Development of a tibial intraneural ganglion cyst after surgery on a peroneal intraneural ganglion in a pediatric patient: further support for the articular origin of intraneural ganglion cysts. Illustrative case

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BACKGROUND The articular origin of intraneural cysts has been previously described and well supported. Intraneural ganglion cysts most commonly occur in adults and in the common peroneal nerve arising from the anterior aspect of the superior tibiofibular joint (STFJ).

OBSERVATIONS The authors report a pediatric patient who developed a tibial intraneural cyst arising from the posterior aspect of the STFJ within months after surgical treatment of a peroneal intraneural cyst from the anterior aspect of the same joint. Surgery consisted of disconnecting the articular branch of the peroneal nerve and decompression of the cyst. Three-month postoperative magnetic resonance imaging showed resolution of the peroneal intraneural cyst and anterior compartment denervation but demonstrated a clinically occult small tibial intraneural ganglion cyst arising from the STFJ with popliteus muscle atrophy.

LESSONS This case underscores the importance of addressing the underlying etiology (articular synovitis) or the communication pathways (articular branches) whenever feasible to mitigate intraneural cyst recurrence. Furthermore, this report reinforces the validity of the articular theory.

https://thejns.org/doi/abs/10.3171/CASE24277

KEYWORDS intraneural ganglion cyst; peroneal and tibial nerve; unified theory; articular branch; case report

Intraneural ganglia,1-4 nonneoplastic mucinous cysts located within the epineurium of nerves, are most frequently observed within the common peroneal nerve at the fibular neck level in adults. Initially described 2 centuries ago, these cysts have been noted for their distinctive and intriguing characteristics.5 The first reported case was briefly described by Beauchêne in 1810.6 The advent of high-resolution magnetic resonance imaging (MRI) has significantly contributed to revealing both typical and atypical clinicoradiological manifestations of intraneural ganglia, thereby enhancing our understanding of this entity. The formation and treatment of this entity have been well explained and substantiated by a unifying articular theory.7-10 This theory supports a synovial joint origin for cyst development: joint fluid (cyst) egresses through a capsular rent and courses along an articular branch into a parent nerve. Typically, patients with peroneal intraneural cysts arising from the superior tibiofibular joint (STFJ) present with symptoms and signs of predominant deep peroneal nerve (DPN) involvement (such as foot drop) due to the articular branch originating from the DPN. With increased joint pressure, the cyst may extend proximally within the common peroneal nerve and even to the sciatic nerve following a path of least resistance. The mainstay of surgical treatment for intraneural cysts has been to disconnect the articular branch and to decompress the cyst. In general, this relatively straightforward approach has frequently led to favorable outcomes (improved pain and function and resolution of the cyst). Though not uniformly adopted, our group has also recommended resection of the synovial surface of the STFJ. This approach has further decreased cyst recurrence without causing joint instability.11 Not identifying and treating the pathologic articular branch connection to the joint has led to high rates of (subclinical or clinically evident) intraneural recurrence. Cyst resection, especially in extensive cysts, is unnecessary and risks neural compromise from intraneural dissection.

Herein, we present a case to illustrate 2 unusual features: 1) 2 intraneural cysts developing (first in the common peroneal nerve and then the tibial nerve) from the same joint metachronously, through 2 different articular branches to 2 different parts of the joint (anterior and posterior aspects of the STFJ), respectively; and 2) the consequence of these cysts in a pediatric (i.e., skeletally immature) patient in whom resection of the STFJ was not performed. We believe that this case
provides additional evidence supporting the articular origin of these intraneural cysts and underscores the importance of treating the joint of origin whenever possible to optimize outcomes.

Illustrative Case
A 6-year-old boy initially presented with spontaneous-onset footdrop persisting for 7 months. Clinical examination revealed British Medical Research Council grade M0 for tibialis anterior, extensor digitorum, and extensor hallucis longus muscles, while peronei function was grade M4. The sensation was decreased on the dorsum of the foot and in the first web space. Tinel’s sign was found at the fibular neck over the lesion. Preoperative MRI demonstrated a peroneal intraneural ganglion cyst (50 × 9.4 × 9.7 mm) arising from the anterior aspect of the STFJ, with muscle denervation of the anterior compartment of the leg (Fig. 1A–C). Surgery consisted of identifying and resecting the articular branch and decompressing the cyst (Fig. 2). Resection of the STFJ was not performed due to the patient’s age and the risk for altered bone growth. Within 3 months, the patient noted rapid improvement of the footdrop. Postoperative MRI showed resolution of the peroneal intraneural cyst and anterior compartment denervation but demonstrated a clinically occult tibial intraneural ganglion cyst (28.4 × 10 × 4.7 mm) arising from the posterior aspect of the STFJ, with popliteus muscle atrophy (Fig. 1D–F). Five months postoperatively, he had grade 4 anterior compartment muscle function and normal peronei.

Patient Informed Consent
The necessary patient informed consent was obtained in this study.

Discussion
Observations
In our case, 2 different nerves with 2 distinct intraneural cysts arising from the same joint were affected. A review of over 1000 cases in the literature reveals only 3 cases of interest. Two cases had 2 intraneural ganglia developing presumably from the same joint: one with concurrent cysts affecting the radial collateral nerve and dorsal collateral nerves of the thumb (at the metacarpophalangeal joint level, though a joint connection was not identified), and the other with intraneural ganglion cysts of the lateral dorsal cutaneous nerve and DPN in the foot, arising from the fourth metatarsal–cuboid and intermetatarsal
portions of a common joint complex. A third case presented with metachronous tibial (first) and peroneal (second) intraneural ganglia arising from the STFJ following disconnection of the tibial articular branch and cyst decompression within the tibial nerve, without resection of the STFJ; the peroneal nerve intraneural cyst remained asymptomatic. Our case, presented here, initially involved a peroneal intraneural cyst. Following resection of the peroneal articular branch to the STFJ, a second intraneural cyst was observed in the tibial nerve. This mechanism of 2 intraneural ganglion cysts arising from the same joint differs from the cross-over phenomenon, where interconnected intraneural ganglion cysts may originate from the same joint through the same articular branch. The neural pathways for the cyst propagation along the peroneal and tibial nerves follow the principles of the articular theory with phasic extension. For the peroneal nerve, ascent (proximal portion of the common peroneal nerve), cross-over (at the sciatic bifurcation), and descent (tibial nerve or other terminal nerve branches) of the synovial fluid connected to the STFJ (or knee joint) via articular branches has been described in detail. Analogously, a cyst can extend along the articular branch of the tibial nerve. Cross-over allows cysts to extend from one nerve to another nerve (primary neural pathway to a secondary one).

The STFJ is innervated by 2 nerves (peroneal and tibial) per Hilton’s law. The articular branch from the peroneal nerve innervates the anterior portion, while the articular branch of the tibial nerve supplies the posterior portion. In the case illustrated, the patient had the classic presentation of predominant DPN involvement (albeit in a 6-year-old individual) and was found to have a typical peroneal intraneural ganglion cyst arising from the STFJ. Surgical treatment

FIG. 2. Intraoperative photographs of the peroneal intraneural cyst surgery. A: A short oblique incision was made, exposing the cystic common peroneal nerve (asterisk, encircled in a blue vessel loop) proximal to the fibular tunnel. B: The dissection was taken in a U-shaped fashion through the fibular tunnel to the STFJ. The cystic-appearing articular branch to the anterior aspect of the STFJ is exposed (red vessel loop). The asterisk indicates the peroneal nerve. C: The cyst within the common peroneal nerve (asterisk) is decompressed through a small pinhole incision (the inset shows mucinous material being evacuated). D: The common peroneal nerve is decompressed. E and F: The articular branch at the level of the anterior STFJ capsule is resected. Pathology confirmed an intraneural ganglion.
consisted of an articular branch disconnection and cyst decompression. The STFJ was not addressed in this pediatric patient because of concerns of negatively affecting skeletal growth. Postoperatively, while the DPN paralysis improved and the peroneal cyst dissipated quickly, the patient developed an occult tibial intraneural ganglion cyst arising from the posterior aspect of the STFJ with popliteus muscle atrophy (a common finding in a tibial intraneural ganglion arising from this joint). This tibial intraneural ganglion cyst developed from the intact articular branch connection between the tibial nerve and the posterior STFJ (and not through the cross-over mechanism previously described). We believe that resection of the peroneal articular branch caused a change in pressure flow in the underlying abnormal STFJ (more joint fluid than usual): this led to the leakage of synovial fluid into the tibial nerve through (a second) the posterior-directed articular branch. These observations further support the articular theory.

Pediatric cases of intraneural cysts are rare. A recent systematic review identified 69 pediatric cases (age < 16 years old) in the literature. In this age group, the common peroneal nerve was the most common nerve affected (90% of cases) and had a high recurrence rate of 17%. This particular patient had evidence of hypermobility in other joints, leading us to wonder if capsular laxity (rather than direct trauma or degenerative changes) predisposed him to the joint problem.

Lessons
The optimal treatment for managing intraneural ganglion cysts is evolving based on the articular theory. Suboptimal results have improved over the past few decades by understanding that the primary pathology is the affected joint via its articular branch connection and that the cyst is the consequence. Surgeons have been resecting the articular branch(es) to eradicate the pathway whenever feasible. However, if the pathologic joint is not addressed (subjected to increased pressure), it can still lead to recurrence of the cyst or progression of symptoms, necessitating comprehensive management strategies that include addressing the underlying joint pathology in addition to the cyst. Three main types of intraneural cyst recurrence patterns based on the initial surgical management of the intraneural ganglion cyst have been described: type I, parent nerve recurrence when treatment focuses on cyst decompression (without addressing the joint or the articular branch connection); type II, articular branch recurrence with incomplete/incorrect articular branch disconnection (without addressing the joint); and type III, joint recurrence when the joint is not correctly addressed. When addressing the STFJ, resecting the anterior (arising from the peroneal nerve) and posterior (arising from the tibial nerve) branches is not readily feasible through the same surgical approach. Therefore, we believe that when performing the STFJ resection, the primary joint pathology is addressed, avoiding the specific treatment of other uninvolved (noncytic) articular branches. As previously explained, STFJ resection was not performed in this case due to the age group of this patient (generally considered in patients older than 16–18 years of age). Based on our extensive experience, we perform disconnection of the primary cystic articular branch and resection of the STFJ in the vast majority of adults with peroneal intraneural ganglion cysts arising from the STFJ; in patients with (small) “nearly invisible cysts,” we tend to disconnect the peroneal articular branch to the STFJ without STFJ resection. We also decompress the cyst, whenever feasible, through a small stab incision.

Follow-up imaging is a useful part of our practice for documenting interval changes and resolution of the cysts and to identify an occult recurrence or a secondary cyst, as in our case. We normally perform MRI at 3 months and 12 months after surgery. In the reported case, we plan for close observation (clinical examination and repeat MRI) 1 year postoperatively. If symptoms such as paresthesia, weakness, or pain occur in the tibial nerve distribution, then ultrasound-guided aspiration of the cyst/STFJ would be performed to minimize symptoms, decrease the size of the cyst, and temporize until the child becomes skeletally mature. At that time, if revision surgery were needed, resection of the STFJ could be considered either by itself or in addition to disconnection of the tibial articular branch to the STFJ.

In conclusion, the particular characteristics of this unusual case (metachronous intraneural cysts in a pediatric patient) allow us to support a mechanism by which 2 different intraneural cysts can emerge from 2 different articular branches coming from the same joint. Finally, we believe this report strengthens the articular theory and expands our experience with surgical treatment.

References
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**Disclosures**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**Author Contributions**

Conception and design: Spinner, Amrami. Acquisition of data: all authors. Analysis and interpretation of data: all authors. Drafting the article: all authors. Critically revising the article: Spinner, Maldonado. Reviewed submitted version of manuscript: Spinner, Maldonado. Approved the final version of the manuscript on behalf of all authors: Spinner. Administrative/technical/material support: Spinner. Study supervision: Spinner.

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