

Case Report

Peroneal Nerve Palsy Due to Subparaneurial Ganglion Cyst, a Rare Variant of Intraneural Ganglion Cyst

Abstract

Intraneural ganglion cysts are rare mucinous cysts originating within the epineurium of peripheral nerves. Although ganglion cysts are the most frequent tumors of the upper and lower extremities, ganglion cysts rarely result in peripheral nerve compression. We report a case of a 30-year-old patient who presented with foot drop due to subparaneurial ganglion cyst, a variant of an intraneural ganglion cyst. Characteristic magnetic resonance imaging findings were essential in the preoperative diagnosis of intraneural ganglion cyst. The common peroneal nerve and its branches were recognized and traced to its bifurcation during the operation. The articular branches were addressed. The mucinous content of the ganglion was typically found to be located within the subparaneurial compartment. Incision of the subparaneurial ganglion cyst was performed, and mucinous content was evacuated. At 2 months after the surgery, paralyzed peroneal nerve was recovered completely. Therefore, early diagnosis of intraneural ganglion, precise identification of the pathology, and proper treatment of the articular branch with atraumatic dissection of ganglion cyst are essential in the successful management of this rare lesion.

Keywords: Common peroneal nerve, cyst, ganglion, intraneural ganglion cyst, peroneal neuropathy

Introduction

Peroneal nerve palsy, the most common peripheral neuropathy of the lower extremity, has multiple causes. Although an extrinsic compression remains the most common cause, traumatic injuries, direct blunt trauma, metabolic diseases such as diabetes mellitus, prolonged bed rest, casting and bracing, and compressive mass lesions (both intraneural and extraneural lesions) may manifest with acute or progressive peroneal nerve neuropathy.^[1] The recent history of significant weight loss, habitual leg crossing, and prolonged squatting are known to be associated with increased risk of peroneal palsy and acute foot drop.^[2]

Intraneural ganglion cysts are rare, nonneoplastic, and mucinous cysts that originate within the epineurium of peripheral nerves.^[3-6] Synovial cyst (also known as ganglion cysts) deriving from synovial joints are well known.^[7] Ganglions are cystic structures lined by flat spindle-shaped cells that contain mucin or fluid.^[8] They may arise from joint capsules, ligaments, tendon sheaths, bursae,

or subchondral bone.^[8] These intraneural ganglion cysts typically result in neurologic deficit due to the displacement of nerve fascicles by cyst contents.^[3,9,10] They occur most commonly in the peroneal nerve. However, many nerves in the vicinity of synovial joints have also been described.^[7,10] The etiology of intraneural ganglion cysts has been poorly understood. Multiple surgical treatments strategies have been developed for intraneural ganglion cyst which has an estimated recurrence rate of 11%.^[11] Spinner *et al.*^[3,9] have introduced a comprehensive explanation for the pathogenesis of intraneural ganglion cyst. They suggested that a capsular defect or pedicle allows joint fluid to pass along an articular branch into the parent nerve. Intra-articular pressure then pushes the synovial fluid along the path with the least resistance, with extending the involvement along the nerve and its branches. The fluid propagation usually advances proximally. However, it can extend distally or in both directions.^[3,9] Here, we present a rare case of peroneal nerve palsy from an intraneural ganglion cyst and discussion on the current pathogenesis of unifying articular (synovial) theory.

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Case Report

A 30-year-old male patient presented a progressive weakness in dorsiflexion of the left foot and ankle for 2 months. In the beginning, vague discomfort was developed insidiously in the left anterolateral shin. He felt some difficulty in walking. This discomfort refrained him from walking or exercising for 2 weeks. However, weakness of dorsiflexion of the left foot was developed and gradually worsened. No pain or paresthesia was associated with the development of progressive weakness of the left foot and ankle dorsiflexion. During the past 2 years, he had a heavy physical exercise for baseball and soccer. However, he denied any physical trauma or accident that might have possibly affected the left knee and ankle. His family and medical histories were unremarkable. The patient was initially treated with nonsteroidal anti-inflammatory drugs with activity modification. However, the weakness of the left foot and ankle was progressively worsened. Complete foot drop developed eventually. He was referred for further evaluation.

On examination, there was grade 1+/5 weakness of ankle dorsiflexion and great toe extension in the left side. Neither restriction of lumbosacral range of motion nor complaints of low back pain was observed. Although minimal tenderness without swelling was present around the left fibular head, Tinel's sign was not evoked along the course of the peroneal nerve. There was no pain, spontaneously or evoked, in association with the passive movement of the knee or ankle. There was no abnormality in sensory examinations. Sciatic root stretch signs were negative.

An electromyogram (EMG) and nerve conduction study of the lower extremity showed a left peroneal nerve neuropraxia at the level of the fibular head. Magnetic resonance imaging (MRI) of the left knee showed a long segmental, high signal intensity lesion on T2-weighted image along the course of the distal common peroneal and proximal deep peroneal nerve [Figure 1]. The lesion appeared low signal intensity on T1-weighted images. It did not show any enhancement after gadolinium [Figure 1]. The high signal intensity in the tibialis anterior muscle suggestive of subacute denervation was not observed. X-ray findings of the left knee were nonspecific. Laboratory examinations including erythrocyte sedimentation rate, C-reactive protein, rheumatoid factor, antinuclear antibody, and creatinine kinase were normal. Considering the course of peroneal nerve palsy and abnormal MRI findings, an exploration of the peroneal nerve was performed.

Following the division of the superficial fascial layers, the common peroneal nerve was identified along its course around the fibular head. It continued distal and deep to the peroneus longus. The connective fascia superficial to the peroneus longus muscle and intermuscular septums were identified and divided. The common peroneal nerve was found to be entrapped by a dense and tight tendinous,

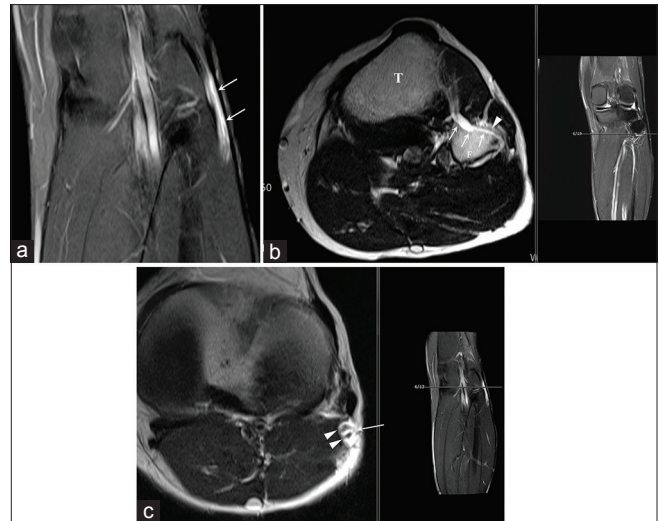


Figure 1: Magnetic resonance imaging of intraneural ganglion cyst involving the common peroneal nerve. (a) T2-weighted coronal image with fat suppression showing a tubular cyst (arrows) circumferentially surrounding the tibial and peroneal division within the distal sciatic nerve. (b) Axial T2-weighted FSE image at the level of the neck of the fibular showing intraneural cyst (arrows) within the articular branch (arrowhead) of the peroneal nerve with cyst extending into the proximal nerve branch to the tibialis anterior muscle, corresponding to the “transverse limb sign” F: Fibular, T: Tibia, TA: Tibialis anterior muscle. (c) Axial T2-weighted FSE image above the level of the fibular showing an intraneural ganglion cyst (arrow) in which the tibial and peroneal division are separately contained

posterior crural intermuscular septum. The nerve was decompressed and released by dividing posterior septum with a deep tendinous fascia beneath the peroneus longus as previously suggested.^[12] Meanwhile, the common peroneal nerve proximal to the musculoaponeurotic arch at the entrance to the fibular tunnel was swollen and encircled with loose connective tissue sheath of paraneurium [Figure 2]. The cystic content was gelatinous and milky, typical of the intraneural ganglion. However, the gelatinous content was located external to the epineurium of common peroneal nerve. Incision of the epineurium was not performed. The gelatinous content was removed after paraneuriotomy with gentle suction. The articular branch to the joint was addressed and divided.

The weakness in dorsiflexion of the toes and ankle was gradually improved by postoperative 2 weeks. It was completely recovered after 2 months. The patient's condition was stable without any recurrent symptom at the 12-month follow-up at an outpatient clinic.

Discussion

Peroneal nerve palsy and intraneural ganglion

Although peroneal nerve palsy is the most common entrapment neuropathy of the lower extremity, peroneal palsy is rarely associated with intraneural ganglion. Since the first case report of an intraneural ganglion of the common peroneal nerve in 1921, such lesion has been reported at various sites, including the ulnar,

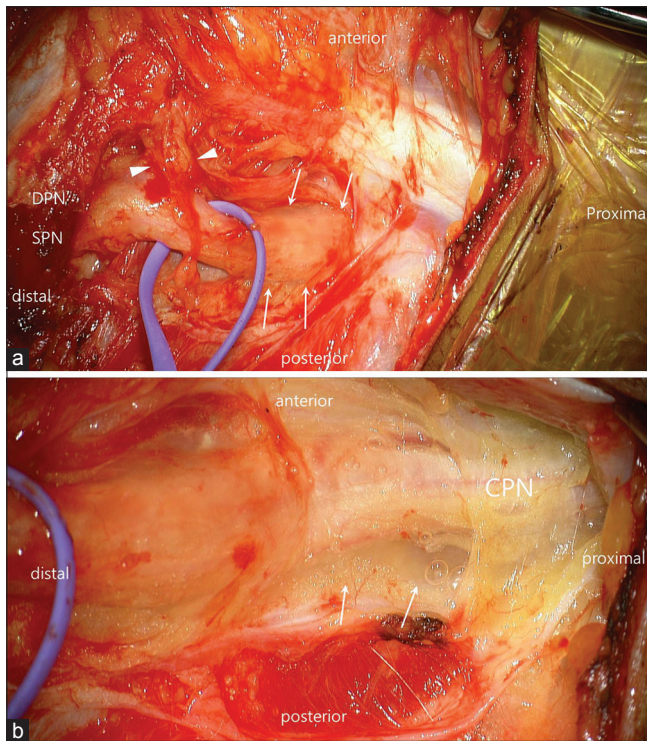


Figure 2: Intraoperative photographs showing the subparaneurial variant of intraneural ganglion cyst. (a) An intraoperative photograph showing the course of common peroneal nerve (arrows) after division of superficial and deep fascias of the peroneus longus muscle. The common peroneal nerve is swollen and encircled by a mucoid, gelatinous cyst. The articular branch of the common peroneal nerve (arrowheads) is addressed. DPN: Deep peroneal nerve, SPN: Superficial peroneal nerve. (b) An intraoperative photograph showing the common peroneal nerve and subparaneurial ganglion cyst. The mucoid, gelatinous content (arrows) was typically located in the subparaneurial space, outside the epineurium of the common peroneal nerve

radial, median, sciatic, tibial and posterior interosseous nerves, all of which tend to occur adjacent to the joint or bursa.^[9,13] Considering these universal occurrence of intraneural ganglia near joints and their tendency to extend proximally, the high percentage of preceding trauma of knee or superior tibiofibular joint (STFJ) abnormalities, the predominance of deep peroneal nerve dysfunction sparing of superficial peroneal nerve, and the frequent finding of a pedicle to the STFJ, Spinner *et al.*^[3,9,14] have suggested a unifying articular (synovial) theory as the etiology for intraneural ganglion cysts. The unifying articular theory describes that intraneural ganglion cysts originate from a neighboring synovial joint. They then propagate thorough a capsular rent into an articular branch and the proximal, parent nerve. Cyst (joint) fluid follows the path with the least resistance. The shape and size of the cyst depend on pressure and pressure fluxes. This unifying articular theory has been substantiated and repeatedly replicated by others in recent years.^[13,15,16]

Intraneural and subparaneurial ganglions

In the diagnosis of intraneural ganglion cysts, high-resolution MRI is essential for morphologic

classification of intraneural ganglion cysts.^[17] Two imaging signs, the signet ring sign^[18] and the transverse limb sign,^[10] have been suggested to be extremely robust in the differential diagnosis of intraneural ganglion cysts and more common extraneural variants.^[17]

Subparaneurial ganglion cyst, a variant of an intraneural ganglion cyst, was first reported by Prasad *et al.*^[17] The subparaneurial compartment is known to be formed by multilaminar sheaths of collagen interposed with adipose that encompasses the nerve completely or partially at different parts along its course.^[17,19-21] Based on their experience with literature review, Prasad *et al.*^[17] have suggested that subparaneurial cyst could have a complex lobulated morphology like intraneural cyst and intraneural ganglion cyst. It can progress from a subepineurial compartment to a subparaneurial compartment. As a pathogenic mechanism, weakness in the epineurium and direct infiltration of the subparaneurial compartment have been suggested.^[17] In the present case, the gelatinous mucinous content of the ganglion cyst was typically located in the subparaneurial compartment [Figure 2]. However, the MRI appearance of subparaneurial cyst in the current case did not show a typical pattern of multi-lobulated morphology. It was cylindrically tubular, encircling the common peroneal nerve. Although the transverse limb sign was observed, the signet ring sign was not observed [Figure 1].

Treatment of intraneural ganglion cyst

Treatment option for peroneal nerve palsy due to peroneal intraneural ganglion is surgery. No authors have recommended conservative treatment because surgical treatment is successful in most cases when it is performed early.^[13] If surgical treatment is delayed, intraneural growth and invasion may cause irreversible axonal damage, resulting in poor outcome of foot drop.^[13] Young age, the absence of nerve damage, and duration of symptoms have been reported to be associated with clinical outcomes.^[13] Patients with foot drop for <4 months have shown favorable outcomes.^[13,22-24] If there is denervation of the tibialis anterior muscle in preoperative EMG examination, complete resolution of peroneal palsy is less likely.^[13]

The surgical procedure involves exploration and decompression of the peroneal nerve.^[13] However, the technique of surgical treatment for intraneural ganglion cysts has been controversial due to the contention surrounding their pathogenesis.^[11] In line with the articular theory,^[3,9,14] Spinner *et al.*^[11,25] have suggested the 4D technique (dissection of the nerve, disarticulation of the tibiofibular joint, decompression of the cyst, and disconnection of the articular branch) to treat articular branch connection and/or the joint with.^[25] Failure to disconnect the articular branch or treat the joint pathology has been found to be a statistically significant factor for recurrence as high as 30%.^[3,9,11,25,26] In addition, treating the

superior tibiofibular joint, the origin of the ganglion cyst, has become a critical part of surgery for STFJ-associated intraneural cyst.^[11] According to Desy *et al.*,^[11] addressing the joint connection and/or the joint pathology might even obviate the need for cyst/nerve decompression. If cyst decompression is attempted, simple cyst incision and evacuation of the mucinous fluid are recommended to avoid iatrogenic nerve injury instead of using more radical procedures such as full cyst excision.^[11]

Conclusions

A rare case of subparaneurial variant of the intraneural ganglion cyst is reported here. MRI examination and understanding the pathophysiology of intraneural ganglion cyst are essential for the early diagnosis and successful treatment of peroneal nerve palsy due to ganglion cyst.

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Conflicts of interest

There are no conflicts of interest.

References

- Poage C, Roth C, Scott B. Peroneal nerve palsy: Evaluation and management. *J Am Acad Orthop Surg* 2016;24:1-0.
- Masakado Y, Kawakami M, Suzuki K, Abe L, Ota T, Kimura A, *et al.* Clinical neurophysiology in the diagnosis of peroneal nerve palsy. *Keio J Med* 2008;57:84-9.
- Allieu PY, Cenac PE. Peripheral nerve mucoid degeneration of the upper extremity. *J Hand Surg Am* 1989;14:189-94.
- Giele H, Le Viet D. Intraneural mucoid cysts of the upper limb. *J Hand Surg Br* 1997;22:805-9.
- Spinner RJ, Atkinson JL, Tiel RL. Peroneal intraneural ganglia: The importance of the articular branch. A unifying theory. *J Neurosurg* 2003;99:330-43.
- Naam NH, Carr SB, Massoud AH. Intraneural ganglions of the hand and wrist. *J Hand Surg Am* 2015;40:1625-30.
- Capek S, Koutlas IG, Strasia RP, Amrami KK, Spinner RJ. An inferior alveolar intraneural cyst: A case example and an anatomical explanation to support the articular theory within cranial nerves. *J Neurosurg* 2015;122:1433-7.
- Janzen DL, Peterfy CG, Forbes JR, Tirman PF, Genant HK. Cystic lesions around the knee joint: MR imaging findings. *AJR Am J Roentgenol* 1994;163:155-61.
- Spinner RJ, Atkinson JL, Scheithauer BW, Rock MG, Birch R, Kim TA, *et al.* Peroneal intraneural ganglia: The importance of the articular branch. Clinical series. *J Neurosurg* 2003;99:319-29.
- Spinner RJ, Desy NM, Amrami KK. Cystic transverse limb of the articular branch: A pathognomonic sign for peroneal intraneural ganglia at the superior tibiofibular joint. *Neurosurgery* 2006;59:157-66.
- Desy NM, Wang H, Elshiekh MA, Tanaka S, Choi TW, Howe BM, *et al.* Intraneural ganglion cysts: A systematic review and reinterpretation of the world's literature. *J Neurosurg* 2016;125:615-30.
- Boyd KU, Brown JM. Injury and compression neuropathy in the lower extremity. In: Mackinnon SE, editor. *Nerve Surgery*. New York: Thieme; 2015. p. 338-90.
- Muramatsu K, Hashimoto T, Tominaga Y, Tamura K, Taguchi T. Unusual peroneal nerve palsy caused by intraneural ganglion cyst: Pathological mechanism and appropriate treatment. *Acta Neurochir (Wien)* 2013;155:1757-61.
- Spinner RJ, Amrami KK, Wolanskyj AP, Desy NM, Wang H, Benarroch EE, *et al.* Dynamic phases of peroneal and tibial intraneural ganglia formation: A new dimension added to the unifying articular theory. *J Neurosurg* 2007;107:296-307.
- Isaacs AM, Midha R, Desy NM, Amrami KK, Spinner RJ. The mechanism underlying combined medial and lateral plantar and tibial intraneural ganglia in the tarsal tunnel. *Acta Neurochir (Wien)* 2016;158:2225-9.
- Sobol GL, Lipschultz TM. Successful surgical treatment of an intraneural ganglion of the common peroneal nerve. *Am J Orthop (Belle Mead NJ)* 2015;44:E123-6.
- Prasad NK, Desy NM, Howe BM, Amrami KK, Spinner RJ. Subparaneurial ganglion cysts of the fibular and tibial nerves: A new variant of intraneural ganglion cysts. *Clin Anat* 2016;29:530-7.
- Spinner RJ, Luthra G, Desy NM, Anderson ML, Amrami KK. The clock face guide to peroneal intraneural ganglia: Critical "times" and sites for accurate diagnosis. *Skeletal Radiol* 2008;37:1091-9.
- Vloka JD, Hadzić A, Lesser JB, Kitain E, Geatz H, April EW, *et al.* A common epineural sheath for the nerves in the popliteal fossa and its possible implications for sciatic nerve block. *Anesth Analg* 1997;84:387-90.
- Macchi V, Tiengo C, Porzionato A, Parenti A, Stecco C, Bassetto F, *et al.* Musculocutaneous nerve: Histotopographic study and clinical implications. *Clin Anat* 2007;20:400-6.
- Andersen HL, Andersen SL, Tranum-Jensen J. Injection inside the paraneural sheath of the sciatic nerve: Direct comparison among ultrasound imaging, macroscopic anatomy, and histologic analysis. *Reg Anesth Pain Med* 2012;37:410-4.
- Spinner RJ, Carmichael SW, Wang H, Parisi TJ, Skinner JA, Amrami KK, *et al.* Patterns of intraneural ganglion cyst descent. *Clin Anat* 2008;21:233-45.
- Al Mufargi YS, Mouch CA, Ziebarth K, Joeris A, Slongo T. An unusual cause of paralysis of the peroneal nerve: A report of 3 cases. *J Pediatr Orthop* 2011;31:e50-2.
- Lowenstein J, Towers J, Tomaino MM. Intraneural ganglion of the peroneal nerve: Importance of timely diagnosis. *Am J Orthop (Belle Mead NJ)* 2001;30:816-9.
- Spinner RJ, Desy NM, Rock MG, Amrami KK. Peroneal intraneural ganglia. Part II. Lessons learned and pitfalls to avoid for successful diagnosis and treatment. *Neurosurg Focus* 2007;22:E27.
- Cobb CA 3rd, Moiel RH. Ganglion of the peroneal nerve. Report of two cases. *J Neurosurg* 1974;41:255-9.