

Superficial Peroneal Intraneural Ganglion Cyst Originating from the Inferior Tibiofibular Joint: The Latest Chapter in the Book

The article by Stamatis et al highlights a novel site for an intraneural ganglion cyst—the superficial peroneal nerve (SPN) in the ankle region (1). Despite recent publications describing a consistent articular joint connection (2,3), these authors did not find one preoperatively or intraoperatively. One must wonder if their assessment is complete, ie, if there is more to the story.

Intraneural ganglion cysts, considered curiosities for nearly 200 years, are now being understood by the articular (synovial) theory (4). Over the past decade “chapters” of evidence have been put forth to elucidate the formation and propagation of intraneural cysts: (1) proposing a theory based on the most common site, the peroneal nerve at the superior tibiofibular joint (STFJ) (2); (2) defining its principles (3); and (3) extrapolating it to rare sites—small- (5) and large- (6) caliber nerves, arising from different joints in the appendicular skeleton (4) and axial spine (7). Any case in which an articular connection is not identified, by definition, challenges the theory, detracts from proof, and potentially sets back progress and momentum.

We offer a different perspective on Stamatis et al’s (1) par-articular cyst based on their beautiful published figures without the benefit of the opportunity to review the entire magnetic resonance (MR) study and to correlate it with the published intraoperative photograph. Despite these limitations, we demonstrate that this case represents a joint-related intraneural ganglion, one that (1) follows the articular (synovial) theory, (2) obeys the principles of this theory, and (3) has features that are generalizable.

Following the Unifying Articular (Synovial) Theory

Although the authors believe that finding a small connection to the inferior tibiofibular joint (ITFJ) is a “reasonable” explanation, we, in fact, believe it is the only explanation. Conveniently, 2 of the 3 MR published images (Figs. 1 and 3 in their article) demonstrate the unrecognized joint connection (Fig. 1). The third image (Fig. 2 in their report), a low-resolution SPIR (spectral presaturation with inversion recovery) image, is proximal to the joint connection that was seen on the lower images; the arrowheads on the original published image point to a small amount of fluid within the most superior aspect of the ITFJ that is contiguous with the fluid seen on the lower images. The cyst arising from the ITFJ on the images provided can be traced to the more lateral nerve branch, labeled by the authors as the intermediate dorsal cutaneous (IDC) on their black-and-white intraoperative photograph. The MR images presented appear to be slightly distal to the bifurcation of the SPN with cyst seen in both terminal branches of the nerve—IDC and medial dorsal cutaneous (MDC) nerves per the authors—based on the intraoperative photograph. Although we are confident that the cyst arises from the ITFJ, the exact point of origin of the cyst (specifically, its relationship to the anterior tibiofibular ligament and tibiofibular syndesmosis) cannot be determined without access to other contiguous axial images and/or other planes. Nor can we comment on the presence or absence of subtle ligamentous injury or bony/joint abnormality.

The formation of a superficial peroneal intraneural cyst from the ITFJ is not intuitively obvious. In fact, at first glance, such an intraneural cyst may appear to be a clinical outlier to the articular

(synovial) theory. On reinterpretation of this case, we believe that it affords a window into understanding the anatomy of the ITFJ, its articular branch anatomy, and the potential pathologies arising from it. First, unlike all other examples of intraneural ganglia thus far reported that are derived from a synovial joint (4), the ITFJ has historically been considered a fibrous joint of the syndesmosis type. Its inferior part has a small superior projection of synovial capsule extending from the ankle joint (8,9). More recently, the ITFJ has been shown to be a true synovial joint in three fourths of cases (10). Second, the SPN is not widely known to have an articular branch to the ITFJ. This articular branch was only relatively recently described (11,12), and was not described in classic anatomic papers (13–15) and textbooks (16–19). The better known deep peroneal and sural nerves also innervate the ITFJ (12,14); some have described articular branches by the saphenous nerve (8) or tibial nerve’s interosseous crural (13) or the deep peroneal’s interosseous branch (12). More detailed information regarding the innervation of this joint is needed. These parent nerves, eg, the superficial peroneal, deep peroneal, sural, tibial, and saphenous nerves, all innervate the ankle joint. Third, although several cases of tibial intraneural ganglia have been reported originating from the ankle joint (20–23), we are unaware of any cyst previously described arising from the ITFJ. Joint fluid and effusions (24,25), which are known to occur in the ITFJ, could be attributed to known joint communications with the ankle (26); however, reports of synovial chondromatosis (27) and osteochondromatosis (28), pigmented villonodular synovitis (29), and arthritic bony erosions (30) specifically affecting the ITFJ, confirm the presence of synovium in the ITFJ and its potential to produce pathology in this location. Therefore, it is logical that the synovial lining in the ITFJ can also be the origin for intraneural (or extraneural) ganglia in this location, especially given its propensity for trauma and degenerative changes.

This case further demonstrates that knowing about the articular theory is not enough and that suspecting an articular branch connection is only the first step. Unless the joint pathology is addressed directly and definitively, the articular branch should be actively looked for, identified, and treated. The relatively small size of the articular branch has been elaborated on in several previous publications (2,4,31). It is easy to understand how this tiny connection might be missed on imaging (Fig. 1) and visually. This articular branch connection has eluded the eyes of expert radiologists and surgeons. Identification of the sometimes clinically occult articular branch has been catalyzed by improvements in imaging techniques and sequences, and experience in the interpretation of images (2,4). MRI has reliably identified joint connections, and computed tomography/MR imaging (CT/MRI) arthrography, communications of the joint of origin with the cyst (31,32). Although the treatment of choice is still being substantiated, it is clear that understanding the pathology is necessary to target the intervention. Resection of the cyst is not necessary (as long as the articular branch is disconnected) (4). This has been demonstrated in cysts of all dimensions, including “extreme” examples, where complete resection would clearly not be feasible (33). Resection of the nerve was performed in this case; this technique, although possible in this particular distal cutaneous nerve, should not be performed widely and should not be generalized, especially to cysts involving major mixed nerves.

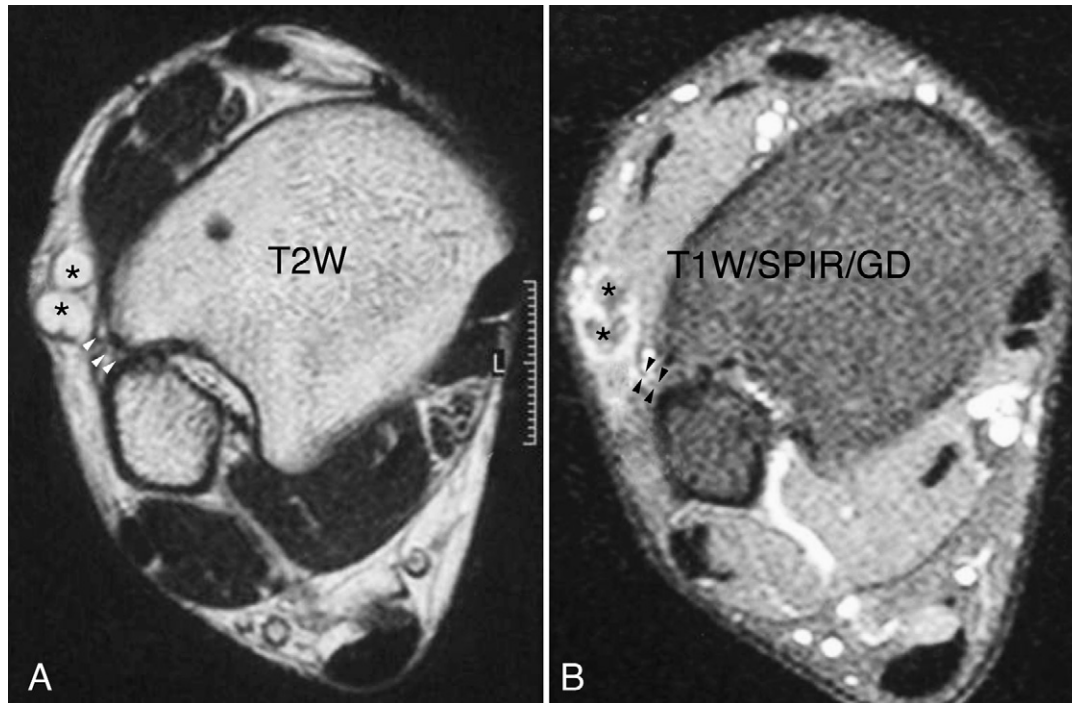


Fig. 1. Reinterpreted MRIs. (A) Axial T2-weighted fast spin-echo (FSE) image at the level of the mid to distal aspect of the inferior tibiofibular joint (ITFJ) shows the intraneural cyst within the terminal branches of the superficial peroneal nerve (asterisks). The cyst origin from the anterior aspect of the ITFJ is shown (arrowheads), as well as the direct connection to the more lateral intraneural cyst. (B) Axial T1-weighted spectral presaturation with inversion recovery (SPIR) after intravenous gadolinium shows the intraneural cyst (asterisks) and anterior joint connection (arrowheads). Note enhancement of cyst wall. (From Stamatis ED, Manidakis NE, Patouras PP. Intra-neural ganglion of the superficial peroneal nerve: a case report. *J Foot Ankle Surg* 49: 400.e1–400.e4, 2010, Figs. 1 and 3.)

Surgical outcomes are improving and recurrences can be eliminated by addressing the articular origin of these intraneural cysts.

Obeying Principles of the Articular Theory

Establishing a joint connection in this case allows the operative photograph to be reinterpreted in a different light, one that can explain the propagation pattern. The finding of interconnected intraneural cysts (“multicystic expansion”) is not coincidental but can easily be explained by a phasic mechanism based on fluid following the path of least resistance—just as it has been in other locations including the peroneal or tibial nerves in the distal thigh (3) or the deep peroneal nerve in the foot (5). In this case, evidence of ascent (phase I), cross-over at the SPN bifurcation (phase II), and descent (phase III) are seen. Because of possible anatomic variations in articular branch anatomy, we offer 2 potential paths of propagation for superficial peroneal intraneural ganglia arising from the ITFJ (Fig. 2): (1) ascent up the IDC, with cross-over at the bifurcation of the SPN and descent down the MDC and (2) ascent up the MDC and descent down the IDC. In their case, the first pathway seems preferred because of the lateral position of the cystic joint connection seen on MRI and the lateral trail of blebs seen in the operative figure. Given the limitations of the data available to us, the second option should be considered given the relatively large dimension of the MDC compared with the IDC. Continued ascent was present in the SPN (“balloon” sign) (34) owing to the effects of increased intra-articular pressure.

Being Generalizable

This case of a superficial peroneal intraneural ganglion has stereotypic features analogous to other cases of intraneural ganglia, ie, those occurring in common and more unusual locations, from the hip (6,35) to the toes (5) and in between. In particular, this superficial peroneal intraneural cyst derived from the ITFJ is analogous to the previously described and better-known peroneal or tibial examples derived from the STFJ. Both the relatively common peroneal intraneural cyst derived from the STFJ and the novel superficial peroneal intraneural cyst from the ITFJ can be considered bookends of each other because of their many similar features. Knowledge of the articular theory and articular branch anatomy would predict that other intraneural cysts (eg, ones within the deep peroneal or sural nerve) could potentially be derived from the ITFJ, just as a tibial intraneural cyst can be derived from the STFJ (36) (Fig. 3). Moreover, application of the articular theory would predict that an isolated superficial peroneal intraneural cyst derived from the STFJ cannot exist but that an intraneural cyst within the SPN or its terminal branch derived from a joint in the foot (37) and ankle region can exist.

In conclusion, this article of a superficial peroneal intraneural cyst near the ankle has implications that transcend beyond the typical case report relating to the pathogenesis (formation and propagation) but also to both diagnosis and treatment of a curious entity. Our conclusions differ from those of the original authors (1). This case, which challenged the underlying principles of the articular theory, after reinterpretation and reconsideration with a different perspective, strengthens and further generalizes it. In

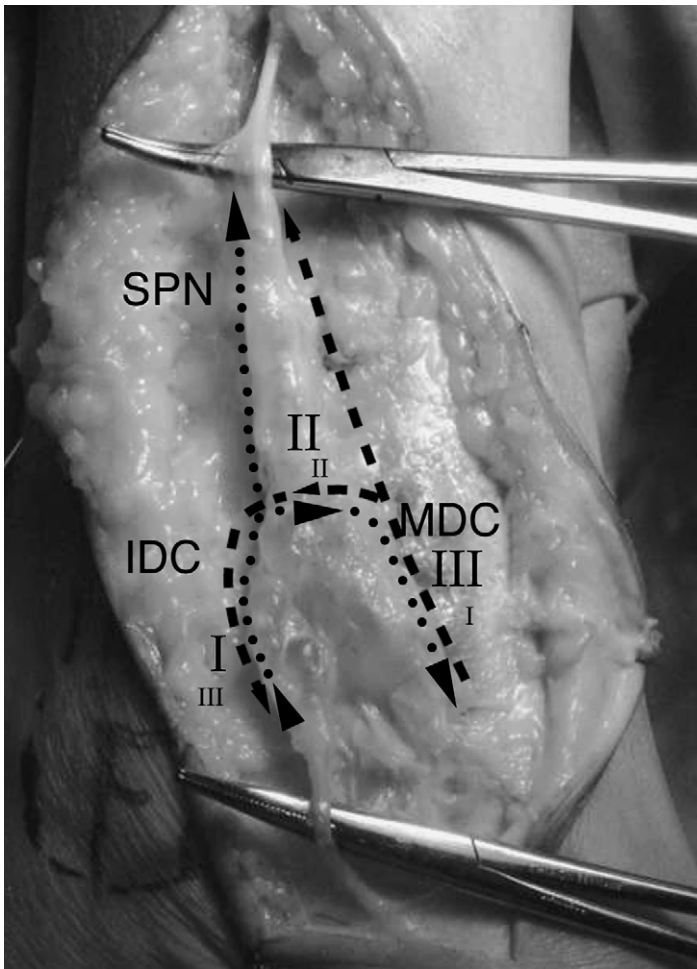


Fig. 2. Reinterpretation of the operative photograph. Two pathways are shown to illustrate possible phasic mechanisms of formation and propagation: (1) Dotted arrows show ascent up the intermediate dorsal cutaneous (IDC) nerve (phase I) from an articular branch; cross-over (phase II) at the bifurcation of the superficial peroneal nerve (SPN) and descent (phase III) down the medial dorsal cutaneous nerve (MDC). (2) Dashed arrows show: ascent (I) up the MDC; cross-over at the SPN bifurcation (II), and descent (III) down the IDC. The larger Roman numerals show the preferred phasic pathway. (From Stamatis ED, Manidakis NE, Patouras PP. Intraneural ganglion of the superficial peroneal nerve: a case report. *J Foot Ankle Surg* 49: 400.e1–400.e4, 2010 [Epub ahead of date], Fig. 4.)

the end, this important case represents the latest chapter in the evolving story of intraneural ganglia.

Acknowledgments

We appreciate the assistance with the editorial illustration, ie, the inspiration from Alexandra Wolanskyj, MD, and the rendering by David Factor (Rochester, MN).

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Dr. Spinner has worked as a consultant/advisor to Mays Medical Ventures.

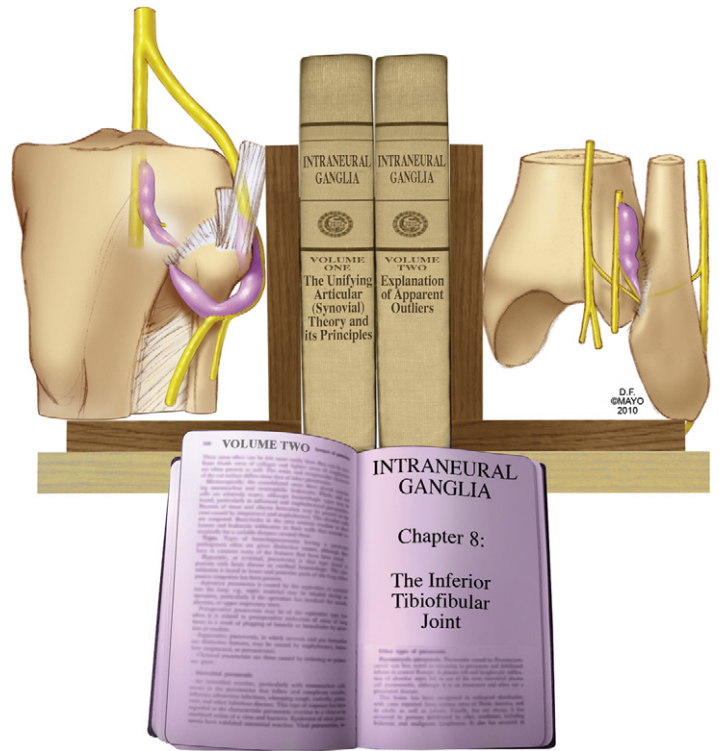


Fig. 3. The latest chapter, “The Inferior Tibiofibular Joint,” in the book *Intraneural Ganglia*. Just as bookends keep books upright, the articular (synovial) theory serves as a buttress to explain the pathogenesis of intraneural cysts. The superior tibiofibular joint (STFJ) (left bookend): The STFJ has served as the prototype for understanding the origin of the most common form of intraneural ganglia—those affecting the peroneal nerve. Subsequently, tibial intraneural cysts were shown to be the posterior counterpart, being derived from the posterior part of the STFJ, whereas the peroneal are derived from the anterior part. The inferior tibiofibular joint (ITFJ) (right bookend): The ITFJ as shown in this case can be the origin of a superficial peroneal intraneural cyst. The anatomy of the superficial peroneal nerve and its branches is purposefully simplified. The drawing is adapted from the anatomic descriptions by Lippert (14) of deep peroneal and sural nerve innervation and Mentzel et al (12) of superficial peroneal nerve innervation to the ITFJ. Based on the articular anatomy, other intraneural cysts within the deep peroneal or sural nerves, for example, could potentially be derived from the ITFJ as well (From Mayo Foundation, 2010; with permission).

References

1. Stamatis ED, Manidakis NE, Patouras PP. Intraneural ganglion of the superficial peroneal nerve: a case report. *J Foot Ankle Surg* 49:400.e1–400.e4, 2010.
2. Spinner RJ, Atkinson JLD, Tiel RL. Peroneal intraneural ganglia. The importance of the articular branch: a unifying theory. *J Neurosurg* 99:330–343, 2003.
3. Spinner RJ, Wang H, Amrami KK, Wolanskyj AP, Benarroch EE, Skinner JA, Rock MG, Desy NM, Scheithauer BW. The dynamic phases of peroneal and tibial intraneural ganglia: a new dimension added to the unifying articular theory. *J Neurosurg* 107:296–307, 2007.
4. Spinner RJ, Scheithauer BW, Amrami KK. The unifying articular (synovial) origin of intraneural ganglia: evolution-revelation-revolution. *Neurosurgery* 65:A115–A124, 2009.
5. Blitz NM, Amrami KK, Spinner RJ. Magnetic resonance imaging of a deep peroneal intraneural ganglion cyst originating from the second metatarsophalangeal joint. A pattern of propagation supporting the unified articular (synovial) theory for the formation of intraneural ganglia. *J Foot Ankle Surg* 48:80–84, 2009.
6. Spinner RJ, Hébert-Blouin M-N, Trousdale RT, Midha R, Russell SM, Yamauchi T, Sasaki S, Amrami KK. Intraneural ganglia in the hip and pelvic region. *J Neurosurg* 111:317–325, 2009.
7. Spinner RJ, Hébert-Blouin NM, Maus TP, Atkinson JLD, Desy NM, Amrami KK. Evidence that atypical juxtafacet cysts are joint derived. *J Neurosurg Spine* 12:96–102, 2010.

8. Moore KL, Dalley AF, Agur AMR. *Clinically Oriented Anatomy*, ed 6, Lippincott Williams and Wilkins, Baltimore, 2010.
9. Hollinshead WH. *Anatomy for Surgeons. The Back and Limbs*, vol. 3, ed 2, Hober Medical Division, Harper & Row, Publishers, New York, 1969.
10. Bartoníček J. Anatomy of the tibiofibular syndesmosis and its clinical significance. *Surg Radiol Anat* 25:379–386, 2003.
11. Champetier J. Innervation de l'articulation tibio-tarsienne (Articulatio talocruralis). *Acta Anat* 77:398–421, 1970.
12. Mentzel M, Fleischmann W, Bauer G, Kinzl L. Ankle joint denervation. Part I: Anatomy—the sensory innervation of the ankle joint. *Foot Ankle Surg* 5:15–20, 1999.
13. Casagrande PA, Austin BP, Indeck W. Denervation of the ankle joint. *J Bone Joint Surg Am* 33:723–730, 1951.
14. Lippert Z. Zur innervation der menschlichen fußgelenke. *Z Anat Entwicklungsgesch* 123:295–330, 1962.
15. Gardner E, Gray DJ. The innervation of the joints of the foot. *Anat Rec* 161:141–148, 1968.
16. Woodburne RT, Burkel WE. *Essentials of Human Anatomy*, ed 9, Oxford University, New York, 1994.
17. Sunderland S. *Nerves and Nerve Injuries*, ed 2, Churchill Livingstone, Edinburgh, 1978.
18. Gray's Anatomy: The Anatomical Basis of Clinical Practise, ed 40, edited by S Standring, Churchill Livingstone Elsevier, Edinburgh, 2008.
19. Sarrafian SK. *Anatomy of the Foot and Ankle. Descriptive Topographic Functional*, ed 2, JB Lippincott Co, Philadelphia, 1993.
20. Mühlbauer W, Stock W, Heiß J. Intraneural wachsendes ganglion am n. tibialis posterior. *Chir Praxis* 20:609–611, 1975/76.
21. Høgh J. Benign cystic lesions of peripheral nerves. *Int Orthop* 12:269–271, 1988.
22. Spinner RJ, Dellon AL, Rosson GD, Anderson SR, Amrami KK. Tibial intraneural ganglia in the tarsal tunnel: is there a joint connection? *J Foot Ankle Surg* 46:27–31, 2007.
23. Spinner RJ, Amrami KK, Hamlat A. Letter to the editor: collaborative follow-up. *Acta Neurochir (Wien)* 150:93–96, 2008.
24. Brown KW, Morrison WB, Schweitzer ME, Parelada JA, Nothnagel H. MRI findings associated with distal tibiofibular syndesmosis injury. *AJR Am J Roentgenol* 182:131–136, 2004.
25. Evans JM, Schucany WG. Radiological evaluation of a high ankle sprain. *Proc (Bayl Univ Med Cent)* 19:402–405, 2006.
26. Vora AM, Haddad SL, Kadakia A, Lazarus ML, Merk BR. Extracapsular placement of distal tibial transfixation wires. *J Bone Joint Surg Am* 86:988–993, 2004.
27. Tibrewal SB, Iossifidis A. Extra-articular synovial chondromatosis of the ankle. *J Bone Joint Surg* 77B:659–660, 1995.
28. Yu GV, Zema RL, Johnson RWS. Synovial osteochondromatosis. A case report and review of the literature. *J Am Podiatr Med Assoc* 92:247–254, 2002.
29. Mori H, Nabeshima Y, Mitani M, Ozaki A, Fujii H, Kuroda R. Diffuse pigmented villonodular synovitis of the ankle with severe bony destruction: treatment of a case by surgical excision with limited arthrodesis. *Am J Orthop* 38:E187–E189, 2009.
30. Karasick D, Schweitzer ME, O'Hara BJ. Distal fibular notch: a frequent manifestation of the rheumatoid ankle. *Skeletal Radiol* 26:529–532, 1997.
31. Spinner RJ, Amrami KK, Rock MG. The use of MR arthrography to detect an occult joint communication in a recurrent peroneal intraneural ganglion. *Skeletal Radiol* 35:172–179, 2006.
32. Omoumi P, de Gheldere A, Leemrijse T, Galant C, Van den Bergh P, Malghem J, Simoni P, C Vande Berg B, Lecouvet FE. Value of computed tomography with delayed acquisitions in the work-up of ganglion cysts of the tarsal tunnel: report of three cases. *Skeletal Radiol* 39:381–386, 2010.
33. Spinner RJ, Hébert-Blouin MN, Rock MG, Amrami KK. Extreme intraneural ganglion cysts. *J Neurosurg* May 21 2010 [Epub ahead of print].
34. Spinner RJ, Amrami KK. Letter to editor. The Balloon Sign. *Acta Neurochir (Wien)* 148:1224–1226, 2006.
35. Spinner RJ, Hébert-Blouin MN, Tanaka S, Amrami KK, Swartz KR, Fee DB, Sugita M. Hip- and pelvic-related intraneural ganglia. *J Neurosurg* 112. 1353–1356.
36. Spinner RJ, Mokhtarzadeh A, Schiefer TK, Krishnan KG, Kliot M, Amrami KK. The clinico-anatomic explanation for tibial intraneural ganglion cysts arising from the superior tibiofibular joint. *Skeletal Radiol* 36:281–292, 2007.
37. Lemont HL, Sabo MA. Origin of foot ganglion. *J Am Pod Med Assn* 90:256–257, 2000.