

Intraneural ganglion cyst: a case report and a review of the literature

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Background. An intraneural ganglion cyst is a non-neoplastic gelatinous cyst, which can be found within the epineurium of a peripheral nerve and lead to signs and symptoms of peripheral neuropathy. Early diagnosis and appropriate surgical treatment of this condition is a key to a successful outcome.

Materials and methods. We describe a case of a patient with peroneal nerve neuropathy caused by an intraneural ganglion cyst. Postsurgically, only partial relieve of the symptoms was achieved because of delayed diagnosis.

Results and conclusions. The aetiology of intraneural ganglia is controversial and historically different pathogeneses of this condition have been described. Diagnosis of these cysts is based on MRI. Multiple surgical treatment strategies have been developed based on different pathogenesis of the formation of these cysts, but neuropathy is reversible only, if this condition is diagnosed and treated early.

Keywords: Intraneural ganglion cyst, peroneal neuropathy, unifying articular theory

INTRODUCTION

An intraneural ganglion cyst is a non-neoplastic gelatinous cyst, which can be found within the epineurium of a peripheral nerve (1). Intraneural ganglion cysts typically lead to signs and symptoms of peripheral neuropathy because of displacing and compression of the nerve fascicles (2). It is important to distinguish this condition from other possible causes of peripheral nerve compression because early

detection and diagnosis lead to better results after surgery (3). The pathogenesis of this condition is controversial and historically it was not completely understood. There are multiple theories regarding formation of these cysts. As a result of various theories, multiple surgical treatment strategies have been developed based on different pathogenesis of the formation of these cysts (4). It led to unsatisfying post-operative results because of high rates of recurrence. During the past few years, substantial evidence has been presented to support the articular (synovial) theory for the pathogenesis and findings observed on magnetic resonance imaging (MRI) (5). We describe a case of a 64-year-old patient with peroneal nerve neuropathy caused by intraneural ganglion.

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Unfortunately, only partial relieve of the symptoms was achieved because of delayed diagnosis.

CASE PRESENTATION

In November 2017, a 64-year-old female patient presented with pain in the left knee and the lateral side of the left calf, and weakness in the left foot. Pain occurred periodically during the past 15 years. During the last three years, the pain was persistent, progressively getting worse. Despite persistent pain, the patient did not visit any health institution for several years. Weakness of dorsiflexion of the left foot developed two years ago. The patient visited her general practitioner, and lumbar radiculopathy was suspected. However, the weakness in the left foot and ankle progressively worsened. Eventually, a complete foot drop developed in August 2017. MRI was performed and intraneural ganglion of the peroneal nerve was suspected. The patient was subsequently referred to our clinic.

A neurological examination revealed profound motor weakness in the left foot – the absence of foot dorsiflexion and finger extension. In addition, hypoaesthesia in the anterolateral side of the left foot in the region of the deep peroneal nerve was observed.

No electromyogram (EMG) was performed. MRI showed a lobular cystic lesion (ganglion cyst) in the left peroneal canal.

Decompression and neurolysis surgery under general anesthesia was performed, during which the common peroneal nerve expanded into an irregular sausage-like shape of over 10 cm in length, from the posterior border of the long peroneal muscle to the back of the knee. No connection of the cyst with the tibiofibular joint was found. The membrane surrounding the cyst was microsurgically dissected and opened parallel to the course of the nerve (Figure), and decompression was performed. The cyst contained light-yellowish gelatinous fluid that it was sent for histopathological examination.

Histopathological examination confirmed the presence of histiocytes and did not detect any tumour cells. The findings were described as reactive changes.

The pain disappeared but the loss of sensation and weakness persisted after surgery.

No follow up was conducted due to lack of contact with the patient.

DISCUSSION

Peroneal nerve palsy is one of the most common peripheral neuropathies of the lower extremity and it has multiple causes. External compression remains the most common cause, but traumatic injuries, direct blunt trauma, metabolic diseases,

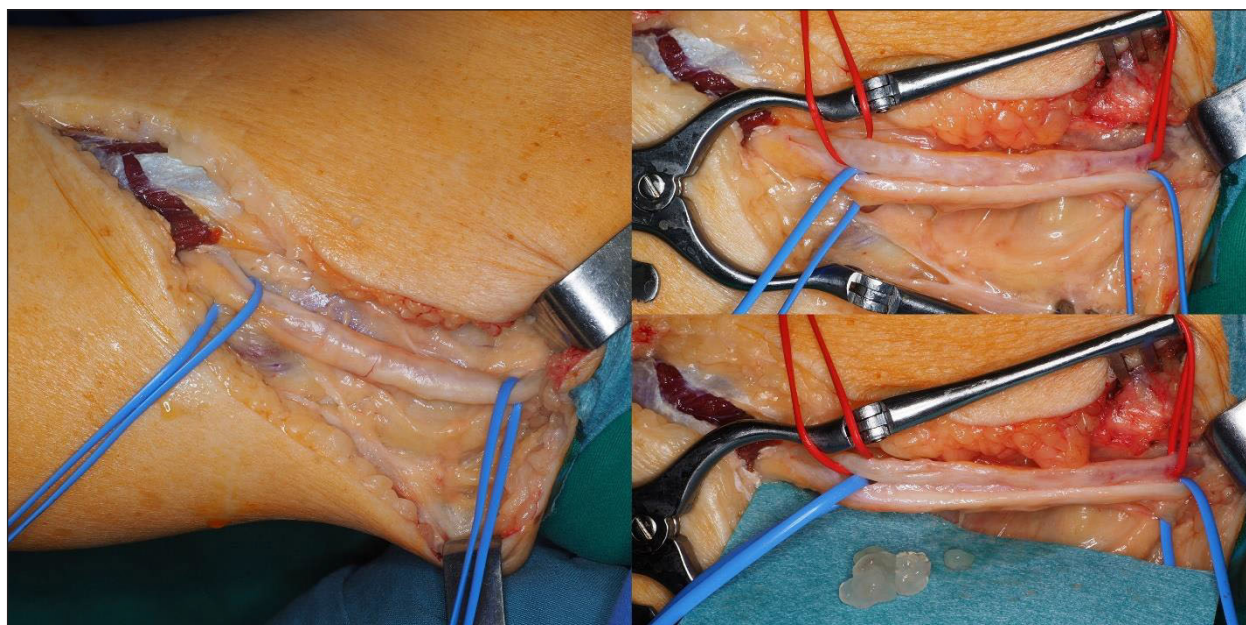


Figure. Intraoperative view of the dissected peroneal nerve, before microsurgical dissection (left), after microsurgical fascicles dissection (upper right), and ganglion decompression (lower right)

prolonged bed rest, and tight casting and bracing may also manifest with acute or progressive peroneal nerve neuropathy (6). However, there are some unusual cases of peroneal nerve palsy like intraneural ganglia.

Intraneural ganglion cysts are usually defined as non-neoplastic mucinous cysts within the peripheral nerve, which are connected to the adjacent joint. Any peripheral nerve can be affected, but the most common location is the peroneal nerve (7). Ganglia are cystic structures lined by flat spindle-shaped cells that contain mucin or fluid. Ganglia may arise from joint capsules, ligaments, tendon sheaths, bursae, or subchondral bone (8). The main difference between intraneural and other ganglia is their relation to the nerve. Since the 1921, when the first case of an intraneural ganglion of the common peroneal nerve was reported, the same lesion has been described at a variety of other sites: the ulnar, radial, median, sciatic, tibial and posterior interosseous nerves (9). All of them occurred adjacent to the joint or bursa.

The aetiology of intraneural ganglia is controversial and different pathogeneses of this condition have been historically described. Some of them, like degenerative and tumoural theories, described intraneural ganglia as cysts, which arise within the nerve sheath. Other theories proposed the concept that extraneural ganglia could invade epineurium, leading to intraneural ganglia formation (10). These theories were not sufficient to explain all cases. Considering the observation of certain typical clinical features of peroneal intraneural ganglia, such as their location near superior tibiofibular joints, the tendency to extend proximally, high occurrence of adjacent joint trauma and degenerative joint disease, predominance of deep peroneal nerve deficit and frequent finding of a pedicle to the joint, Spinner et al. suggested a unifying articular (synovial) theory with substantial evidence of the formation of these cysts.

According to this theory, articular fluid travels through capsular rents from the joint directly into the epineurium of articular branch, where it follows the path of least resistance proximally, up the deep peroneal branch and the deep peroneal portion of the common peroneal nerve (11). They identified three sequential phases of peroneal intraneural ganglia cyst formation: primary ascent,

sciatic nerve cross-over, and terminal branch descent (12). The evidence of the articular branch from the adjacent superior tibiofibular joint was observed by MRI in a multi-centre clinical case series of 24 patients (13). In 2015, Desy et al. performed a large systematic review of the literature and MR images on intraneural cysts which further supported the unifying articular (synovial) theory and retrospectively confirmed joint connections in 27 of 79 case reports that were previously unrecognized by authors. Trauma was identified in 13% of cases, and most of the joints associated with intraneural cysts were degenerative (14).

Diagnosis of these cysts is based on MRI. They are small in size and have a typical tubular configuration along the course of the involved nerve or its branches. Sometimes the joint connection can be well observed (3). The differential diagnoses for these cysts may include nerve sheath tumours, atypical Baker's cyst, and extraneural ganglion. An atypical vascular or lymphatic malformation could be also considered (15).

Surgical treatment is undoubtedly the first-choice treatment for a peroneal intraneural ganglion. No authors have recommended conservative treatment because, when performed early, surgical treatment is usually successful (10). Surgical exploration and decompression of the peroneal nerve is the most frequently procedure. Spinner et al. proposed the 4D technique: dissection of the nerve, disarticulation of the tibiofibular joint, decompression of the cyst, and disconnection of the articular branch (13). However, traumatic dissection of the nerve to perform radical resection of the ganglion is in many cases associated with a higher risk of fascicular damage. The paralyzed peroneal nerve is capable of recovering even when a residual ganglion is present after surgery (10). The main point of surgical treatment to prevent recurrent cyst formation is identification and disconnection of the articular nerve branch. A recurrence rate of 13% was identified following isolated cyst decompression without articular branch disconnection, whereas primary surgery consisting of articular branch ligation or disconnection led to 6% recurrence rate. No intraneural recurrence was observed after surgical procedures that addressed the adjacent joint (5). However, disarticulation of the joint may also be an unnecessary and traumatic procedure leading to delayed

recovery. Muramatsu et al. proposed five key points for the successful treatment of peroneal intraneural ganglion: correct early diagnosis by MRI, surgery within four months of the occurrence of foot drop, simple exoneural dissection, microsurgical epineurotomy and drainage of the cyst, and disruption of the articular branch (10).

CONCLUSIONS

Intraneural ganglion cysts can be differentiated from other peripheral nerve lesions and effectively diagnosed by MRI. The neuropathy is reversible if this condition is diagnosed and treated early. To prevent recurrence and further cyst formation, it is very important to find the articular connection and disconnect it during surgery (6, 8, 15).

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References

- Panwar J, Mathew A, Thomas BP. Cystic lesions of peripheral nerves: are we missing the diagnosis of the intraneural ganglion cyst? *World J Radiol.* 2017 May 28; 9(5): 230–44.
- Desy NM, Wang H, Elshiekh MAI, 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(3): 615–30.
- Lang CJ, Neubauer U, Qaiyumi S, Fahlbusch R. Intraneural ganglion of the sciatic nerve: detection by ultrasound. *J Neurol Neurosurg Psychiatry.* 1994 Jul; 57(7): 870–1.
- Desy NM, Lipinski LJ, Tanaka S, Amrami KK, Rock MG, Spinner RJ. Recurrent intraneural ganglion cysts: pathoanatomic patterns and treatment implications. *Clin Anat N Y N.* 2015 Nov; 28(8): 1058–69.
- Tehli O, Celikmez RC, Birgili B, Solmaz I, Celik E. Pure peroneal intraneural ganglion cyst ascending along the sciatic nerve. *Turk Neurosurg.* 2011; 21(2): 254–8.
- Kim D, Choi J-G, Son B-C. Peroneal nerve palsy due to subparaneurial ganglion cyst, a rare variant of intraneural ganglion cyst. *Asian J Neurosurg.* 2018; 13(4): 1225–8.
- Spinner RJ, Carmichael SW, Wang H, Parisi TJ, Skinner JA, Amrami KK. Patterns of intraneural ganglion cyst descent. *Clin Anat N Y N.* 2008 Apr; 21(3): 233–45.
- Janzen DL, Peterfy CG, Forbes JR, Tirman PF, Genant HK. Cystic lesions around the knee joint: MR imaging findings. *AJR Am J Roentgenol.* 1994 Jul; 163(1): 155–61.
- 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 Sep; 155(9): 1757–61.
- Spinner RJ, Scheithauer BW, Amrami KK. The unifying articular (synovial) origin of intraneural ganglia: evolution-revelation-revolution. *Neurosurgery.* 2009 Oct; 65(4 Suppl): A115–124.
- 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 Aug; 107(2): 296–307.
- Spinner RJ, Atkinson JLD, Scheithauer BW, Rock MG, Birch R, Kim TA, et al. Peroneal intraneural ganglia: the importance of the articular branch. Clinical series. *J Neurosurg.* 2003 Aug; 99(2): 319–29.
- Keser N, Akpınar P, Is M, Aktas I. Irreversible foot-drop as a consequence of neglected knee pain in an adolescent with a peroneal intraneural ganglion cyst. *World Neurosurg.* 2018 Mar; 111: 307–10.
- 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 Aug; 107(2): 296–307.
- Liang T, Panu A, Crowther S, Low G, Lambert R. Ultrasound-Guided Aspiration and Injection of an Intraneural Ganglion Cyst of the Common Peroneal Nerve. *HSS J.* 2013 Oct; 9(3): 270–4.

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INTRANEURINIS GANGLIONAS: KLINIKINIO ATVEJO PRISTATYMAS IR LITERATŪROS APŽVALGA

Santrauka

Tikslas. Intraneurinis ganglionas – tai nepyktybinis, „drebučių“ konsistencijos turiniu užpildytas mucininis darinys, randamas periferinių nervų viduje, ilgainiui sukeliantis periferinės kompresinės neuropatijos simptomus ir požymius. Ankstyva diagnozė ir tinkamas chirurginis gydymas – esminiai veiksniai, siekiant sėkmingo ligos gydymo.

Medžiaga ir metodai. Straipsnyje pateikiamas šėivinio nervo intraneurinio gangliono sukeltos kompresinės neuropatijos klinikinis atvejis. Po chirurginio gydymo pasiektas tik dalinis pagerėjimas dėl per vėlai nustatytos diagnozės.

Rezultatai ir išvados. Intraneurinių ganglionų etiologija nėra visiškai aiški, todėl sukurta keletas teorijų, paaiškinančių šių darinių patogenezę. Diagnostika remiasi magnetinio rezonanso tomografijos tyrimu. Literatūroje randama daug šios patologijos gydymo būdų, atsižvelgiant į įvairias patogenezės teorijas. Ligos simptomai neatsinaujina tik esant ankstyvai diagnozei ir gydymui.

Raktažodžiai: intraneurinis ganglionas, šėivinio nervo neuropatija, apibendrinanti sąnarinė teorija