전골간신경증후군과 유사한 증상을 보인 근위정중신경병증 2예

배초롱, 이아리, 권희규

고려대학교 의과대학 고려대학교안암병원 재활의학과교실

Proximal Median Neuropathy Mimicking Anterior Interosseous Nerve Syndrome: A Report of Two Cases

Cho Rong Bae, Ahry Lee, Hee-Kyu Kwon

Department of Physical Medicine & Rehabilitation, Korea University Anam Hospital,
Korea University College of Medicine, Seoul, Korea

Anterior interosseous nerve (AIN) syndrome (AINS) is a clinical condition that arises due to injury of the AIN and presents characteristic weakness of the flexor pollicis longus, radial half of the flexor digitorum profundus, and/or the pronator quadratus. An identical clinical manifestation may also result from fascicular lesions more proximally within the median nerve or brachial plexus (pseudo-AINS). In case of pseudo-AINS due to focal proximal median lesion, however, it is hard to differentiate from the lesion to AIN itself by electrodiagnostic examination. This is a report of two cases of proximal median neuropathy following an axillary brachial plexus block, which clinically mimics AINS. This report shows the importance of knowledge about the fascicular somatotopy in AINS diagnosis.

Key Words: anterior interosseous nerve syndrome, brachial plexus block, electrodiagnosis

Introduction

Brachial plexus block (BPB) is a commonly used technique for providing regional anesthesia to the upper extremities. There are several advantages of BPB over general anesthesia, such as superior pain management and fewer systemic effects.¹ The axillary

approach provides effective anesthesia distal to the elbow; therefore, it is preferred for forearm and hand surgery.¹ Complications with BPB may also occur, including nerve damage due to direct needle trauma, local anesthetic toxicity, or hematoma. The reported incidence of nerve injury following axillary block is variable (0.2% to 19%), and the severity of symptoms may also range from minor transient pain to permanent sensory or motor deficit.² In a retrospective study involving 13 cases of infraclavicular brachial plexus injury following axillary regional block, the median nerve was most often affected.³

The anterior interosseous nerve (AIN) is a motor

Received January 17, 2018

Revised March 29, 2018

Accepted April 13, 2018

Corresponding Author: Hee-Kyu Kwon

Department of Physical Medicine & Rehabilitation, Korea University Anam Hospital, Korea University College of Medicine, 73 Incheon-ro, Seongbuk-gu, Seoul 02841, Korea

Tel: 82-2-920-6471, Fax: 82-2-929-9951, E-mail: hkkwon@korea.ac.kr

Table 1. Nerve Conduction Study Findings

Motor nerve conduction						
Side	Nerve	Stimulation site	Recording site	Latency (msec)	Amplitude (mV)	NCV (m/s)
Left	Median	Wrist	APB	3.4	13.2	55.2
		Elbow	APB	7.2	13.1	–
	Ulnar	Wrist	ADM	2.9	12.0	58.0
		Below elbow	ADM	6.0	11.5	55.5
		Above elbow	ADM	7.8	11.2	–
Sensory nerve conduction						
Side	Nerve	Stimulation site	Recording site	Onset/peak latency (msec)	Amplitude (uV)	Distance (cm)
Left	Median	Wrist	III digit	2.8/3.6	29.3	14
		Palm	III digit	1.1/1.8	34.9	7
	Ulnar	Wrist	V digit	2.6/3.4	24.8	14

NCV: nerve conduction velocity, APB: abductor pollicis brevis, ADM: abductor digiti minimi, –: not applicable

branch of the median nerve that leaves the median nerve trunk at the forearm level. It innervates the flexor pollicis longus (FPL), the pronator quadratus (PQ), and the radial half of the flexor digitorum profundus (FDP). Paralysis of the AIN, therefore, presents with characteristic weakness of the pincer movement of the thumb and/or index finger, middle finger, and forearm pronation, which is referred to as AIN syndrome (AINS).⁴

This report describes two cases of proximal median neuropathy following an axillary BPB, which clinically mimics AINS.

Case Report

1) Case 1

A 61-year-old man was referred for electrodiagnostic examination (EDX) to the Department of Physical Medicine and Rehabilitation on March 23, 2016, with complaints of flexion difficulty of the left thumb for 6 months. He was injured while working on September 30, 2015, and was diagnosed with a rupture of the extensor tendon with a coexisting fracture on his left index finger. He underwent a pinning and tenorrhaphy operation under axillary BPB on the same day at a private orthopedic clinic. Weakness of the left thumb developed immediately after the surgery and showed no improvement over the ensuing 6 months. Hand

magnetic resonance imaging (MRI) revealed preserved flexor tendons of the left fingers. He was referred to the authors for further evaluation.

The Medical Research Council scale for his left thumb interphalangeal (IP) joint flexion and index finger distal IP joint flexion was grade 0. No sensory abnormalities were reported, and all deep tendon reflexes were normoactive.

An EDX was performed on March 23, 2016. On the nerve conduction study, compound muscle action potential (CMAP) and sensory nerve action potential of the left median and ulnar nerves were within normal limits (Table 1). On needle electromyography, abnormal spontaneous activities and no motor unit action potential were noted in the left FPL and PQ muscles. Normal motor unit potentials were noted in all the other muscles examined (Table 2). EDX findings were compatible with a left anterior interosseous neuropathy. However, ultrasonography of the AIN revealed no abnormal findings. Clinical history, EDX, and sonography resulted in a final diagnosis of a left median neuropathy, probably at the axillary level with predominant involvement of fascicles to AIN. Nine months later, in December 2016, he reported some improvement in his left thumb flexion strength to grade 2.

Table 2. Needle Electromyography

Side	Muscle	Insertional activity	Spontaneous activity	Motor unit action potentials	Recruitment pattern
Left	Deltoid	Normal	-	Normal	Normal
	Brachioradialis	Normal	-	Normal	Normal
	Pronator teres	Normal	-	Normal	Normal
	Triceps brachii	Normal	-	Normal	Normal
	Flexor carpi ulnaris	Normal	-	Normal	Normal
	First dorsal interosseous	Normal	-	Normal	Normal
	Flexor pollicis longus	-	F&P(+++)	-	Absent
	Pronator quadratus	-	F&P(++~+++)	-	Absent
	C5/6, C6/7 PVMs	Normal	-		

F: fibrillation potentials, P: positive sharp waves, PVM: paravertebral muscle, -: not applicable

2) Case 2

A 54-year-old woman presented to the Department of Physical Medicine and Rehabilitation on October 29, 2014, with a 5-month history of weakness in right thumb flexion. She underwent an A1 pulley release operation under axillary BPB to release right trigger thumb on May 24, 2014, at a private orthopedic clinic. Weakness in the right thumb developed immediately after the surgery and persisted for several months. She underwent re-operation; however, the integrity and gliding of the right FPL tendon were intact. She was referred for EDX to rule out brachial plexus injury or AINS.

Muscle strength for her right thumb IP joint flexion was grade 0. She was unable to perform a proper "OK" sign when asked to form an 'O' with her right thumb and index finger. There was no sensory loss.

An EDX was performed on October 29, 2014. Motor nerve conduction study of the right median nerve with abductor pollicis brevis muscle recording was within normal limits. The right median CMAP with FPL muscle recording (using needle electrode), however, revealed prolonged latency and low amplitude, compared to the left side (normal side-to-side difference: CMAP amplitude < 0.7 mV; CMAP onset latency < 5.1 %) (Table 3).⁵ On needle electromyography, abnormal spontaneous activities and no motor unit action potential were noted in the right FPL and PQ muscles. Normal motor unit potentials were noted in all the other muscles examined (Table 4). Findings on EDX

were compatible with a right anterior interosseous neuropathy. However, considering full clinical history, she was finally diagnosed with a right median neuropathy, probably at the axillary level with predominant involvement of fascicles to AIN. Her symptom(s) gradually improved, exhibiting recovery of right thumb flexion strength to grade 4 at the 3-month follow-up.

Discussion

Focal injury of the AIN is an uncommon peripheral neuropathy. Because the FPL and/or FDP to the index finger are involved, patients are not able to flex the IP joint of the thumb and the distal IP joint of the index finger, and complain of difficulty in attempting to pick up small objects using the first two digits. The PQ is also paralyzed; however, it is difficult for patients to notice the weakness and on manual muscle testing because the PQ muscle could be overwhelmed by pronator teres muscle action.⁶ Because the AIN lacks cutaneous sensory fibers, patients with pure AIN palsy will not complain of a sensory defect.⁷ The signs and symptoms have various etiologies, including inflammation, compression and trauma of the AIN itself.⁷

An identical clinical manifestation, however, may also result from a lesion in the more proximal part of the median nerve or brachial plexus, which were noted in our cases. Causes of these pseudo-anterior interosseous neuropathies include supracondylar fractures of the

Table 3. Nerve Conduction Study Findings

Motor nerve conduction						
Side	Nerve	Stimulation site	Recording site	Latency (msec)	Amplitude (mV)	NCV (m/s)
Right	Median	Wrist	APB	2.4	6.9	51.3
		Elbow	APB	6.3	6.8	–
		Elbow	FPL (needle)	3.8*	3.7*	–
	Ulnar	Wrist	ADM	2.5	8.3	64.5
		Below elbow	ADM	5.6	8.3	66.6
		Above elbow	ADM	7.1	8.1	–
Left	Median	Wrist	APB	2.5	6.9	50.0
		Elbow	APB	6.3	6.7	–
		Elbow	FPL (needle)	2.5	6.6	–
Sensory nerve conduction						
Side	Nerve	Stimulation site	Recording site	Onset/peak latency (msec)	Amplitude (uV)	Distance (cm)
Right	Median	Wrist	III digit	3.1/3.8	21.0	14
		Palm	III digit	1.2/1.8	24.6	7
		Wrist	I digit	2.0/2.8	22.0	8
	Ulnar	Wrist	V digit	2.4/3.1	22.5	14
	Superficial radial	Wrist	Snuff box	1.8/2.4	26.1	10
	LABCN	Elbow	Forearm	2.2/2.7	15.5	12

*Abnormal value. NCV: nerve conduction velocity, APB: abductor pollicis brevis, ADM: abductor digiti minimi, FPL: flexor pollicis longus, LABCN: lateral antebrachial cutaneous nerve, –: not applicable

Table 4. Needle Electromyography

Side	Muscle	Insertional activity	Spontaneous activity	Motor unit action potentials	Interference pattern
Right	Brachioradialis	Normal	–	Normal	Normal
	Pronator teres	Normal	–	Normal	Normal
	Triceps brachii	Normal	–	Normal	Normal
	Flexor carpi radialis	Normal	–	Normal	Normal
	Flexor carpi ulnaris	Normal	–	Normal	Normal
	First dorsal interosseous	Normal	–	Normal	Normal
	Flexor pollicis longus	–	F&P(++)	–	Absent
	Pronator quadratus	–	F&P(++)	–	Absent
	C5/6, C6/7 PVMs	Normal	–		

F: fibrillation potentials, P: positive sharp waves, PVM: paravertebral muscle, –: not applicable

humerus or of the proximal radius, catheterization of the brachial artery or venipuncture at the antecubital fossa, and neuralgic amyotrophy (acute brachial neuropathy). A plausible explanation is that the injury is restricted to certain fascicles contributing to the AIN more distally.⁸

Within a peripheral nerve, individual nerve fibers are clustered in fascicles. Internal longitudinal organization (whether cable or plexiform structure) and somatotopic organization of the fascicles (functional clustering of nerve fibers) within nerves have long been studied

because of clinical importance. Various studies have provided evidence for somatotopic clustering of nerve fibers. In distal portions of nerves, fascicles are mainly organized in a cable structure, and each fascicle contains motor or sensory fibers innervating specific skin areas or muscles. More proximally, nerve fibers intermingle and form a plexiform structure. Despite many variations in the fascicle array, however, somatotopic clustering of nerve fibers is largely maintained within most of the length of the nerve. As a result of this somatotopic arrangement, partial focal nerve lesions can

produce similar clinical manifestations that are not consistent with the classic rules of localization.⁸

The internal neural topography of fascicles forming the AIN in the median nerve were mapped by Jabaley et al.⁹ They examined the median nerves of fresh cadavers by serial section and microscopic interfascicular dissection. They found that fascicles of AIN are observed on the ulnar-dorsal surface in upper forearm level and travel proximally, retaining the same somatotopic arrangement on the outer perimeter. Later, Pham et al.⁴ used high-resolution magnetic resonance neurography to investigate fascicular lesions in spontaneous AINS patients and reported precise somatotopic organization of fascicles in the upper arm median nerve trunks. Only a partial area of dorsal and lateral aspect of the nerve cross-section were involved, and this lesion area accorded with the motor fascicles that form the AIN further distally.⁴

At the level of axillary block, the cords of the brachial plexus have already divided into major terminal nerves, and these are distributed around the axillary artery. Therefore, to achieve sufficient anesthesia with axillary BPB, multiple needle position adjustments and injections are needed. Mechanical trauma from needle and injection agents can occur during the process.^{1,2} Other potential mechanisms of injury, such as direct injury, compression by hematoma, and use of tourniquet, cannot be fully excluded. However, because both of our cases have undergone finger operations, direct surgical injury is unlikely to cause direct AIN injury, and was confirmed by ultrasonography in one case. In case of hematoma formation due to injury of vessel, nerves are compressed gradually and this often leads to a delayed onset of symptoms after the procedure. Furthermore, tourniquet injury is known to usually affect the radial nerve with/without the median and ulnar nerves.^{2,3} Therefore, considering that symptoms occurred immediately after surgery and that injuries were isolated to median nerve, the median neuropathy mimicking anterior interosseous neuropathy confirmed by EDX were considered as a

problem with the proximal median nerve, probably at axillary level, and not a problem with the AIN itself. Brachial plexus injury is usually confirmed by EDX, but EDX is sometimes nonspecific or non-localizing, especially in cases of mild abnormalities. Therefore, although not performed in our cases, imaging modalities such as MRI and ultrasound can be useful complementary tools for assessing pathologies and localizing lesions of brachial plexus injury.

Recognition of fascicular somatotopy has clinical importance, not only in terms of the clinical manifestations, but also in the surgical repair of nerve lesions. Treatments for nerve lesions depend on various factors, such as injury site, degree of damage, and duration between injury and repair. Conservative management is recommended if there is no axonal degeneration. However, surgical treatment should be considered if there is severe axonal degeneration and no improvement with conservative treatment for a few months.^{2,10} Both patients in our cases exhibited spontaneous recovery to some extent after few months of onset, although neurological deficit persisted in case 1. In case of AINS due to focal proximal median lesion, it is difficult to differentiate from lesion to AIN itself by EDX. Therefore, without the concept of fascicle somatotopy, lesions can be mislocalized, which is particularly important when determining treatment.

References

1. Mian A, Chaudhry I, Huang R, Rizk E, Tubbs RS, Loukas M: Brachial plexus anesthesia: A review of the relevant anatomy, complications, and anatomical variations. *Clin Anat* 2014; 27: 210-221
2. Kim HJ, Park SH, Shin HY, Choi YS: Brachial plexus injury as a complication after nerve block or vessel puncture. *Korean J Pain* 2014; 27: 210-218
3. Tsao BE, Wilbourn AJ: Infraclavicular brachial plexus injury following axillary regional block. *Muscle Nerve* 2004; 30: 44-48
4. Pham M, Baumer P, Meinck HM, Schiefer J, Weiler M, Bendzus M, et al: Anterior interosseous nerve syndrome: fascicular motor lesions of median nerve trunk. *Neurology* 2014;

- 82: 598-606
5. Vucic S, Yiannikas C: Anterior interosseous nerve conduction study: normative data. *Muscle Nerve* 2007; 35: 119-121
 6. Nagano A: Spontaneous anterior interosseous nerve palsy. *J Bone Joint Surg Br* 2003; 85: 313-318
 7. Chin DHCL, Meals RA: Anterior interosseous nerve syndrome. *J Am Soc Surg Hand* 2001; 1: 249-257
 8. Stewart JD: Peripheral nerve fascicles: anatomy and clinical relevance. *Muscle Nerve* 2003; 28: 525-541
 9. Jabaley ME, Wallace WH, Heckler FR: Internal topography of major nerves of the forearm and hand: a current view. *J Hand Surg Am* 1980; 5: 1-18
 10. Spinner RJ, Kline DG: Surgery for peripheral nerve and brachial plexus injuries or other nerve lesions. *Muscle Nerve* 2000; 23: 680-695