Case Report

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Acute Motor and Sensory Axonal Neuropathy (AMSAN) Associated with COVID-19 Infection; A Case Report

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Abstract

Myalgia and headache are relatively common in COVID-19 disease, but a serious neurological disease is uncommon. In this case, we describe the symptoms and clinic of AMSAN, a rare variant of Guillain Barre syndrome (GBS) due to COVID 19. We presented a case of AMSAN, a rare variant of GBS, in a 46-year-old male patient with poor overall condition that did not recover after COVID-19 disease, loss of strength and decreased sensation in distal limbs. electromyography-nerve conduction study findings were suggestive acute motor and sensory axonal neuropathy. Cerebrospinal fluid analysis was elevated protein with a normal white blood cell count. The clinical diagnosis of AMSAN supported by results of diagnostic testing such as cerebrospinal fluid and electromyography-nerve conduction study. We added another GBS case due to Covid-19 infection to the literature. It should always be kept in mind that GBS may develop after the COVID-19 disease.

Keywords: AMSAN, COVID-19, Guillain Barre Syndrome, neuropathy

Introduction

COVID 19 diseases surprised the scientific world with various clinical findings. Its plain findings range from mild viral syndrome with asymptomatic infection, fever, myalgia or cough to severe pneumonia that requires a ventilator, worsens rapidly and leads to early death (1). The spike proteins of the virus determine tissue tropism by using the angiotensin converting enzyme type 2 (ACE-2) receptor to bind to cells. The ACE-2 receptor can be found in nervous system tissue and endothelial cells, between tissues of many other organs (2).

Myalgia and headache are relatively common in COVID-19 disease, but a serious neurological disease is rare. Similar to other coronaviruses, severe acute respiratory syndrome coronavirus (SARS-CoV) and Middle East respiratory syndrome coronavirus (MERS), SARS-CoV-2 have been found to have central and peripheral nervous system manifestations (3, 4). Studies have reported that the most common neurological symptoms among individuals infected with COVID-19 are ischemic stroke, Guillain-Barré Syndrome (GBS), and ICU syndrome-related encephalopathy (5).

The most common variant forms of Guillain-Barré syndrome are acute inflammatory demyelinating polyradiculoneuropathy (AIDP), Acute motor axonal neuropathy (AMAN), Acute motor and sensory axonal neuropathy (AMSAN), Miller Fisher syndrome (MFS), Bickerstaff brainstem encephalitis (BBE). In a meta-analysis, acute AIDP variant, one of the subtypes of Gullian Barre syndrome, was demonstrated to be associated with COVID 19 more frequently in the published cases. However, other subtypes were observed very rarely (6).

In this case, we describe the symptoms and clinic of AMSAN, a rare variant of Guillian Barre due to COVID 19.

Case Report

A 46-year-old male patient presented to the emergency department with complaints of shortness of breath and cough. The patient was hospitalized due to hypoxia and positive SARS-CoV2 PCR test. He had a history of Type 2 diabetes mellitus, hypertension, hypothyroidism and asthma. Dexamethasone 8 mg/day, IL-1 antagonist (Anakinra subcutaneous (100 mg/day)), ceftriaxone 2 gr twice/day, Clarithromycin 500 mg twice/day was given when he was hospitalized due to COVID-19 disease. He was discharged on oxygen therapy. When the patient was discharged, there was loss of strength in his lower extremities, but thought to be caused by dexamethasone

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treatment and malnutrition. He was admitted to the emergency service again because his general condition did not improve and progressive, symmetrical weakness and loss of strength in both lower extremities increased.

On physical examination, the patient did not have fever, and his blood pressure was 110/60 mm/hg, heart rate was 70 beats/minute, respiratory rate was 12/minute, and oxygen saturation in room air was 70%. During the hospitalization of the patient, his consciousness was slow and his orientation and cooperation were poor. In the muscle strength examination, 4/5 in the bilateral upper extremity distal and 3/5 in the bilateral lower extremity distal weakness were observed according to the Medical Research Council (MRC) scale. Deep tendon reflexes were hypoactive. Drop foot was present in both feet bilaterally. There was a decrease in vibration and fine touch sensation in the distal ankle joints.

In laboratory examination;
Laboratory examination results were as follows: serum glucose 188 mg/dL; blood urea nitrogen: 10 mg/dL; creatinine 0.9 mg/dL; alanine aminotransferase 8 IU/L; aspartate aminotransferase 5 IU/L; sodium 138 mmol /L; potassium 3.5 mmol/L; hemoglobin 8.9 g/dL white blood cell count 7420 cells per microliter, erythrocyte sedimentation rate 11 mm/hr, C-reactive protein 52.1 mg/L, procalsitonin 0,2 ug/L, hemoglobin A1c 8.9 g/dL. Negative glucose and ketone were observed in complete urinalysis **(Table 1).** Potassium chloride (40 mEq KCL in 1000 cc isotonic saline) replacement was given to the patient with hypokalemia. Blood and urine culture was performed from the patient with elevated CRP on admission. On the third and fifth days of hospitalization(respectively), the CRP level was 84.5 -111.6, procalcitonin 0.5-0.2. Multidrug resistance klebsiella pneumonia was grown in the blood culture taken at his hospitalization. The patient was started on 150 mg Polymyxin E (Colistin) twice a day and 1 gr Meropenem three times a day. CRP and procalcitonin levels decreased in the follow-ups.

In the follow-ups, the potassium level returned to normal. Since the patient's hemoglobin level was low, vitamin B12 1278 ng/L, folic acid 12 ug/L, percent tranferrin saturation 71, the patient's anemia was evaluated as anemia of chronic disease. The patient's TSH level was 150 mU/ml. The dose of Levothyroxine was increased, and the TSH level decreased in the follow-ups. The respiratory rate was 12/minute and the oxygen saturation was 70% in room air, which may have been due to the patient's severe hypothyroidism.

Cervical and brain magnetic resonance imaging (MRI) was performed. Brain MRI was normal, bulging was present between C2-7. Electromyography (EMG) was performed with the preliminary diagnosis of critical illness myopathy and Gullian Barre. There was right upper ulnar motor and sensory loss. The motor action potential of the right median nerve was extremely low, and the motor action potentials of the tibialis anterior were extremely low. In needle EMG, there were widespread denervation potentials in the right and upper and lower extremities. Findings are consistent with severe motor and sensory axonal neuropathy (AMSAN) in the acute phase (table 2.). In Cerebrospinal fluid white blood cell count was 4, albumin was 52.8mg/dL (normal:0-30), microprotein was 99.68mg/dL (normal:15-40).

Plasmapheresis treatment was started for the patient with the diagnosis of AMSAN. Plasma exchange was done with albumin. Before plasma exchange, the estimated plasma volume (EPV) was calculated from the patient's weight

Table 1: Laboratory values of the AMSAN patient associated with COVID-19

	VALUE	REFERANCE RANGE
COMPLETE BLOOD COUNT		
Total leucocyte count 10^3 uL	7,42	4,5-10
Neutrophils. %	62,3	42-75
Lymphocytes %	19,1	12-48
Haemoglobin. %	8,9	13-17
Haematocrit. %	26,2	40-49
Platelets. 10^3 uL	279	150-450
ELECTROLYTES		
Sodium mmol/L	138	136-145
Potassium mmol/L	3,5	3,5-5,1
Chloride mmol/L	96	98-107
OTHERS		
Glycemia mg/dL	188	70-110
Urea Nitrogen mg/dL	10	6-20
Serum Creatinine mg/dL	0,90	0,70-1,20
AST U/L	5	0-40
Albumin g/L	27,0	35-52
Percent saturation of transferrin	71	15-50
Ferritin ug/L	722	30-400
B12 vitamin ng/L	1278	197-771
Folic Acid ug/L	12	3,8-26,8
D-Dimer ug/L	532	0-500
CRP ug/L	52,1	0-5
Erythrocyte Sedimentation Rate (ESR) mm/h	62	0, -15
CEREBROSPİNAL FLUİD		
CSF Albumin mg/dL	52,8	0-30
CSF Sodium	144	
CSF Chloride	118	
CSF Glucose mg/dL	134	40-70
CSF Microprotein mg/dL	99,6	15-40
CSF WBC 10^3 uL	0,004	
CSF PMN % CSF PMN # 10^3 uL	25 0,001	
CSF RBC 10^6 uL	0	
CSF CULTURE	NEGATİVE	

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Table 2: EMG findings of an AMSAN patient associated COVİD- 19

MOTOR NERVE CONDUCTION STUDIES				
NERVE	LAT	AMP	CV	F-M LAT
MEDIANUS MOTOR RIGHT				
WRIST – APB	2,63 ms	1,65 mV		15,1 ms
ELBOW - WRIST	7,94 ms	0,87 mV	50,8 m/s	
PERONEUS MOTOR RIGHT				
AB. KNEE- FIB. HEAD	5,46 ms	0,026 mV		
TIBIALIS MOTOR RIGHT				
ANKLEE – ABD. HAL	4,58 ms	2,8 mV		51,3 ms
KNEE - ANKLE	15,6 ms	0,23 mV	41,5 m/s	
ULNARIS MOTOR RIGHT				
WRIST - ADM	35,1 ms	-		

SENSORY NERVE CONDUCTION STUDIES			
NERVE	PEAK LAT	AMP	CV
MEDIANUS SENSORY RIGHT			
DIG III-WRIST	3,06 ms	16,6 uV	62,0 m/s
SURALIS SENSORY RIGHT			
MID. LOWER LEG – LAT. MALLEOLUS	2,74 ms	2,7 uV	62,5
ULNARIS SENSORY RIGHT			
DIG V-WRIST	-	-	-

and hematocrit using the formula EPV = $(0.065 \times \text{wt [kg]}) \times (1 - \text{Hct})19$ (average 1-1.3 plasma volume change for each session). After the first day of plasmapheresis, status epilepticus developed in the patient, anticonvulsant treatment (keppra 1000mg twice/day) was started and the patient was intubated. No pathology was detected in the EEG of the patient. Since it was the weekend, EEG could be taken two days later.

The EEG may therefore be normal. The patient was extubated after the 5th session of plasmapheresis. After the 7th session of plasmapheresis. The patient was not hypoxemic before the seizure. The patient was intubated because he could not protect his breathing during the seizure. The patient who was extubated in the 9th session of plasmapheresis was re-intubated from the 10th session. While the first reason for intubation was status epilepticus, the patient was later intubated due to respiratory failure. The patient was extubated 3 days after the third intubation. The patient, whose muscle weakness relatively improved during the follow-ups, was discharged on the fourth day of his extubation (31st day of hospitalization) at the request of himself and his relatives.

Conclusion

In this study, we reported a case diagnosed as AMSAN, a rare variant form of Guillian Barre after COVID-19. We considered that GBS could be a possible diagnosis because of bilateral weakness and relatively symmetrical limb weakness and monophasic course after COVID 19 in our patient. We confirmed our diagnosis as a result of EMG and CSF sampling. COVID-19 often presents with respiratory symptoms, but acute cerebrovascular diseases, seizures, anosmia, meningitis, encephalitis, and skeletal muscle involvement are neurological manifestations (7). Recently, the increase in the number of Guillain Barre cases in COVID 19 disease is noteworthy (8). In the same meta-analysis, 80.5% of the patients had AIDP variant of GBS, and 3 patients had only AMSAN syndrome. The studies on coronaviruses have demonstrated that these viruses have neurotrophic and neuroinvasive properties. It is also unclear whether COVID-19 causes the production of antibodies to specific gangliosides, which usually occurs in certain forms of GBS (9).

GBS is defined as a rare, but potentially fatal, immune-mediated disease of nerve roots that is usually triggered by infections. Diagnosis of GBS in SARS-CoV-2 is particularly difficult because symptoms such as shortness of breath and fatigue can be misinterpreted as secondary to SARS-CoV-2, delaying GBS assessment. Since our patient was hospitalized in the intensive care unit, the symptoms were considered as critical illness myopathy in the first place and it was a late case.

As a result, GBS and its variants should be kept in mind in the presence of symptoms related to peripheral nervous system disorders such as decreased or absent reflexes, elevated and loose paralysis in COVID 19 patients.

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