring, which can create a wide field of injury. In zones of anatomic nerve entrapment, such as the tarsal tunnel, this may be secondary to the increased volume within a confined space of limited compliance, thereby accelerating symptoms of compression and ischemia. From a treatment standpoint, they can be managed surgically by either decompression or complete excision. In the case reported here, given the intrafascicular extension of the ganglion with ongoing nerve compression as well as higher rates of tumor recurrence following ganglion decompression throughout the body, we proceeded with an intrafascicular dissection to protect the individual branches and provide a complete tumor excision (1). The most common site of intraneural ganglion cyst occurrence is at the level of the fibular neck involving the peroneal nerve. In a review article, 60.6% of all intraneural ganglion cysts are found in the common peroneal nerve, and only 3.1% are found involving the tibial nerve (2).

In this case report, we describe a case of an intraneural ganglion cyst involving the tibial nerve within the tarsal tunnel. Given the relative infrequency of the diagnosis and the potential for delay in proper diagnosis and treatment, we review the details of the case and the available literature for appropriate management and related surgical outcomes of intraneural ganglion cysts.

Case Report

A 56-year-old healthy male presented for evaluation of left foot and ankle pain of 1-year duration. He indicated that the pain had gotten progressively worse over the prior 6 months and had been especially limiting over the past 2 months. He stated that pain was present throughout the day with standing and walking but was particularly bothersome at night when trying to sleep, causing him to sleep with his legs hanging over the side of the bed and feet on the floor to alleviate the pain. He described the pain as burning in nature, starting at the level of the ankle and radiating to the toes. He had also noted numbness along the lateral aspect of the sole of his left foot. He denied any overt

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Address correspondence to: John M. Giurini, DPM, Chief, Division of Podiatric Surgery, Beth Israel Deaconess Medical Center, One Deaconess Road, Boston, MA 02215.
E-mail address: jgiurini@bidmc.harvard.edu (J.M. Giurini).

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injury or trauma to the foot. He had noted no redness or bruising but did describe a feeling of fullness around the ankle. Previous treatment had consisted of physical therapy and acupuncture without relief. He was also treated for gout and received an intra-articular injection of the left ankle, also without improvement.

His medical history was unremarkable; his surgical history, however, was remarkable for arthroscopic knee surgery. He was prescribed gabapentin for his foot pain, which he took at night. He related that it did help slightly with the pain. He was extremely active, participating in running several times per week, hiking, and cycling, and worked as a motivational speaker, which required travel and standing for hours at a time several times per month.

On examination, he had strongly palpable pulses at the level of the dorsalis pedis and posterior tibial arteries bilaterally. His dermatological examination showed no edema, erythema, or ecchymosis of the left foot or ankle. On musculoskeletal examination, he was found to have full range of motion of the ankle joint and subtalar joint without crepitus. He had +5/5 muscle strength in all 4 muscle groups. His stance evaluation showed a mild collapse of the medial longitudinal arch with mild excessive pronation. His neurologic exam showed intact vibratory sensation, but there was decreased sharp/dull sensation along the plantar aspect of his left foot along the distribution of the lateral plantar nerve. He had a positive Tinel's sign posterior to the medial malleolus with radiation to the toes.

Diagnostic Imaging

Multiplanar magnetic resonance (MR) images of the left ankle were performed with administration of intravenous contrast using a mass/infection MR ankle protocol. A 2.1 × 1.7 × 4.4-cm soft tissue mass was found to abut and efface the tibial nerve in the proximal tarsal tunnel between the flexor hallucis longus and tibialis posterior tendon (Fig. 1 *A* and *B*). The mass was well circumscribed, with multilobulated margins and multiple nonenhancing internal septations. However, the site of origin of the mass could not be entirely determined on imaging. On short T1 inversion recovery (STIR) images, the mass demonstrated a hyperintense signal and was iso- to hypointense to skeletal muscle on T1 fat-saturated images. There was no significant enhancement after contrast administration. The tibial nerve was seen above and below the mass and appeared to be thickened, with possible eccentric displacement of the fascicles by the cyst.

On coronal STIR and T1 fat-saturated postcontrast images, there appeared to be a tail extending from the mass into the tarsal tunnel, although no definite articular connection was identified (Fig. 2). The distal edges of the lesion lay at the level of the tibiotalar joint and posterior talus, with the bulk of the mass lying at the level of the distal tibia (Figs. 3 and 4).

Surgical Intervention

With the patient under general anesthesia and the use of a thigh tourniquet, a curvilinear incision was made extending from the distal third of the leg posterior to the medial malleolus to the porta pedis. Through a combination of blunt and sharp dissection, the flexor retinaculum was identified and transected, allowing identification and mobilization of the neurovascular bundle. A bulbous structure was identified in close proximity to the tibial nerve distally. Using loupe magnification, the epineurium was split, and longitudinal strands of cystic material were found enveloped within fibers of the tibial nerve. Cystic material was carefully dissected away from nerve fascicles. Dissection was carried distally, where there was noted to be extension into the medial subtalar joint. A small portion of the joint capsule was removed with the entire cyst and sent as a specimen for pathological analysis. The cyst was then followed proximally along the course of the tibial nerve. The cyst was separated from the nerve at this level and submitted to pathology in toto. Because of the resection of the epineurium and intrafascicular dissection, a Neuragen Neurowrap (Integra Life Sciences Corp., Plainsboro, NJ) was placed around the tibial nerve to prevent adhesions to the tendons or soft tissue of the medial ankle and allow normal nerve gliding (3–6). A nerve protector was placed over the nerve just before

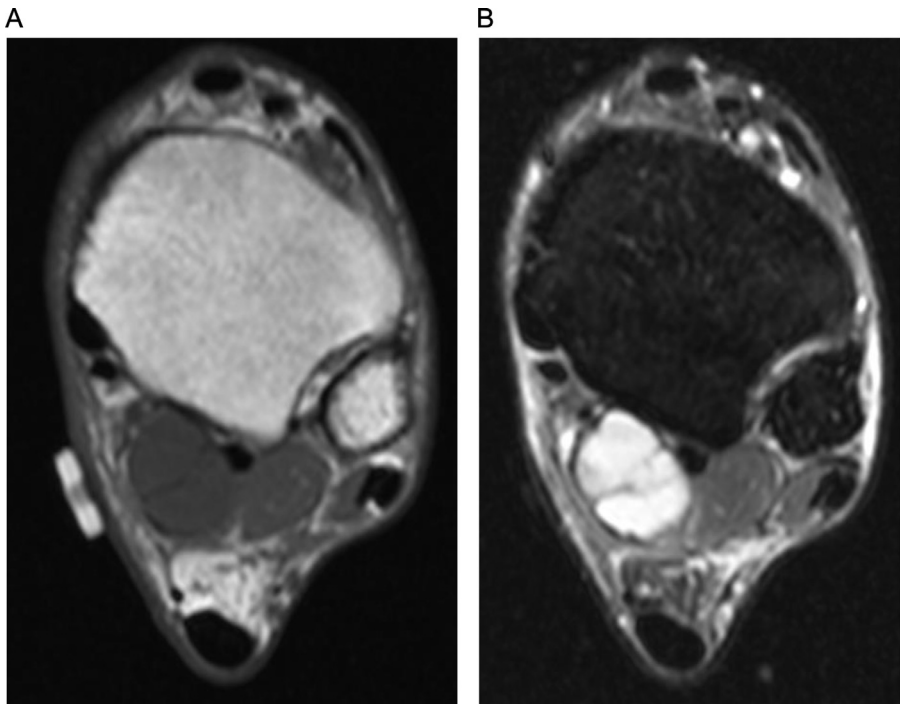

Fig. 1. A well-circumscribed, multi-lobulated mass is seen on the axial T1 (*A*) and STIR (*B*) images between the flexor hallucis longus and posterior tibial tendons. Note the multiple septations on the STIR image (*B*).

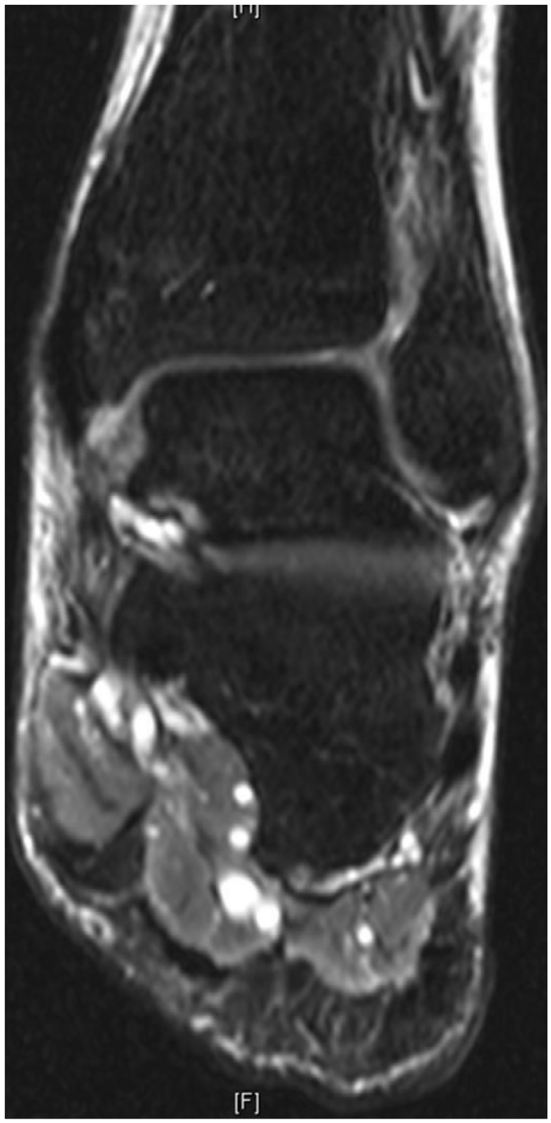

Fig. 2. The coronal STIR image shows a possible connection of the cyst with the subtalar joint. However, this was not deemed definitive on any view.

the tibial nerve branching into medial and lateral bundles and held in place with fibrin glue (Figs. 5–8).

Postoperatively, the patient was placed in an long legged plaster splint and maintained without weightbearing to allow for primary healing of the incision. He was advanced to a controlled ankle movement boot after 2 weeks and allowed to begin partial weightbearing. He was transitioned to full weightbearing at 4 weeks and allowed to begin full range of motion of the ankle with light exercise. He completed a course of physical therapy and returned to full activity at 2 months (7–10).

The patient is 2 years postoperative from his procedure and has returned to his presurgical activities including running, hiking, and motivational speaking. He no longer experiences pain in his foot during either the day or night and is completely off all medications, specifically gabapentin. He maintains +5/5 muscle strength in all 4 muscle groups. His only complaint is of slight residual numbness on the plantar aspect of his foot.

Pathology

The specimen consisted of multiple fragments of soft tissue aggregating to $3.6 \times 3.0 \times 1.0$ cm, which were entirely submitted for microscopic evaluation. An apparently multiloculated cyst (devoid of a synovial lining) composed of a dense collagenized fibrous wall with foci of myxoid change and intraluminal mucoid material, diagnostic of a ganglion cyst, was identified (Fig. 9). Adjacent to the cyst was fibrofatty tissue, skeletal muscle with atrophic changes, dense fibrous tissue, and some embedded peripheral nerve segments (Fig. 10 *A* and *B*). There was also a small focus of non-necrotizing granulomatous inflammation (acid-fast bacteria and fungal stains were negative).

Discussion

Intraneural ganglion cysts are benign mucinous lesions that form in proximity to peripheral nerves in the upper and lower extremities of the body. They are relatively uncommon. The most common site of these lesions involves the common peroneal nerve and the superior tibiofibular joint of the lower extremity. Desy et al (2) performed a review of the world's literature documenting 648 intraneural cysts in 645 patients. Twenty cases (3.1%) involved the tibial nerve. Other locations of intraneural cysts in the foot and ankle have been described, but these are much rarer (Table).

The etiology of intraneural cysts remains unknown and somewhat controversial, although there is increasing evidence and literature to suggest that these cysts develop from an articular nerve branch (11–14). It is presumed that a capsular defect creates a conduit between a synovial joint and the articular branch of the adjacent nerve with the cyst. The fluid then flows along a path of least resistance along the epineurium proximally, but it can also dissect distally. This is the mechanism described by Spinner et al (15) as occurring in the common peroneal nerve and the proximal tibiofibular joint through methylene blue dye studies performed in cadavers**.** As with the cause of most ganglion cysts, it is presumed that a traumatic event leads to the initial capsular defect. This is easy to understand in the case of the common peroneal nerve and its superficial nature as it crosses the fibular neck. In our patient, no preceding history of overt trauma was given. However, our patient was very active physically, spent extensive hours on his feet in the course of his work, and had a pronated foot type, which is known to exacerbate tarsal tunnel syndrome (16). This combination in the setting of lack of orthotic support may have led to repetitive trauma to the tibial nerve and the subtalar joint. Additionally, it is possible there may have been an episode of unperceived trauma that went untreated resulting in a capsular defect within the subtalar joint. It has been postulated that because of the location of these nerves, mechanical trauma can serve as the initiating factor in bringing about these changes in the nerve and may explain the atrophic changes noted in the surrounding skeletal muscle (13).

The signs and symptoms of an intraneural ganglion cyst are similar to those associated with peripheral neuropathy, namely tingling, radiating pain along with areas of numbness along the distribution of the affected nerve (16). Additionally, clinical examination and palpation may detect an area of fullness. This is especially true in a confined space such as the tarsal tunnel as in this case. Therefore, paresthesias are the primary complaint, along with a positive Tinel's sign. Additionally, there may be numbness along the distribution of the medial or lateral plantar nerve, suggesting selective compression of these nerves. In our patient, these paresthesias interfered with the patient's physical activity, which became increasingly more difficult. His most bothersome symptom occurred at night when trying to sleep, again a hallmark symptom of neuropathy. He was able to sleep only with his legs hanging off the edge of the bed. One explanation may be that by hanging his feet off the bed, it avoided any pressure on the tarsal tunnel and further irritation of the nerve. It was this symptom that ultimately resulted in the patient seeking treatment.

Although the diagnosis of an intraneural ganglion cyst is primarily clinical, diagnostic studies are important in outlining the size and extent of the

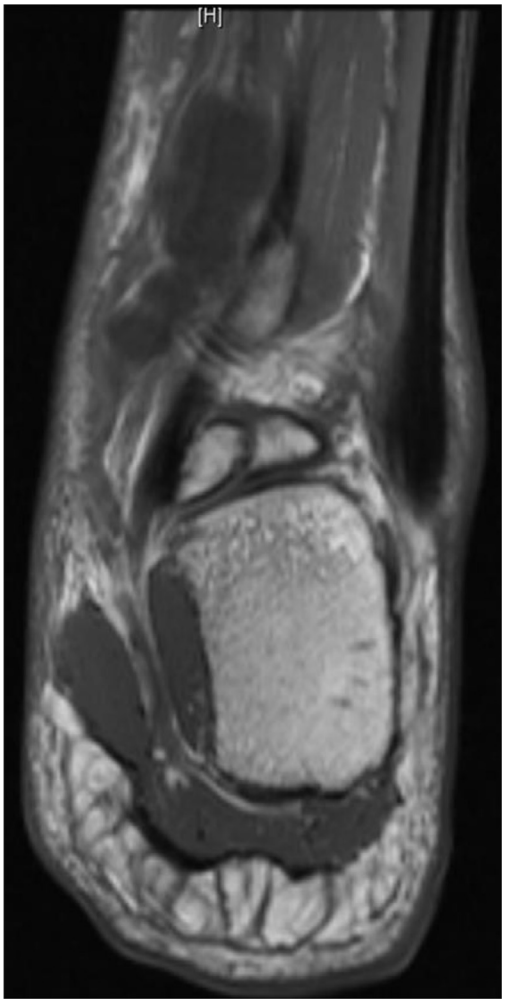

Fig. 3. The tibial nerve can be seen above and below the cyst. It is noted to be thickened and displaced.

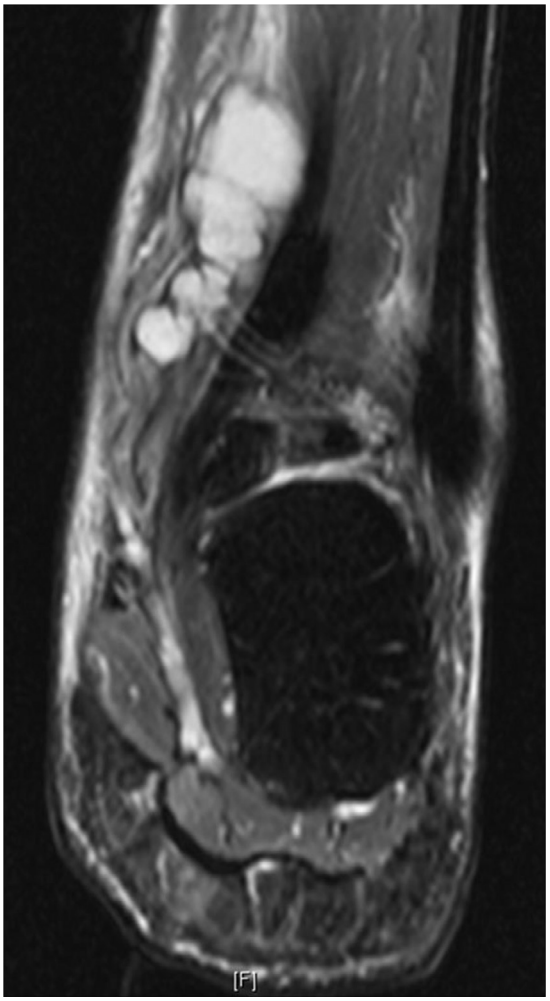

Fig. 4. The multilobulated nature of the cyst can once again be seen along with its full extension from proximal to distal into the tarsal tunnel.

cyst as well as the potential source of the cyst. Plain radiographs are generally of little value in the diagnosis of a cyst. However, they may show degenerative changes in the joint which may suggest the cause and source of the cyst. Ultrasound may also be helpful in identifying a soft tissue mass

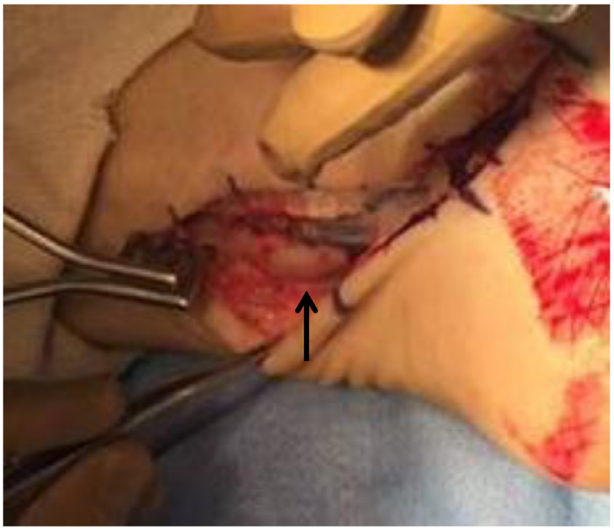

Fig. 5. The intraneural cyst is seen in close proximity to the neurovascular bundle (arrow) within the tarsal tunnel.

Fig. 6. The tibial nerve can be seen protected and gently retracted by vascular loop (large arrow) away from the cyst (small arrow).

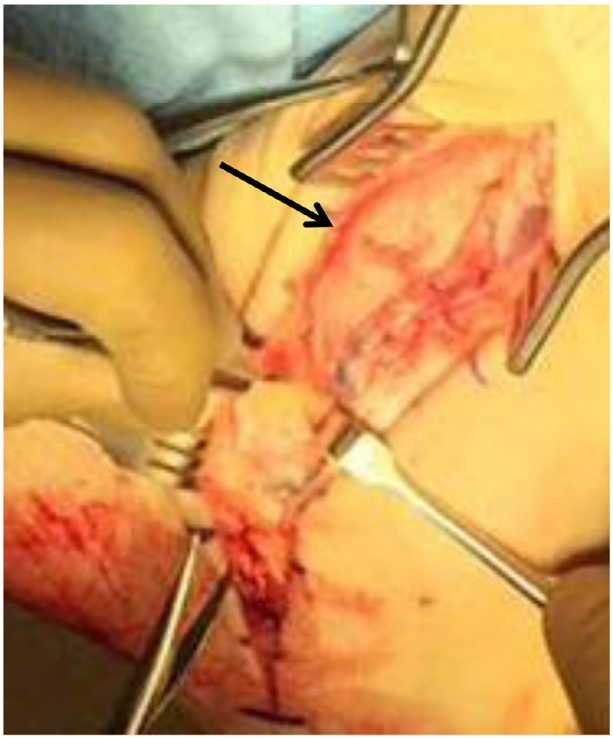

Fig. 7. The cyst is shown here extending from the distal aspect of the tarsal tunnel proximally to the distal third of the leg. Note its continued proximity to the tibial nerve (arrows).

Fig. 8. The tibial nerve is shown completely released and following complete removal of the intraneural cyst.

or space-occupying lesion. However, it provides little information on the source. The study of choice is MR imaging. This should be performed with and without contrast. Additional views beyond the standard cuts through the joint may be necessary to identify the articular branch of the nerve that serves as the conduit for the fluid. This is important for surgical planning and reducing the chances of recurrence. The MR images will identify the extent of the cyst and its proximity to the involved nerve and potentially other vital structures.

Additional preoperative studies such as nerve conduction studies (NCS) or electromyography (EMG) may be helpful in identifying and documenting areas of focal neurological deficits. Focal deficits may also show selective involvement of nerve branches. In the case of the tibial nerve, NCS may show selective involvement of the medial plantar nerve, lateral plantar nerve, or both, which may explain specific

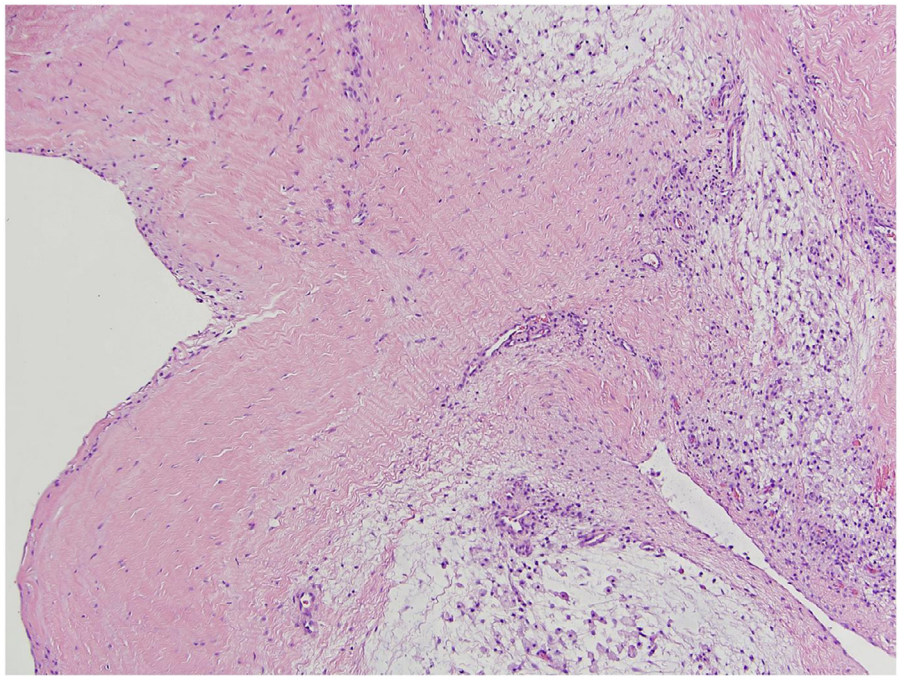

Fig. 9. The ganglion cyst consisted of a fibrous wall (without synovial lining) with intraluminal mucoid material and associated foci of myxoid change (magnification × 10; hematoxylin and eosin stain).

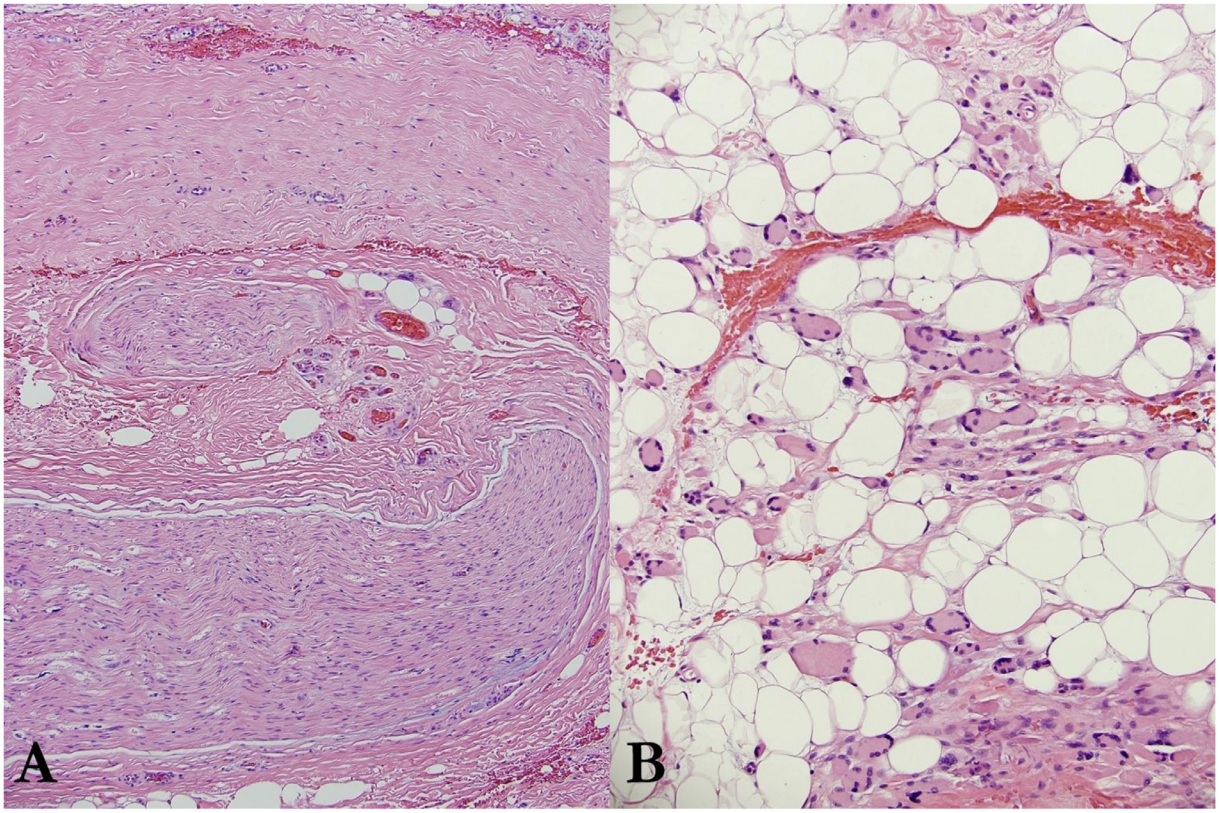

Fig. 10. Soft tissue adjacent to the ganglion cyst showed peripheral nerve segments embedded within dense fibrous tissue (*A*, hematoxylin and eosin stain, original magnification 10×) and foci of fibrofatty tissue with atrophic skeletal muscle fibers (*B*, magnification × 20; hematoxylin and eosin stain).

symptoms. Performing these studies preoperatively will also allow for comparison to studies performed postoperatively, which may demonstrate improvement, worsening, or no change in nerve function in an objective manner. In the systematic review of 645 cysts by Desy et al (2), >50% had documented EMG and NCS preoperatively. Surgical decisions were not based on neurophysiologic studies, and the current literature does not cite these studies as necessary for the diagnosis or treatment of intraneural cysts (14,17).

Our patient did not undergo EMG or NCS pre- or postoperatively. Although this did not affect or impede our surgical approach, the lack of these studies may have prevented us from fully evaluating the success of our surgery. The patient had preoperative complaints of decreased sensation on the plantar aspect of his foot. Postoperatively, he continues to have similar complaints, although his pain is fully resolved and he has no muscle weakness. Because of the lack of neurophysiologic studies, we are not able to objectively determine if nerve function has improved or worsened or is unchanged. Additionally, we are unable to determine the prognosis for return of sensation. He has been evaluated by a neurologist who does not recommend studies at this time, as they will not change course of treatment. He was told it may take as long as 2 years for sensation to possibly return.

Treatment consists of surgical exploration with or without removal of the cyst. Because the currently accepted theory is that the origin of the cyst is through an articular branch of the nerve supplying a joint,

Table
Occurrence of intraneural ganglion cyst in the lower extremity by nerve: review of the world literature*

Cyst	Occurrence	Incidence in Nerve of Lower Extremity (%)	Incidence in All Nerves (%)
Tibial	20	4	3
Sural	11	2	1.7
Superficial peroneal (ankle)	3	0.7	0.5
Tibial and medial plantar	2	0.4	0.3
Medial plantar	12	2.4	1.9
Lateral plantar	1	0.2	0.2
Plantar nerve	2	0.4	0.3
Deep peroneal (foot)	2	0.4	0.3
Superficial peroneal (foot)	1	0.2	0.2
Intermediate dorsal cutaneous	1	0.2	0.2
Lateral dorsal cutaneous	2	0.4	0.3

Total intraneural cysts, N = 648; total intraneural cysts in the lower extremity, n = 499 (77%); total intraneural cysts at the level of the ankle and distal, n = 57 (11% of lower extremity, 8.7% of all nerves).

* Adapted from Desy et al (2).

it is recommended that the articular branch of the nerve be identified and ligated, thus depriving the cyst of the source of the fluid (9). When the articular branch is identified and ligated, the cyst can be evacuated of its fluid and left in situ with relief of systems. If the surgeon elects to remove the cyst, the articular branch of the nerve should still be ligated to avoid recurrence (8,18). There has also been a recent report of endoscopic release of the cyst (19). In the present case, the cyst was dissected free from the tibial nerve starting at the tarsal tunnel and continued proximally until the entire cyst was mobilized. Once fully mobilized, the cyst was once again located distally, where it was found to communicate with the subtalar joint. Although an articular branch was not specifically visualized, the connection between the joint and the cyst was identified, and a periosteal flap was elevated separating the cyst from the joint at this level. The periosteal flap and joint connection were then ligated to prevent recurrence of the cyst.

Potential complications of surgery are those commonly associated with any type of surgery involving peripheral nerves, that is, residual numbness, paresthesia, and persistent pain including complex regional pain syndrome (20). Additionally, because of the proximity of other structures in the tarsal tunnel, injury to the tibialis posterior tendon or artery has also been described. Careful dissection under tourniquet control and the use of loupe magnification to improve visualization can be helpful in reducing these risks (10).

In conclusion, this case report discusses the diagnosis, treatment, and review of the literature of an intraneural ganglion cyst of the tibial nerve resulting in tarsal tunnel syndrome. Typical symptoms include those associated with neuropathy such as numbness and burning, shooting pain. In advanced cases, muscle weakness and atrophy may occur. MR imaging with and without contrast is critical to the diagnosis, as is surgical planning. A connection between the cyst and the articular branch of the nerve is diagnostic of the condition. Neurophysiologic studies, while helpful, are not required to diagnose and treat intraneural ganglion cysts. Treatment is surgical in nature, although total excision of the cyst is not always necessary. It is important, however, to identify and ligate the articular branch that feeds the cyst to reduce the likelihood of recurrence. Once ligation is done, the cyst can be evacuated of its contents and left in situ. This may reduce the risk of complications commonly associated with tarsal tunnel surgery. Alternatively, in the case of large, extensive cysts, the surgeon may choose to remove the cyst in toto.

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