E-ISSN: 2707-8353
P-ISSN: 2707-8345
IJCRO 2022; 4(1): 132-135
Received: 08-05-2022
Accepted: 10-06-2022

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Peroneal intraneural ganglion cyst

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DOI: https://doi.org/10.22271/27078345.2022.v4.i1b.105

Abstract
Peroneal Intraneural Ganglion Cyst is a rare condition with very few cases reported in literature. A 55 year old middle-aged lady presented with complaints of pain over lateral aspect of right knee and leg for the previous one month. She also complained about the inability to extend the ankle and the toes for one week. There was no recent history of trauma. She had noticed the tingling and numbness over right foot for the last one month. The typical radiological features and atypical clinical course, favored the diagnosis of Peroneal Intraneural Ganglion Cyst. After all necessary investigations, she underwent definitive management, i.e. Dissection of the Common peroneal nerve along with Neurolysis, decompression of the Ganglion Cyst, isolation and disconnection of the articular branch of the nerve and Disarticulation of the proximal Tibiofibular Joint. At the latest follow up (6 months) she has completely recovered from her foot drop and there are no signs of recurrence of the ganglion cyst.

Keywords: Peroneal intraneural ganglion cyst, ganglion, cyst

Introduction
Ganglion cysts are benign mucinous lesions that typically occur near joints that by their location and size may affect neighboring nerves. These cysts may be classified as intraneural or extra-neural. Although rare, intraneural ganglia are located within the substance of the nerve and may cause direct nerve compression. Extra-neural ganglions are more common which may extrinsically compress nerves [1].

Misdiagnosis of intraneural and extra-neural ganglia, including failure to identify the joint origin, are common problems. Preoperative distinction of these entities is both possible and important for effective patient management. Intraneural ganglia are uncommon and radiologists and surgeons may encounter them only very rarely, if at all, in their careers. As a result, the often-small joint connection may be unrecognized and untreated, resulting in a high rate of recurrence [2].

The pathogenesis of peroneal intraneural ganglia, long a source of controversy, is becoming clear. Typical pathoanatomic features have been substantiated to support a joint origin for these cysts i.e the unifying articular (synovial) theory [1, 2].

Predictable magnetic resonance imaging (MRI) findings can demonstrate the consistent anatomic findings and help identify the type of cyst (intraneural or extra-neural) and a joint connection [3].

MRI therefore can clarify the pathogenesis of these cysts. Central to our understanding of the propagation of these peroneal intraneural cysts is their U or J shaped articular branch that serves as a conduit for cyst fluid to dissect from the superior tibiofibular joint and dependent upon intra-articular pressures ascend preferentially up the common peroneal nerve (and at times into the sciatic nerve) rather than descend down the terminal branches of the peroneal nerve [4].

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 postoperative 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 MRI [5].

We describe a case of a 55-year-old patient with peroneal nerve neuropathy caused by intraneural ganglion cyst. Full relief of the symptoms was achieved because of accurate early diagnosis and appropriate surgical intervention.

The purpose of this article is to describe the clinical and radiological features of Peroneal Intraneural Ganglion Cyst, and also focus on the treatment options best suited to treat it.

Case presentation

A 55-year-old female patient presented with pain in the right knee and the lateral side of the right calf, and weakness in the right foot. Pain occurred intermittently during the previous 1 month. It was persistent, and progressively getting worse. Weakness of dorsiflexion of the right foot developed one week prior to her seeking consultation. The patient visited her general practitioner, and lumbar radiculopathy was suspected. However, the weakness in the right foot and ankle progressively worsened. Eventually, a complete foot drop developed (Fig 1 & Fig 2). The patient presented to our Outpatient Department at this stage. On Clinical examination it was seen that no thickening of the CPN was found but there was a boggy swelling over the lateral aspect of the knee. Sensation was altered but present over L4, L5 and S1 dermatome. EHL/EDL/TA was 1/5, FHL/FDL/TP was 4/5. X rays revealed degenerative Osteo-arthritic changes in the knee joint. A neurological examination revealed profound motor weakness in the right foot – the absence of foot dorsiflexion and finger extension. In addition, hypoesthesia in the anterolateral side of the right foot in the region of the deep peroneal nerve was observed. Also, a high stepping gait was noted because of right sided foot drop. MRI was performed and intraneural ganglion of the peroneal nerve was suspected (Fig 3A & B). No electromyogram (EMG) was performed. MRI showed a lobular cystic lesion (ganglion cyst) in the right peroneal canal.

Surgery consisting of Dissection of the CPN with neurolysis, disconnection of the articular branch of the CPN, Decompression of the ganglion cyst and Disarticulation of Proximal Tibiofibular Joint was under spinal anesthesia was proposed as per technique described by Spinner *et al*., Mura-Matsu *et al*. (ref) (Fig 4, 5 & 6). With the patient in lateral decubitus position and under tourniquet control, a Curvilinear incision was made along the course of the CPN. Dissection of the CPN was done to identify the connection of the cyst with the tibiofibular joint. The membrane surrounding the cyst was micro surgically dissected and opened parallel to the course of the nerve, and decompression was performed. The cyst contained whitish gelatinous fluid. The pain disappeared almost immediately post operatively but the loss of sensation and weakness persisted for few months after surgery. Foot Drop splint was applied after the surgery. Follow up was conducted at regular intervals, which showed gradual recovery of ankle dorsiflexion power. At the latest FU (6 Months) there is no neurological deficit. Patient can do full dorsiflexion of the right ankle and toes and the high stepping gait has disappeared. (Fig 7).

Fig 1: At Full plantar flexion. Preoperative Clinical Picture of the patient

Fig 2: At Full Dorsiflexion. Preoperative Clinical Picture of the patient. Foot Drop noted

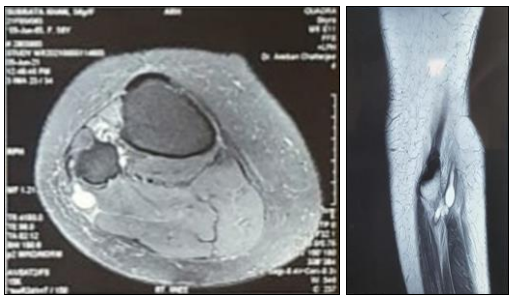

Fig 3 A & B: Diffuse thickening and signal alteration of the common peroneal nerve in the popliteal Fossa. Appear Cystic in nature

Fig 4: Intra op picture showing Partial Disarticulation of proximal tibio-fibular joint being done. Joint space is pointed.

Fig 5: intra operative picture showing Epineurotomy being done. Cyst material visualized. Suctioned.

Fig 6: Intra operative picture showing the CPN after completion of decompression

Fig 7: 6 months Follow Up. Patient can do near full Dorsiflexion of the right ankle

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, chronic granulomatous diseases like leprosy, 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 etiology of intraneural ganglia is controversial and different pathogeneses of this condition have been historically described. Some of them, like degenerative and tumoral theories, described intraneural ganglia as cysts, which arise within the nerve sheath. Other theories proposed the concept that extra-neural 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 multicenter clinical case series of 24 patients [13]. In 2015, Desy *et al*. performed a large systematic review of 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 tumors, atypical Baker's cyst, and extra-neural 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 rewarding [10]. Surgical exploration and decompression of the peroneal nerve is the most frequently performed 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 [2, 3]. 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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