스트렙토코쿠스비리단스에 의한 수막척수신경근염: 증례보고

도희재, 권소현, 서한길

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A Case of Meningomyeloradiculitis Caused by a Rare Pathogen, Viridans Streptococci

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Meningitis due to viridans streptococci is rare, and post-meningitis spinal involvement resulting in myelopathy or polyradiculopathy is an uncommon neurologic complication of bacterial meningitis. For that reason, there are no reports of post-meningitis spinal involvement due to viridans streptococci. We report the case of a man with meningomyeloradiculitis who presented with flaccid paraplegia after viridans streptococci meningitis. He was diagnosed with meningomyeloradiculitis through clinical, electrodiagnostic, laboratory, and imaging studies. Among the studies conducted, the electrodiagnostic studies were most useful in confirming the diagnosis. Eventually, his motor power in the bilateral lower extremities improved and he was able to walk by himself with a cane. This report is presumably the first to present a case of meningomyeloradiculitis after viridans streptococci infection. Clinicians should be aware of the related complications post-meningitis and perform evaluations of unusual symptoms not associated with meningitis.

Key Words: meningitis, meningomyeloradiculitis, viridans streptococci

Introduction

The viridans streptococci group, which is considered to be a low-virulence pathogen, is a bacterium commonly found in the human oral cavity and gastrointes-

tinal tract.¹ It is known to be related to minor dental problems as well as serious endocarditis and bacteremia, especially in neutropenic patients with cancer.^{1,2} However, of the possible complications that may arise, meningitis due to viridans streptococci is rare, with an incidence of only 0.1% in children and 2.8% in adults with bacterial meningitis.^{1,3} Moreover, post-meningitis spinal involvement resulting in myelopathy or polyradiculopathy is an uncommon neurologic complication of bacterial meningitis.^{4,5} For that reason, there have been no previous reports of post-meningitis spinal involvement due to viridans streptococci.

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We report the case of a man with meningomyeloradiculitis who presented with flaccid paraplegia after viridans streptococci meningitis.

Case Report

A previously healthy 38-year-old man presented with loss of consciousness and generalized tonic clonic seizure, and was admitted to the emergency room. He reported having a headache and the sensation of chills for the five days prior to admission. At the emergency room, his mental status, according to the Glasgow Coma Scale E1V2M2, was stupor. His pupils were fixed and his bilateral extremities were flaccid, i.e., grade 1 on the Medical Research Council (MRC) scale. Brain magnetic resonance imaging (MRI) showed a 3 cm lobulated mass in his right temporal lobe and diffuse leptomeningeal enhancement in his left lateral ventricle (Fig. 1). These findings were clinically considered to be a brain abscess combined with meningoencephalitis. The results of a cerebrospinal fluid study demonstrated bacterial meningitis. Ostitomeatal unit computed tomography (OMU CT) was performed, and periodontoid lucency, indicative of periodontitis, was observed. He

was treated with antibiotics and had an operation for removal of the brain abscess. Streptococcus species, viridans group, was cultured from the blood and abscess.

Although the patient was alert on postoperative day six, he still had flaccid arms and legs and left facial palsy. To investigate his motor weakness, electrodiagnostic studies and spine MRI were performed. There was no abnormal finding in nerve conduction studies (NCS) and needle electromyography (EMG) of left arm and leg. In the spine MRI conducted to investigate his motor weakness, equivocal thin enhancement along the spinal cord and nerve roots at the lower thoracolumbosacral spine with a small central protrusion at L1-2 and an extrusion at L4-5 was observed (Fig. 2).

One month postoperatively, he was transferred to the Department of Rehabilitation Medicine for rehabilitation. Upon cranial examination, he was found to have ptosis, esotropia, and lateropulsion in the left eye with left facial palsy. Although he recovered motor power in his bilateral upper extremities (MRC grade 5), his lower extremities were still flaccid (MRC grade 1). On sensory examination, he complained of pain and hypesthesia

Fig. 1. Brain T2-weighted magnetic resonance imaging demonstrated a 3 cm lobulated mass (arrow) in the right temporal lobe and diffuse leptomeningeal enhancement in the left lateral ventricle.

Fig. 2. Spine enhanced magnetic resonance imaging demonstrated equivocal thin enhancement along the spinal cord (arrow head) and nerve roots at the level of the lower thoracolumbosacral spine (arrow) with a small central protrusion at L1-2 and an extrusion at L4-5.

sia bilaterally from the thigh to the medial side of the ankle. The deep tendon reflexes at all joints were decreased. Pathologic reflexes were not observed. Bladder and bowel functions were intact.

An electrodiagnostic study was performed to investigate peripheral nerve root involvement as a possible cause of the bilateral weakness in the lower extremities. Sensory NCS in the bilateral superficial peroneal nerve and sural nerve revealed normal results (Table 1). Motor NCS in the bilateral extensor digitorum brevis muscle and abductor hallucis muscle also were observed to be normal (Table 1). F waves at both abductor

hallucis muscles were normal, though the H reflexes were non-responsive at both soleus muscles. In a left facial nerve motor conduction study, all muscles innervated by the facial nerve revealed no response. In addition, the left blink reflex, which was evoked at the level of the supraorbital nerve, was absent. In needle EMG at both lower extremities, there were denervation potentials in the bilateral iliopsoas, rectus femoris, tensor fasciae latae, tibialis anterior, left peroneus longus, right biceps femoris, and gluteus maximus, with no voluntary motor unit action potential (MUAP) activity (Table 2). Moreover, on the left side of the face, the

Table 1. Nerve Conduction Study

Nerve	Stimulation site	Recording site	Latency (ms)		Amplitude (μV or mV)		CV (m/s)	
			Rt	Lt	Rt	Lt	Rt	Lt
Sensory nerve conduction								
Superficial peroneal	Ankle	Foot	2.55	2.05	10.30	12.80	-	-
Sural	Lower leg	Lateral malleolus	3.15	3.00	18.50	15.10	-	-
Motor nerve conduction								
Common peroneal	Ankle	EDB	5.90	5.95	3.70	3.00		
	Fibular head		13.25	13.05	3.70	2.80	40.80	42.30
Tibial	Ankle	AH	5.35	6.05	7.40	8.00		
	Popliteal fossa		14.90	15.65	6.30	7.40	41.40	41.10

All latencies are onset latencies. Unit of amplitude is used as μ V on sensory nerve conduction and mV on motor nerve conduction. CV: conduction velocity, Rt: right, Lt: left, EDB: extensor digitorum brevis, AH: abductor hallucis

Table 2. Needle Electromyography

Muscle	ASA			MUAP			Interference
	IA	Fib	PSW	Amp	Dur	Poly	
Rt IP	N	2+	2+	-	-	-	NA
Rt RF	N	2+	2+	-	-	-	NA
Rt VM	N	None	None	-	-	-	NA
Rt TA	N	2+	2+	-	-	-	NA
Rt PL	N	None	None	-	-	-	NA
Rt TFL	N	2+	2+	-	-	-	NA
Rt GCM	N	None	None	-	-	-	NA
Rt BIC FEM (L HEAD)	N	2+	2+	-	-	-	NA
Rt Gmax	N	2+	2+	-	-	-	NA
Lt IP	N	2+	2+	-	-	-	NA
Lt RF	N	2+	2+	-	-	-	NA
Lt VM	N	None	None	-	-	-	NA
Lt TA	N	3+	3+	-	-	-	NA
Lt PL	N	2+	2+	-	-	-	NA
Lt TFL	N	3+	3+	-	-	-	NA
Lt GCM	N	None	None	-	-	-	NA

ASA: abnormal spontaneous activity, MUAP: motor unit action potential, IA: insertional activity, Fib: fibrillation, PSW: positive sharp wave, Amp: amplitude, Dur: duration, Poly: polyphasic motor units, Rt: right, Lt: left, N: normal, NA: no activity, IP: iliopsoas, RF: rectus femoris, VM: vastus medialis, TA: tibialis anterior, PL: peroneus longus, TFL: tensor fasciae latae, GCM: gastrocnemius, BIC FEM (L HEAD): biceps femoris long head, Gmax: gluteus maximus

frontalis and orbicularis oris had denervation potentials with no MUAP activity or single interference patterns. In addition to NCS and needle EMG, somatosensory evoked potentials (SEP) in the both tibial nerves were performed. While the waveform was W-shaped complex and the latency of P40 was 40.10 ms within normal range in the left SEP, the amplitude of P40 was 0.68 μ V, which was smaller than the lowest reference value. It meant any defect in the somatosensory pathway from tibial nerve to cortex. In conclusion, the electrodiagnostic study revealed bilateral myeloradiculopathy involving the L2-S1 roots with severe axonotmesis that was clinically indicative of meningomyeloradiculitis, and left facial neuropathy with severe axonotmesis.

At discharge, motor power in both of the patient's hips had recovered (MRC grade 3). After 6 months, the patient's proximal and distal motor power in the bilateral lower extremities had improved to MRC grade 4~5 except in both ankles; however, the patient was able to walk with a cane.

Discussion

Streptococcus is a gram-positive spherical bacterium and a heterogeneous group that includes *Streptococcus pneumoniae*, viridans streptococci, β -hemolytic streptococci and others.⁶ Although *S. pneumoniae* is the most common pathogen for bacterial meningitis, and is thought to cause 21~31% of all adult cases of bacterial meningitis, meningitis caused by viridans streptococci is rare, occurring in only 2.8% of cases.^{1,3,6}

The neurologic complications of bacterial meningitis that have been reported in the literature include hearing deficits, seizures, hydrocephalus, cranial nerve palsies, hemiparesis, and quadriparesis. However, spinal involvement in spinal meningitis or myeloradiculitis after nontuberculous bacterial meningitis is uncommon.⁴ In nontuberculous bacteria, *Streptococcus pneumoniae*, *Neisseria meningitidis*, *Borrelia burgdorferi*, and *Brucella* have been reported as pathogens for post-meningitis spinal involvement.^{4,5,7-9} Several mechanisms

have been considered in regards to the pathogenesis, but none have been conclusively determined.⁴ First, as a result of vasculitis or systemic hypotension, impaired blood flow have been considered. Second, constrictive arachnoid adhesions might cause radicular pain and progressive cord dysfunction. Finally, lesion of the anterior horn cells has been considered as a polio-myelitic form. However, a direct infective pathology, such as the pressure of meningeal exudate on the nerve, is doubtful because in most cases the purulent cerebrospinal fluid is clear.⁷ Post-meningitis polyradiculopathy might have a good prognosis without therapy.⁵ Beneficial effect of treatment with corticosteroids is unknown.⁵ On the other hand, it is important to maintain oxygenation, blood pressure, fluid balance and nutrition in spinal cord involvement.¹⁰ Unlike the polyradiculopathy, post-meningitis spinal cord involvement has an unfavorable prognosis with residual abnormalities such as spasticity or bowel and bladder dysfunction.¹⁰

In this patient, meningitis was caused by viridans streptococci, a rare pathogen for meningitis, and resulted in meningomyeloradiculitis, an extremely uncommon neurologic complication of bacterial meningitis. Because of the rarity of the pathogen and this complication, this report is presumably the first case of meningomyeloradiculitis after viridans streptococci infection in the literature, confirmed by clinical, electrodiagnostic, laboratory and imaging studies. There have been a few reports involving the *Streptococcus* species, but not involving viridans streptococci. Kikuchi et al.⁴ reported post-meningitis cauda equine syndrome that presented with flaccid paraplegia, saddle anesthesia, and bladder and bowel dysfunction due to *Streptococcus pneumoniae*. Although the neurologic complication was diagnosed by enhanced MRI of the brain and spine, the report was limited in that the sensory NCS and brainstem auditory-evoked potentials were normal and needle EMG was not performed. In that respect, our report is clinically important in terms of the neurophysiological information it provides on the disease. On the other hand, the results of the electrodiagnostic

study should be carefully considered. Unlike legs, his motor weakness in arms, left oculi and left face may be caused by meningoencephalitis through clinical courses, brain MRI and electrodiagnostic studies. Peculiarly, during needle electromyography, all muscles in the lower extremities revealed no activity, despite the motor nerve conduction studies for some muscles being normal and an absence of definite muscle atrophy. The reason for this may be due to an effect of the lesion on the central and the peripheral nervous systems.

In conclusion, this report demonstrated a rare complication of meningomyeloradiculitis after viridans streptococci infection. Clinicians should be aware of related complications following meningitis and perform evaluations of unusual symptoms that are not associated with meningitis.

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