OLGU SUNUMU

A Case of Poststreptococcal Acute Motor and Sensory Axonal Neuropathy

Poststreptokokkal Akut Motor ve Sensoryel Kksonal Möropati Olgusu

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SUMMARY

The characteristic of the typical Guillain-Barré syndrome (GBS) is an ascending symmetrical and demyelinating polyradiculoneuropathy. Acute motor and sensory axonal neuropathy (AMSAN) is a less common variant of GBS. Here, we describe an atypical AMSAN case developed after a beta-hemolytic streptococcal infection. Also, repeated electrophysiological test findings were discussed.

Keywords: Polyneuropathy, GBS, streptococcus, atypical.

ÖZET

Tipik Guillain-Barré sendromunun (GBS) karakteristiği assendan, simetrik ve demiyelinizan poliradikülonöropati olmasıdır. Akut motor ve sensoryel aksonal nöropati (AMSAN) GBS'nin daha az görülen bir çeşididir. Burada, beta-hemolitik streptokok enfeksiyonu sonrası gelişen atipik bir AMSAN olgusunu tanımladık. Ayrıca tekrarlı elektrofizyolojik bulgularını tartıştık

Anahtar Sözcükler: Polinöropati, GBS, streptokok, atipik.

INTRODUCTION

Post-infectious acute polyneuropathies may develop after various viral or bacterial infections. Several pathogens can be responsible such as Campylobacter jejuni, Mycoplasma pneumoniae, Cytomegalovirus and Epstein Barr virus (1). Group A beta-hemolytic streptococcus (GABHS) has rarely been reported as a responsible infectious agent (2). The characteristic clinical feature of Guillain-Barre sydrome (GBS) is an ascending symmetrical demyelinating polyradiculoneuropathy. However, unusual forms such as acute motor axonal neuropathy (AMAN), acute motor and sensory axonal neuropathy (AMSAN) were recorded. Although it is symmetrical polyneuropathy, asymmetrical features of GBS have been described (1, 3).

Here, we described an atypical AMSAN case developed after a GABHS infection.

CASE REPORT

A previously healthy 38-year-old male was diagnosed as GABHS tonsillopharyngitis verified by a throat culture, and was given ampicillin 1000 mg/day. On the second day of treatment, a generalized maculopapular skin rash developed on his body and ampicillin treatment cancelled and prednisolone was started at a dose of 16 mg/day. Three days later, he admitted another neurologist for weakness of right foot. Right peroneal mononeuropathy was observed in electromyography (EMG) and prednisolone dose was increased to 64 mg/day.

Because of a weakness in his left hand had developed after two days the patient was admitted to our outpatient clinic. Routine biochemical assays, vasculitis and cancer-related biomarkers, protein and immune electrophoresis were normal. Nerve conduction studies showed prolonged distal and F latencies, asymmetrical reduction of compound motor action potential amplitudes. Sensory nerve studies showed mild prolonged latencies. Needle EMG showed widespread acute denervation potentials in muscles of upper and lower extremities (Table 1).

These findings revealed both demyelinating and axonal polyneuropathy in motor and sensory fibers with significant asymmetry in upper and lower limbs. In cerebrospinal fluid examination, the protein level was 50 mg/dl and a cell reaction was not detected. GM1 and/or GD1a antibody was found negative. Intravenous immune globulin therapy was administered at a dose of 0.4 gr/kg/ day for five days. His clinical findings were fairly normal on examination after one month. Also, the last EMG examination showed that motor latencies in both upper and lower extremities were shortened, amplitudes in upper extremities were improved after 1 month. In lower extremities, amplitude asymmetry in motor nerves was found. Sensory nerve conductions were markedly improved (Table 1).

Table 1. Electromyographic findings of patient. Both motor and sensory nerve amplitudes were reduced in first evaluation. Markedly improvement was obtained in second evaluation.

Motor conduction	Terminal latency (ms) First	CMAP (mV) First	Velocity (m/s) First	F latency (ms) First	Terminal latency (ms) Second	CMAP (mV) Second	Velocity (m/s) Second	F latency (ms) Second
R. Median Ankle Elbow	6.5 11.4	2.8 2.7	50.8	34.7	5.1 9.9	5.5 5.1	52.7	34
R. Ulnar Ankle Below elbow Above elbow	3.3 8.7 10.5	3.4 2.7 1.2	48.1 66.7	39.4	3.09 8.3 10.4	8.4 8.8 8.3	53.6 50.9	34.1
L. Median Ankle Elbow	5.2 9.4	1.9 0.5	59.5	32.7	5.1 9.9	5.1 5.2	52.1	
L Ulnar Ankle Below elbow Above elbow	4.4 7.4 9.1	1.6 1.5 1.1	80 74.1	38	3.7 8.1 9.7	3.03 3.4 2.3	61.6 62.9	41
R Tibial Ankle Poplitea	7.8 18.8	6.8 4.1	46.4	69.7	5.4 15.8	4 2.3	46.2	55.2
R Peroneal Ankle Neck of fibula Behind knee	8.2 18.3 19.8	0.6 0.6 0.5	37.4 69	60.5	6.7 16.2 18.6	0.5 0.6 0.6	41.1 42.6	
L Tibial Ankle Poplitea	9.5 21.5	4.5 3.4	41.7	69.7	6.7 17.9	2.9 2.5	42.2	57.2
L Peroneal Ankle Neck of fibula Behind knee	6.7 15.8 17.5	5.1 5.4 5.1	44 57.1	62.5	4.7 13.4 15.3	3.6 2.9 3	41.4 52.6	58.2
Sensory conductions:	Onset latency (ms)	SNAP (μV)	Velocity (m/s)		Onset latency (ms)	SNAP (μV)	Velocity (m/s)	
R Median	3.2	12.7	49.7		2.4	33	53.3	
R. Ulnar	2.6	5.8	48.5		2.3	45	47.8	
L Median	3.1	16.8	47.5		1.9	62	60.6	
L Ulnar	2.9	10.3	46.6		2.3	29	46	
R Sural	3	9.9	36.7		2.2	17	74.9	
L Sural	2.3	21.2	51.3		2.4	25	55.3	

DISCUSSION

Our patient displayed acute onset of right sided weakness on admission mimicking mononeuropathy, which is not typical for GBS. Classical electrophysiological findings of GBS are slower motor nerve conduction, conduction blocks, and excessive temporal dispersions of compound muscle action potentials (CMAP) (1,4). The AMAN and AMSAN that have poor prognosis are axonal variant of GBS. Campylobacter jejuni is associated with these variants (3). These variants may develop in two patterns. The first pattern affects distal and the other one effects proximal parts of the nerves (5). The first one affects nodal-paranodal regions of axon and it causes dysfunction of voltage gated sodium channel with probably accumulation of GM1 and / or GD1a antibody and it rapidly recovers. The other one causes severe axonal Wallerien-like dejeneration, its recovery is slow and not complete (5-9). After one month, weakness and hypoestesia fairly recover in our patient. EMG showed that demyelinating findings markedly improved and amplitudes of CMAP were increased in upper extremities but they were decreased in lower extremities. Sequential EMG examination can be used for the assessment and follow-up of

neuropathy (10, 11).

In this case, nodopathy and /or paranodopathy and axonal degeneration especially in long nerves can develop together. The rapid recovery of clinical findings made us exclude vasculitic neuropathy (12). We think that two patterns of AMSAN have developed together in this case. Electrophysiological recovery in the lower extremities may be late due to long nerves. Traverso et al. has reported a case of mononeuritis multiplex result of sequelae of

streptecoccal infection. In this case, sural nerve biopsy have been showed necrotizing vasculitis. They have mentioned that the disease course was progressive and poor (13). Asymmetry of symptoms and mononeuropathy multiplex principally suggests vasculitic etiology (12). However, this course can be rarely observed in the variants of GBS (14).

As a result of, A beta hemolytic streptococcal infection can rarely be the cause of the variants of AMAN, AMSAN or vasculitic neuropathy.

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