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Motor axonal polyneuropathy in the course of ulcerative colitis: A case report

Ülseratif kolitin seyrinde görülen nadir bir extraintestinal bulgu: Motor aksonal polinöropati

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We describe an axonal motor polyneuropathy in a patient with ulcerative colitis. Symptoms of neuropathy occurred during active colitis. Electrophysiological study showed motor axonal degeneration. After treatment with steroid added to mesalazine, the patient had a gastrointestinal recovery and neurological symptoms were improved. Axonal motor polyneuropathy is an unusual extraintestinal manifestation of ulcerative colitis, and is probably associated with an autoimmune process

Key words: Axonal degeneration, ulcerative colitis

Biz ülseratif kolitli bir hastada axonal motor polinöropati tanımladık. Nöropati semptomları kolitin alevlenmesiyle ortaya çıktı. Motor aksonal dejenerasyon elektrofizyolojik çalışmayla gösterildi. Mesalazin tedavisine eklenen steroid sonrasında hastanın barsaktaki hastalık hali düzeldi ve nörolojik semptomları kayboldu. Axonal motor polinöropati ülseratif kolitin nadir bir extraintestinal bulgusudur. Muhtemelen bu bulgu otoimmün bir reaksiyona bağlıdır.

Anahtar kelimeler: Aksonal dejenerasyon, ülseratif kolit

INTRODUCTION

Ulcerative colitis (UC) is characterized by recurring episodes of inflammation limited to the mucosal layer of the colon. The severity of the symptomatology often correlates with the anatomic extent of disease. UC has various extraintestinal manifestations (1-3). Among extraintestinal complications, neurologic involvement is relatively rare. Up to 3% of patients may have various non-iatrogenic neurologic involvements including peripheral neuropathy, myelopathy, myasthenia gravis and cerebrovascular disorders (4). There are a few reports about axonal polyneuropathy in UC (5,6). We report an adult male patient with UC who had an axonal motor polyneuropathy.

CASE REPORT

A 47-year-old man developed rectal bleeding, tenesmus, abdominal pain and weight loss in May 2004. Colonoscopy and colonic biopsy established UC. The disease involved intestine from the rectum to the splenic flexura. Since the disease was severe, we started oral prednisone (40 mg/d), and oral (2 g/d) and enema (4 g/d) mesalazine. He went into remission in July 2004, after which we continued therapy with only enema mesalazine.

The exacerbation of the disease occurred in September 2004 with bloody diarrhea, tenesmus, and abdominal pain. He simultaneously complained about weakness of arms and legs. On examination, he was dehydrated, respiratory and cardivascular systems were normal, abdominal tenderness was on all quadrants of abdomen, sound of intestine was increased, and liver and spleen were within normal size. On the neurologic examination, mentation and cranial nerve functions were normal, as were ocular funduscopy and cerebellar tests. He could not stand or walk by himself. Weakness and decreased muscle tone were determined in legs and arms. Muscle strength was 3+ to 4+ in the arms, and 2+ to 3+ in the legs by manual muscle testing. Muscular atrophy was remarkable in the legs. Perceptions of temperature, touch and vibration were normal. Deep tendon reflexes were decreased and pathologic reflex was absent.

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Peripheral white blood cell count was 11,000/mm^3 leukocytes, neutrophil 9,500/mm^3, hemoglobin 8.5 g/dl, hematocrit 30%, and red blood cell count 4,000,000/mm^3. Erythrocyte sedimentation rate was 60 mm/h, and C-reactive protein was 31.3 mg/dl. Serum iron concentration and total iron binding capacity were 25 and 200 µg/dl, respectively. Platelet counts, glucose, serum urea, serum sodium, serum potassium, transaminase, albumin, fibrinogen, prothrombin time, rheumatoid factor, antinuclear antibody, anti-ds DNA, p-ANCA, c-ANCA, antimitochondrial antibody, anti-Ro, anti-La, immunoglobulins, Ig A and Ig G antigliadin antibodies, Ig A antiendomysium antibody, antibodies to

Table 1. Electrophysiological findings of the patient

Motor Nerve Conduction Studies	**Latency (ms)**	**Amplitude**	**Velocity (m/s)**	**Normal Limit (Velocity: m/s, TL: ms)**
Peroneal Nerve				
TL (Ankle-EDB)	4.5	0.8* µV		5.2 ms
Knee-Ankle		0.7* µV	37	41.4 m/s
Pop-Knee		0.7* µV	36	41.0 m/s
F wave	Ø*			51.2 m/s
Tibial Nerve				
TL (mm-AH)	5	1.1* µV		5.4 ms
Pop-Ankle		1* µV	33	40.2 m/s
F wave	58*			52.0 m/s
Median Nerve				
TL (W-APB)	3.3	3* µV		3.9 ms
Forearm		3* µV	43	45.8 m/s
Upper arm		2.5* µV	45	53.7 m/s
Axilla		2* µV	46	56 m/s
F wave	33*			30.1 ms
Ulnar Nerve	**Latency (ms)**	**Amplitude**	**Velocity (m/s)**	**Normal Limit (Velocity: m/s, TL: ms)**
TL (W-ADM)	3.6	3.5* µV		3.6 m/s
Forearm		2* µV	44	45.2 m/s
Upper arm		2* µV	40	43.7 m/s
Sensory Nerve Conduction Studies				
Median Nerve				
W - DII (Antidromic)	2.7	30 µV	48	41.2 m/s
E - DII (Antidromic)	5.2	20 µV	45	43.7 m/s
Ulnar Nerve				
W- DV (Antidromic)	1.9	25 µV	50	42.2 m/s
E - DV (Antidromic)	4.7	15 µV	48	44.3 m/s
Sural Nerve (Orthodromic)	2.4	15 µV	48	34.7 m/s
Somatosensory Evoked Potentials (SEP)				
Tibial SEP				
mm- Scalp (P40)	40	2 µV		41.6 ms
mm- T12 (N21)	22	1.5 µV		22.1 ms
Median SEP				
W - Scalp (N20)	21	4.2 µV		21.6 ms
W - Cervical 7 (N13)	12.5	3.1 µV		13.4 ms
W - Erb (N9)	8.9	7 µV		9.7 ms

TL: Terminal latency. EDB: Extensor digitorum brevis. AH: Abductor hallucis. FH: Fibular head, mm: Medial malleolus, Pop: Popliteal, ADM: Abductor digiti minimi, APB: Abductor pollicis brevis, W: Wrist, E: Elbow, A: Axilla, DII: Digit II, DV: Digit V, T: Thoracic vertebrae,
*Above the laboratory limits, All records were made bilaterally using surface electrodes. As there was no difference between the values of the two sides, only the results of one side are presented

Campylobacter jejuni, Yersinia enterocolitica and Borrelia, cryoglobulin, hemoglobin A1C, serum vitamins A, B1, B6, B12 and folic acid levels, serum ferritin, urine for Bence Jones protein, serum cortisol, serum calcium, thyroid stimulating hormone, Gruber-Widal test, cultures of blood and stool, detection of Giardia lamblia, Salmonella, Shigella, Escherichia coli and Yersinia enterocolitica on stool, Clostridum difficile toxin-A, electrocardiogram, telecardiography and abdominal ultrasound were normal or negative. Sural nerve biopsy was not performed because the patient did not consent to the intervention. Because electrophysiological findings of the sural nerve were available and sensory conduction investigations were completely normal, we did not insist on biopsy.

Electrophysiological Findings

In our study, after onset of the symptoms, we performed the nerve conduction velocities (NCVs) of motor and sensory nerves in upper and lower extremities. NCV findings are shown in Table 1.

Sensory conduction investigations in upper and lower extremities were normal. Motor NCV investigations were revealed as motor polyneuropathy with axonal degeneration, which was relatively worse in lower than in upper extremities. Concentric needle electromyography (EMG) was performed in the proximal and distal sites of both upper and lower extremity muscles. Rare fibrillation and positive sharp waves were observed in the muscles of lower extremities. The recruitment pattern was reduced with normal duration of motor unit potentials (MUP). Somatosensory evoked potentials were normal.

We added 60 mg prednisone to mesalazine therapy. After two months, his general and neurological status were markedly improved.

DISCUSSION

The clinical and histological findings in this patient established a diagnosis of UC. The neuropathy character in this patient was an axonal motor polyneuropathy. The diagnosis of neuropathy was made according to NCV.

Extraintestinal complications of inflammatory bowel disease (IBD) are common, but neurologic involvement is rarer than that of other systems (4). In this patient, the neuropathic manifestations paralleled the course of UC severity, revealing that the neuropathy did not coincide with UC.

The majority of neuropathies in UC are related to an immunologic mechanism, although non-immunologic mechanisms also have a role (4). Poor intake of vitamin B12 and folic acid or inadequate absorbtion of these vitamins can cause peripheral neuropathy and subacute degeneration of spinal cord (7), but vitamin B-12 and folic acid levels were in normal limits in our patient. Sulfasalazine therapy can cause sensorimotor neuropathy (8). As sulfasalazine was not in our therapy regimen, the neuropathy was not associated with sulfasalazine. A peripheral neuropathy can occur after a prolonged administration of metronidazole. Neuropathy is improved after cessation of the drug (6). Metronidazole was not used when the neurologic symptoms occurred.

Peripheral neuropathy often occurs as a side effect of medication or as a manifestation of systemic disease, i.e. diabetes mellitus, sepsis, carcinoma. There are few reports of peripheral neuropathy with UC. Chronic polyneuropathy is clinically similar to chronic inflammatory demyelinating neuropathy (9), perineuritis (10), Guillain-Barre syndrome (11,12), mononeuropathy multiplex (13), subacute autonomic neuropathy (14), and axonal mononeuritis (3).

However, an axonal motor polyneuropathy in UC as found in the present case was described in a few adult patients. In a retrospective study conducted by Larrode et al. (5), a peripheral sensorimotor neuropathy developed in four patients with a parallel course to IBD. One case was secondary to vitamin B-12 deficiency, another was caused by metronidazole neurotoxicity and in the remaning two cases the polyneuropathy was caused by the activity of IBD itself. In one of these two cases, the nerve biopsy demonstrated an axonal neuropathy. In addition, in a case report published by Greco et al. (6), a six-year-old girl with UC had an axonal sensorimotor polyneuropathy. Three months after treatment with steroid, she had a gastrointestinal recovery and neurological symptoms were improved. After six months, she presented a full clinical recovery with normal gait.

For these reasons, we conclude that our patient's neuropathic manifestations were associated with UC. An axonal motor polyneuropathy can be an extraintestinal manifestation of UC.

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