

신경 내 결절종에 의한 총비골 신경병증: 2예에서 관찰된 전리생리학적 소견에 근거한 신경 섬유속 손상 평가와 수술소견

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Common Peroneal Neuropathy Caused by Intraneural Ganglion Cyst, Electrophysiological Findings Related to Fascicular Involvement and Operative Findings in Two Cases

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Foot drop can occur as a manifestation of a number of pathological and clinical conditions and is a fairly common disability encountered in electrodiagnostic clinics. This report describes two cases of atraumatic foot drop. Both cases were electrophysiologically diagnosed as common peroneal neuropathy around the fibular head. Radiographic and surgical findings confirmed intraneural ganglion cyst of common peroneal nerve and the surgical resection was performed. Although the radiographic and surgical findings were compatible with the lesion of common peroneal nerve, the precise analysis of the electrophysiologic findings could differentiate the fascicular involvement in two cases.

Keywords: Common peroneal neuropathy, Electrodiagnosis, Intraneural ganglion cyst

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Introduction

The common peroneal nerve (CPN) is the most commonly damaged nerve in the lower extremity [1]. The nerve damage mostly occurs around the fibular head level, where the nerve is the most vulnerable as it passes through fibular tunnel and due to

its superficial location. Common peroneal neuropathy at the fibular head can result from various causes such as trauma (e.g. fracture, forcible stretch injury), prolonged compression (e.g. casting, surgical procedure under heavy sedation) and structural mass lesions, either intrinsic or extrinsic [1]. Although it is a rare entity, ganglion cyst can act as a mass lesion that can lead to pe-

ripheral neuropathies. Hereby, we report two cases of common peroneal neuropathy caused by intraneural ganglion cyst (IG) and their detailed neurophysiological findings.

Case reports

Case 1

A 60-year old male patient presented with 2-month history of atraumatic right foot drop. Before the foot drop started, he had experienced pain in the right upper lateral calf region 3 months ago but did not seek medical attention then. He had no history of low back pain nor the radiation of pain into his hip or thigh. On the physical examination, the Medical Research Council (MRC) scale for right ankle dorsiflexion and great toe extension were grade 0 and right ankle inversion, knee extensor and hip abductor were grade 5. Diminished light sensation on the right first webspace was noted but other areas of the right foot dorsum and the distributions of sural and saphenous nerve were intact. There was no palpable mass lesion but the Tinel's sign over the right fibular head was positive. The straight leg raise (SLR) test was

negative, bilaterally.

The electrophysiological study was examined in the standard method with the segmental conduction study of CPN. In the nerve conduction study (NCS) (Table 1), right peroneal compound muscle action potential (CMAP) and deep peroneal sensory nerve action potential (SNAP) were unobtainable. Right mixed common peroneal and superficial peroneal SNAPs were within the normal range. In needle electromyography (EMG) (Table 1), fibrillation potentials, positive sharp waves (F&P) and no motor unit potential were noted in the deep peroneal nerve (DPN) innervated muscles but superficial peroneal nerve innervated muscles, peroneus longus, revealed normal findings. Sonography and magnetic resonance image (MRI) demonstrated lobulating cystic lesion along the course of CPN and the proximal portion of DPN around the fibular head (Fig. 1A and B). Electrodiagnostic examination (EDX) combined with radiographic findings suggested right complete neuropathy of deep peroneal portion of CPN. Excision of the IG, resection of the articular branch of CPN, and neurolysis were performed. And the surgery confirmed the articular branch to the proximal tibiofibular joint and the stage III peroneal IG, which extended proximally to the above fibular head level (Fig. 2) [2]. In a follow-up study at 3-month after the surgery, there was no interval change in NCS, but electrophysiological evidence of reinnervation taking place at the right extensor digitorum longus was noted in the needle EMG. In addition, the MRC scale of right ankle dorsiflexion and great toe extension were improved to grade 2. In the follow-up study at 6-month after the surgery, the peroneal motor response at tibialis anterior recording was evoked with prolonged latency and low amplitude and reinnervation evidences were

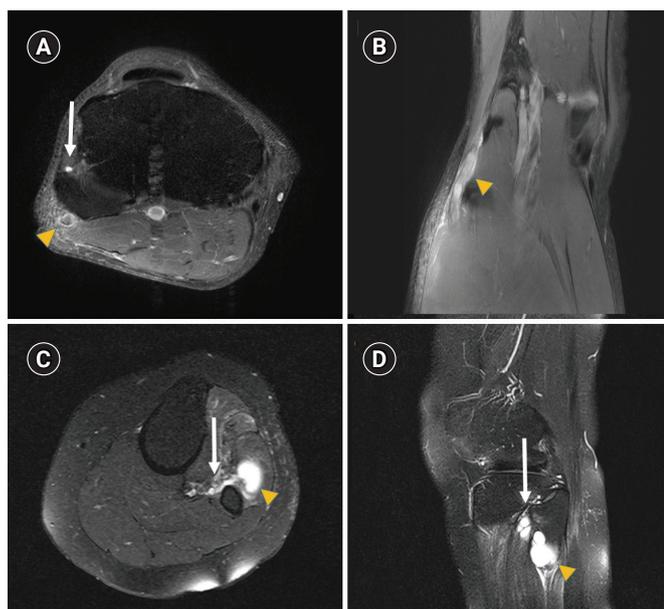


Fig. 1. T2-weighted magnetic resonance images of the knee in case 1 (A) axial, (B) coronal view and case 2 (C) axial, (D) coronal view. MRI of case 1 (A and B) demonstrates the lobulated cystic lesion of intraneural ganglion cyst (arrowhead) extending along the right common peroneal nerve and arising from the proximal tibiofibular joint (arrow). MRI of case 2 (C and D) demonstrates the multi-lobulating cystic lesion of intraneural ganglion cyst (arrowhead) extending along the left common peroneal nerve and arising from the proximal tibiofibular joint (arrow).

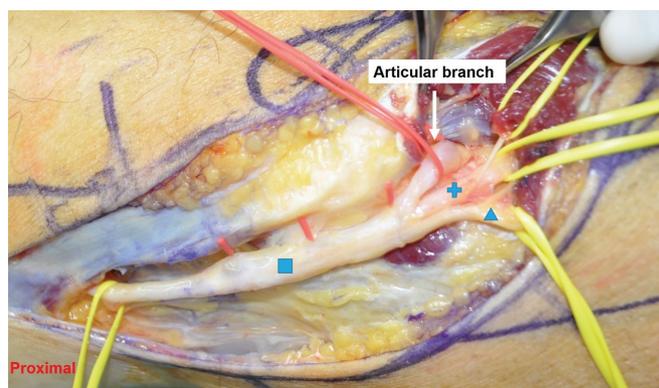


Fig. 2. Intraoperative clinical photographs of case 1, showing the right common peroneal nerve trifurcating into superficial peroneal nerve (triangle), deep peroneal nerve (cross) and enlarged articular branch (arrow) and intraneural ganglion invading the common peroneal nerve (square), intraneural ganglion cyst stage III.

Table 1. Nerve Conduction and Needle Electromyography Data (Case 1)

Nerve conduction (Pre-operation)								
Motor		Stimulation site	Recording site	Latency (msec)	Amplitude (mV)	NCV (m/sec)		
Side	Nerve							
Right	Peroneal	Ankle	EDB		NR*			
	Peroneal	Fibular head	TA		NR*			
	Tibial	Ankle	AH	4.5	16.5	46		
Sensory		Stimulation site	Recording site	Latency (msec)		Amplitude (µV)	Distance (cm)	
Side	Nerve			Onset	Peak			
Right	SPN	Calf	Ankle	2.7	3.5	10.6	14	
	DPN	Calf	1st web space			NR*	14	
	MCPN	Popliteal fossa	Fibular neck	1.0	1.6	21.5	6	
	Sural	Calf	Ankle	2.5	3.7	13.1	14	
Follow-up study: 6-months after the surgery								
Motor		Stimulation site	Recording site	Latency (msec)	Amplitude (mV)	NCV (m/sec)		
Side	Nerve							
Right	Peroneal	Ankle	EDB		NR*			
	Peroneal	Fibular head	TA	4.0	3.0*	35		
		Popliteal fossa			6.0	2.9*		
Needle Electromyography								
Side	Muscle	Insertional Activity	Spontaneous Activity	Motor Unit Action Potentials				RP
				Normal	Polyphasia	Amplitude	Duration	
Pre-Operation								
Right	Tibialis anterior	I/A	F&P(++)					No MUAP
	Ext. digitorum longus	I/A	F&P(++)					No MUAP
	Ext. hallucis longus	I/A	F&P(++)					No MUAP
	Ext. digitorum brevis	I/A	F&P(++)					No MUAP
	Peroneus longus	N	-	N				F
	Tensor fascia lata	N	-	N				F
Follow-up study: 3-months after the surgery								
Right	Tibialis anterior	I/A	F&P(++~+++)					No MUAP
	Ext. digitorum longus	I/A	F&P(++~+++)		P			2-3 MUAPs
	Ext. hallucis longus	I/A	F&P(+++)					No MUAP
	Ext. digitorum brevis	I/A	F&P(+++)					No MUAP
	Biceps femoris (short)	N	-	N				F
	Tensor fascia lata	N	-	N				F
Follow-up study: 6-months after the surgery								
Right	Tibialis anterior	I/A	F&P(+++)		Multiphasic P			4 MUAPs
	Ext. digitorum longus	I/A	F&P(+++)		Multiphasic P	Large		4-5 MUAPs
	Ext. hallucis longus	I/A	F&P(++)			Small		1-2 MUAPs
	Ext. digitorum brevis	I/A	F&P(+++)					No MUAP

*NR: no response, EDB: extensor digitorum brevis, TA: tibialis anterior, AH: abductor hallucis, SPN: superficial peroneal nerve, DPN: deep peroneal nerve, MCPN: mixed common peroneal nerve
 Ext: extensor, N: normal, I/A: increased insertional activities, F&P: fibrillation potentials & positive sharp wave, P: polys, RP: recruitment pattern, F: full, MUAP: motor unit action potentials

shown in the tibialis anterior, extensor digitorum longus, extensor hallucis longus but not at the extensor digitorum brevis in the needle EMG (Table 1). Clinically, the MRC scale of right ankle dorsiflexion was improved to grade 3.

Case 2

A 62-year old female patient presented with a 6-week history of left foot drop. The symptom started after the abrupt pain in the left upper lateral calf region, with no clear history of a traumatic event. She had no history of low back pain nor the radiation of pain into her hip or thigh. The MRC scale for left ankle dorsiflexion and great toe extension were grade 0 and ankle eversion was grade 1. All the other muscles of left lower limb were normal. Hypoesthesia on the left foot dorsum was observed but the sural and saphenous nerve distributions were intact. There was no palpable mass lesion but the Tinel's sign over the left fibular head was positive. The SLR test was negative bilaterally.

In the NCS (Table 2), although the peroneal motor response with tibialis anterior recording was evoked with low amplitude, the left peroneal CMAP with extensor digitorum brevis recording and mixed common peroneal and superficial peroneal SNAPs were unobtainable. In needle EMG (Table 2), F&P were noted in all the CPN innervated muscles with predominant involvement of deep peroneal portion, such as tibialis anterior and extensor digitorum brevis. Sonography and MRI demonstrated lobulating cystic lesion along with the CPN around the fibular head with increased signal intensity of adjacent CPN (Fig. 1-C and D). EDX combined with radiographic findings suggested left incomplete but severe common peroneal neuropathy with predominant involvement of deep peroneal portion. Excision of the IG and resection of the articular branch of CPN was performed. And the surgery confirmed the articular branch to the proximal tibiofibular joint and the stage III peroneal IG, which extended proximally to the above fibular head level (Fig 3) [2]. In addition, IG was found to have been ruptured, forming the adhesion around the nerve to tibialis anterior, DPN and CPN with the ruptured cystic material. A follow-up study at 4-month after the surgery showed clinical and electrophysiological improvement. There was no significant interval change in NCS. But in needle EMG, recruitment patterns were improved in the tibialis anterior and reinnervation has been taking place down to the left extensor hallucis longus (Table 2). In the follow-up study at 1-year after the surgery, previously unobtainable peroneal motor response was evoked in the extensor digitorum brevis with prolonged latency, low amplitude, and decreased conduction velocity but the superficial peroneal and mixed peroneal sensory responses were unobtainable. In needle EMG, reinnervation evidence to the ex-

tensor digitorum brevis was observed (Table 2). Two years after the surgery, she was almost fully recovered with the left ankle dorsiflexion and great toe extensor muscle strength with the MRC scale of grade 5 and only mild paresthesia remained on the left foot dorsum.

Discussion

Ganglion cysts are cystic lesions that originate from tendon sheath or joint capsule. IGs are fluid-filled formations within the epineural sheath of peripheral nerves and can cause nerve compression as in our cases [3,4]. Although the mechanism is not fully understood, the most accepted hypothesis on formation of the peroneal IG is the 'articular theory' by Spinner et al. [2]. CPN has been reported to branch out at least three articular branches during its course and one of which provides sensory information from the proximal tibiofibular joint [5]. According to the 'articular theory', the IG is formed by one-way communication between the proximal tibiofibular joint and the articular branch of CPN. Due to predisposing conditions such as a traumatic event or abnormal joint pathology, the capsular defect at the proximal tibiofibular joint might have been formed and the articular branch of CPN becomes a conduit. Consequently, the cystic fluid enters the epineurium of the articular branch of CPN and extends towards the less resistant CPN and its branches as pressure is applied. And it has been reported that IG can even extend proximally to the sciatic nerve [2]. In both of our cases, the articular branch of CPN was observed in the radiographic imaging studies and confirmed intraoperatively. Resection of the ar-

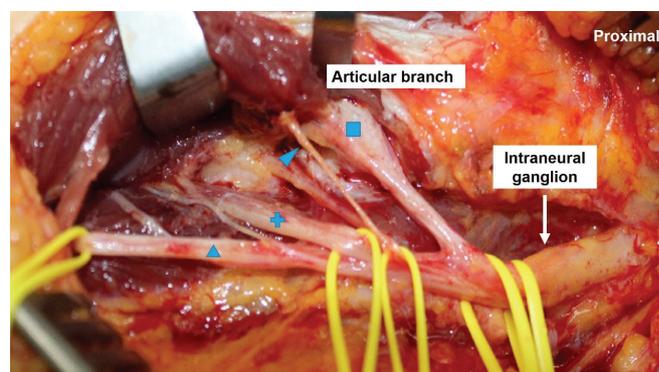


Fig. 3. Intraoperative clinical photographs of case 2, showing the left common peroneal nerve trifurcating into superficial peroneal nerve (triangle), deep peroneal nerve (cross), enlarged and fibrosed articular branch (square), branches to the tibialis anterior muscle (arrowhead) and the intraneural ganglion invading the common peroneal nerve (arrow), intraneural ganglion cyst stage III.

Table 2. Nerve Conduction and Needle Electromyography Data (Case 2)

Nerve conduction (Pre-operation)								
Motor Side	Nerve	Stimulation site	Recording site	Latency (msec)	Amplitude (mV)	NCV (m/sec)		
Left	Peroneal	Ankle	EDB		NR*			
	Peroneal	Fibular head	TA	3.6	0.68*	50.0		
		Popliteal fossa	TA	5.0	0.46*			
	Tibial	Ankle	AH	3.5	7.8	49.2		
Sensory								
Side	Nerve	Stimulation site	Recording site	Latency (msec)		Amplitude (uV)	Distance (cm)	
Left	SPN	Calf	Ankle	Onset	Peak	NR*	14	
	MCPN	Popliteal fossa	Fibular neck			NR*	6	
	Sural	Calf	Ankle	2.8	3.5	11.3	14	
Follow-up study: 1-year after the surgery								
Motor Side	Nerve	Stimulation site	Recording site	Latency (msec)	Amplitude (mV)	NCV (m/sec)		
Left	Peroneal	Ankle	EDB	6.3*	0.2*	20.0*		
	Peroneal	Fibular head	TA	2.9	4.3	56.0		
		Popliteal fossa	TA	3.8	4.0			
Needle Electromyography								
Side	Muscle	Insertional Activity	Spontaneous Activity	Motor Unit Action Potentials				RP
				Normal	Polyphasia	Amplitude	Duration	
Pre-Operation								
Left	Tibialis anterior	I/A	F&P(++~+++)		P			1-2 MUAP
	Ext. digitorum brevis	I/A	F&P(++)					No MUAP
	Peroneus longus	I/A	F&P(++~+++)		P			R
	Biceps femoris (short)	N	-	N				F
	Tensor fascia lata	N	-	N				F
	Flexor digitorum longus	N	-	N				F
Follow-up study: 4-months after the surgery								
Left	Tibialis anterior	N	F&P(++)		P	Large		R
	Ext. hallucis longus	I/A	F&P(+)		P			DIS
	Ext. digitorum brevis	I/A	F&P(+)					No MUAP
	Peroneus longus	I/A	F&P(++)		P	Large		R
Follow-up study: 1-year after the surgery								
Left	Tibialis anterior	N	-		P		Long	R
	Ext. digitorum longus	I/A	P(+)		P	Large	Long	R
	Ext. digitorum brevis	I/A	F&P(+)		P			DIS
	Peroneus longus	I/A	P(+)		P		Long	R

*NR: no response, EDB: extensor digitorum brevis, TA: tibialis anterior, AH: abductor hallucis, SPN: superficial peroneal nerve, MCPN: mixed common peroneal nerve

Ext: extensor, N: normal, I/A: increased insertional activities, F&P: fibrillation potentials & positive sharp wave, P: polys, RP: recruitment pattern, F: full, R: reduced, DIS: discrete, MUAP: motor unit action potentials

ticular branch stalk to the proximal tibiofibular joint was performed and no sign of recurrence was observed during the follow up period.

In both cases, patients experienced pain in the upper lateral calf region before the foot drop symptom started. Young et al. reported that the patients with common peroneal neuropathy due to the IG were significantly associated with pain at the lateral knee in comparison with the patients with no evidence of IG on MRI [6]. Spinner et al. described that this poorly localized lateral knee pain can be caused by pathologic conditions of the proximal tibiofibular joint [2]. Although it is a rare disease entity, the history of upper lateral calf pain may be the clinical feature that requires further evaluation such as sonography or MRI, if there is no clear proximate cause for common peroneal neuropathy.

The CPN derives from the dorsal branches of L4, L5, S1 and S2 and it descends as a main division of the sciatic nerve. It courses obliquely along the lateral side of the popliteal fossa and winds around the fibular head where it is vulnerable [1]. When exiting the fibular tunnel, the CPN is generally known to bifurcate into the DPN and SPN. However, some studies report that the CPN bifurcation be trifurcation, by including the articular branch. The idea of trifurcation is based on the cadaveric study on branching patterns of CPN. In the study, the articular branch branched out from DPN just within 3mm distal to the bifurcation point in all the limbs examined [2].

At the fibular head level, the intraneural topography of CPN is that the fascicles destined for DPN has tendency to lie more medial, whereas the fascicles destined for the SPN are more lateral. And the fascicles to the articular branch is situated medially, close to the DPN and it sometimes merges with the fascicles to the tibialis anterior and even innervates the tibialis anterior alone in some cases [2,7]. Due to this proximity, predominant involvement of DPN innervated muscles such as tibialis anterior muscle has been reported on previous cases of peroneal IG [3,8]. And this also applies to our cases and supports the previous observations that fascicles of the CPN comprising the DPN may be more vulnerable than those of the SPN. In case 1, IG extended proximally up to 5 centimeters of the CPN portion (Stage III), but only the DPN innervated muscles showed denervation potentials in needle EMG and the mixed and superficial peroneal sensory responses were within the normal range in NCS. Predominant involvement of the fascicles to DPN is also shown in the case 2. However, the ankle eversion was weak and involvement of the SPN was also observed in the NCS and needle EMG. The surgical findings with diffuse adhesion of the CPN and SPN due to the ruptured IG and the mixed common peroneal SNAP abnormality which indicate the lesion extending

proximally to the fibular head might explain the involvement of the laterally located fascicles of SPN in the CPN.

As mentioned ahead, CPN originates from the sciatic nerve with the root values of L4, L5, S1 and S2 [1]. As a result, foot drop may result from the proximal lesions such as sciatic neuropathy, lumbosacral plexopathy and radiculopathy. In a retrospective series of 217 patients presented with foot drop, common peroneal nerve lesions (30.6%), L5-radiculopathies (19.7%) were the two most common subgroups among the peripheral neurogenic origin group [9]. Therefore, to accurately distinguish the causes of foot drop is often difficult. Both of our patients had no history of low back pain nor the radiating pain and SLR was negative in the initial physical examination. NCS of the tibial motor and sural sensory responses were within the normal range and the peroneal division of the sciatic nerve innervated muscle (short head of biceps femoris muscle) and non-peroneal L5-innervated muscles (tensor fascia lata and flexor digitorum longus) showed normal EMG potentials.

A very few cases of CPN due to IG have been reported with detailed neurophysiology. This case report emphasizes correct diagnosis based on electrophysiologic, radiographic and operative findings with the follow up studies. Although the radiographic and surgical findings were compatible with the lesion extending into the CPN, the precise analysis of the electrophysiologic findings could differentiate the fascicular involvement within the CPN in two cases, deep peroneal portions only versus predominant involvement of DPN of CPN.

Intraneural ganglion cyst (IG) is not an entity that we encounter often. However, an early diagnosis of the IG and to rule out the other general causes of common peroneal neuropathy is important, because early surgical treatment of the IG is crucial to the prevention of the neurological deterioration with improved outcomes. In our cases, no recurrence was observed and improvement in the clinical symptom and the electrophysiologic findings were obtained in the follow-up studies after the surgery.

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