Case Report / Olgu Sunumu

Alcoholic Neuropathy with Superimposed Focal Entrapment Neuropathies

Fokal Tuzaklanma Nöropatileri ile Birlikte Olan Alkolik Nöropati

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Abstract

We reported here a case of alcoholic polyneuropathy with superimposed focal entrapment neuropathies. A 55-year-old male patient was admitted to the electrophysiology unit with the diagnosis of left ulnar neuropathy. He had a previous history of chronic alcoholism. Nerve conduction studies revealed bilateral carpal tunnel syndrome, bilateral ulnar neuropathy at the elbow, left peroneal neuropathy at the fibular head, prolonged distal motor latencies and moderately reduced motor conduction velocities in multiple motor nerves, slowed conduction velocities in multiple sensory nerves and multiple prolonged F-waves. Gene analysis did not reveal deletion on chromosome 17p11.2-12. We wanted to share our findings with other clinicians to get their opinion regarding possible diagnosis of the case.

Keywords
Peripheral neuropathy, HNPP, chronic alcoholism, electrodiagnosis

Öz

Bu yazıda fokal tuzaklanma nöropatileri ile birlikte olan alkolik nöropatili bir hasta sunulmuştur. Elli beş yaşında erkek hasta sol ulnar nöropati tanısı ile kliniğimiz elektrofizyoloji ünitesine kabul edildi. Hasta anamnezde önceye ait kronik alkol alışkanlığı öyküsü veriyordu. Sinir iletim çalışmaları hastada bilateral karpal tünel sendromu, dirsekte bilateral ulnar nöropati, solda fibula başı bölgesinde peroneal nöropati, distal motor latanslarda uzama ve orta derecede azalmış motor iletim hızları, çok sayıda duyusal sinir ileti hızlarında yavaşlama ve çok sayıda uzamış F yanıtı latansı varlığını ortaya koydu. Genetik analizlerde 17p11.2-12 kromozomda delesyon saptanmadı. Bulgularımızı hastanın muhtemel tanısı hakkındaki görüşlerini almak için diğer klinisyenlerle paylaşmak istedik.

Anahtar Kelimeler
Periferik nöropati, HNPP, kronik alkolizm, elektrodiagnoz

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Introduction

Peripheral neuropathy (PN) is a common disorder associated with significant morbidity. Differential diagnosis of peripheral neuropathies may sometimes be hard and confusing. We report here a case of alcoholic polineuropathy mimicking hereditary neuropathy with liability to pressure palsies (HNPP). Alcoholic neuropathy with superimposed focal entrapment neuropathies has not been reported previously. The present case report may therefore be of interest.

Case Report

A 55-year-old male patient was admitted to the electrophysiology unit with the diagnosis of left ulnar neuropathy. He complained of muscle weakness and hypoesthesia in his left hand lasting about 6 months. He had a previous history of alcohol abuse (35-70 cL 45% ethyl alcohol per day). He denied previous history of trauma, drug abuse or any predisposing metabolic disease. He also denied family history of neuropathies. On physical examination, there was atrophy in the hypothenar and dorsal interosseous muscles of both hands. Manuel muscle testing demonstrated diminished muscle contraction (4/5) in his all intrinsic hand muscles as well as diminished muscle contraction of distal musculature of both lower extremities. Sensation was diminished in the right and left C5-8 and L1-S1 dermatomes. Tendon reflexes were absent bilaterally.

Nerve conduction studies (NCS) and electromyography (EMG) were conducted using standard techniques (1), by a Neuropack M1 electromyograph (Nihon Kohden, Tokyo, Japan), with skin temperature maintained above 30 °C. Antidromic sensory nerve action potentials (SNAP) for median and ulnar sensory nerves were recorded with standard ring electrodes. Compound muscle action potentials (CMAP) and sural nerve SNAPs were recorded using disc electrodes. NCS revealed increased distal motor latencies of the right median, right and left ulnar and right peroneal nerves. There was a reduction in motor nerve conduction velocities in the right peroneal and tibial nerves. There was a reduction in CMAP amplitude of the right peroneal nerve. Focal slowing of nerve conduction were recorded at the elbow segment in the right and left ulnar and median nerves. CMAP was not elicited at the fibular head segment in the right peroneal nerve. Mildly prolonged F-wave latencies were recorded from the right and left ulnar, right tibial, and left median nerves. F-wave was not elicited from the right peroneal nerve. There was a reduction in sensory nerve conduction velocities in the right and left median and right ulnar nerves without a reduction in SNAP amplitudes. Ulnar SNAP was absent on the left side and sural SNAPs were absent bilaterally (Table 1). Because of the patient's intolerance, motor nerve conduction was not studied on the left lower limb. Needle EMG revealed increased numbers of polyphasic motor unit potentials and discrete activity in the recruitment pattern of the right and left abductor pollicis brevis, right abductor digiti minimi (ADM), deltoid and tibialis anterior tendons and left vastus lateralis muscles. Whole electrophysiological data revealed generalized, primarily demyelinating mixed sensory and motor neuropathy with superimposed focal entrapment neuropathies. Gene analysis was suggested to the patient for correct diagnosis of his disorder. Gene analysis did not reveal deletion on chromosome 17p11.2-12. The patient underwent a rehabilitation program including muscle strengthening, flexibility and range of motion exercises. Wrist splints and activity modifications including avoiding repetitive hand movements and minimizing time leaning on elbows, crossing legs, and kneeling were recommended to the patient. He was consulted with a psychiatrist for his alcohol abuse and was prescribed 125 mg/day disulfiram. Vitamin B and magnesium supplementations were also prescribed. Muscle strength was improved after a 6-week rehabilitation program, but he refused disulfiram treatment and his alcohol abuse was not treated. He also refused follow-up examinations. Informed consent was obtained from the patient.

Discussion

Alcoholic neuropathy is one of the most common forms of PN. It is a mixed sensory and motor disorder, involving predominantly the distal segments and the legs. In alcoholic neuropathy, the nerve conduction abnormalities are typical of axonal neuropathy. The amplitude of the CMAP and SNAP is markedly reduced, whereas the motor and sensory NCSs are mild slow, and the H-reflex and F-wave latencies are mildly prolonged (1-3).

HNPP is one of the major forms of hereditary motor sensory neuropathies. HNPP is associated with a deletion on chromosome 17p11.2-12. Electrophysiologically, HNPP is characterized by a generalized demyelinating neuropathy with superimposed focal entrapment neuropathies. In HNPP patients, NCSs reveal a diffuse increase in distal motor latencies, moderately reduced motor conduction velocities as well as focal slowing of nerve conduction at common sites of entrapment (4-6).

Electrophysiological findings in our patient revealed generalized, primarily demyelinating, mixed

Table 1. Motor and sensory nerve conduction studies

Motor nerve conduction study

Nerve-record	Stimulation site	Onset latency (ms)	Norm onset latency	Amplitude (mV)	Norm amplitude	Segment	Distance (mm)	Velocity (m/s)	Norm velocity
R Ulnar- ADM									
	Wrist	3.42	<3.3	9.68	>7.0				
	Below elbow	7.98		9.67		Wrist-below elbow	240	52.6	>49.9
	Above elbow	11.80		8.65		Below elbow -Above elbow	130	34	>39.6
F resp		34.65	<32.0						
L Ulnar-ADM									
	Wrist	3.44	<3.3	10.96	>7.0				
	Below elbow	7.10		8.98		Wrist-below elbow	210	57.4	>49.9
	Above elbow	5.79		3.67		Below elbow-Above elbow	135	23.3	>39.6
F resp		35.12	<32.0						
R Median-APB									
	Wrist	4.36	<3.8	11.22	>4.3				
	Elbow	8.80		10.89		Wrist-elbow	255	57.4	>49.7
	Above elbow	11.50		9.95		Elbow-Above elbow	120	44.4	>49.9
F resp		31.21	<32.0						
L Median-APB									
	Wrist	3.76	<3.8	17.62	>4.3				
	Elbow	9.06		15.17		Wrist-elbow	265	50	>49.7
	Above elbow	11.62		14.96		Elbow-Ab.Elb	110	42.9	>49.9
F resp		33.03	<32.0						
R Peroneal-EDB									
	Ankle	6.16	<5.8	2.21	>3.6				
	Fibular head	17.25		1.04		FH-Ankle	340	30.7	>40.9
	Poplitea	NR				Pop-FH			
F resp		NR	<52.0						
R Tibial-AH									
	Ankle	4.45	<5.8	11.22	>3.6				
	Poplitea	16.40		4.21		Ankle-Pop	400	33.5	>39.6
F resp		56.34	52.0						

Table 1. Continue

Sensory nerve conduction study							
Nerve	Segment	Onset latency (ms)	Amp (μV)	Norm amp	Distance (mm)	Velocity (m/s)	Norm velocity
R Median	Wrist-2nd Digit	2.92	10.50	>10	125	42.8	>49.4
L Median	Wrist-2nd Digit	2.88	10.20	>10	120	41.7	>49.4
R Ulnar	Wrist-5th Digit	NR		>7			>37.3
L Ulnar	Wrist-5th Digit	2.46	12.10	>7	95	38.6	>37.3
R Sural	Calf-Lat Malleol	NR		>5			>33.8
L Sural	Calf-Lat Malleol	NR		>5			>33.8

NR: No response, R: Right, L: Left, ADM: Abductor digiti minimi, APB: Abductor pollicis brevis, EDB: Extensor digitorum brevis, AH: Abductor hallucis, NR: No response, Norm: Normal values of our electrophysiology unit

sensory and motor neuropathy with superimposed focal entrapment neuropathies. Although the electrophysiological data was consistent with HNPP, the patient denied family history of neuropathies and, gene analysis did not reveal deletion on chromosome 17p11.2-12. In the light of these clinical data and electrophysiological findings, we suspected the diagnosis of alcoholic neuropathy with superimposed focal entrapment neuropathies. Hong et al. (7) reported that the distribution and severity of the background electrophysiologic abnormalities were closely related to the topography of common entrapment or compression sites, which suggests the possible pathogenetic role of subclinical pressure injury at these sites in the development of the distinct background polyneuropathy in HNPP. Similarly, subclinical pressure injury at common entrapment or compression sites of nerves affected by alcoholism may be the cause of pressure palsies in our patient. This case remained as a mystery for us because the gene analyses did not support the diagnosis, but our clinical and electrophysiologic findings resembled HNPP. We wanted to share our findings with other clinicians to get their opinion regarding possible diagnosis of the case.

Ethics

Informed Consent: Consent form was filled out by all participants.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: H.G., B.N., H.R.E., Concept: H.G., B.N., Design: H.G., B.N., Data Collection or Processing: H.G., B.N., Analysis or Interpretation: H.G., B.N., Literature Search: H.G., A.K., H.R.E., Writing: H.G., B.N.

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