Recurrent Herpes Zoster Myelitis

Recurrent zoster myelitis is quite rare. We present a previously healthy 27-year-old woman who developed recurrent attacks of myelopathy shortly after the characteristic skin rashes of herpes zoster. Magnetic resonance imaging studies demonstrated each lesion in the spinal cord at the same segments as the skin lesions. She had two attacks at opposite sites at the same spinal cord level and complete recovery after being treated with intravenous acyclovir. We suspect that direct invasion of varicella zoster virus was the cause of recurrent myelopathy in our patient.
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INTRODUCTION

Herpes zoster viral infection is a sporadic illness caused by the varicella zoster virus (VZV) and is characterized by a vesicular dermatomal rash (1). Although the vast majority of cases with herpes zoster are uncomplicated and self-limiting, neurologic complications such as postherpetic neuralgia, segmental sensory loss or motor paresis, encephalitis, myelitis, multifocal leukoencephalopathy and cerebral vascular occlusion may develop (2). Among them, myelitis is rare, and recurrent cases are even more rare (3~5). We present a patient who showed characteristic benign recurrent zoster myelitis. Our patient appears to be unique in that each spinal cord lesion demonstrated on magentic resonance imaging (MRI) was preceded by a typical skin lesion of herpes zoster at the same spinal segment.

CASE REPORT

A 27-year-old, previously healthy woman was admitted to the Department of Neurology of Yonsei Medical Center because of chest tightness, dyspnea, and paresthesia and weakness in her left arm and leg. About sixty days prior to admission, she was evaluated at the Department of Neurosurgery at Chonju Presbyterian Medical Center because of weakness in her right arm and leg, sensory loss in her right occiput and neck (C_{3-4} dermatomal distribution), and paresthesia below her neck, which developed four days after painful vesicular eruptions on her right posterior auricular area. An immediate T2-weighted MRI study of the cervical spine showed multifocal high signal intensity lesions throughout the long segments of the cervical cord (C_{2-7}) (Fig. 1-A); the lesion was eccentric to the right (Fig. 1-B). Her cerebrospinal fluid (CSF) contained 8 white blood cells, while the protein and glucose were normal. After diagnosis of zoster myelitis, intravenous acyclovir (1500 mg/day for 7 days) and dexamethasone (20 mg/day) were administrated. Oral acyclovir was continued for 10 days after the course of the intravenous treatment. Over the ensuing days, her weakness and sensory change improved.

Forty days after the first vesicular eruptions on her right posterior auricular area, she again developed painful grouped vesicular eruptions on her left paramedian posterior neck area. Seven days later, she was readmitted to Chonju Presbyterian Medical Center when she noticed tightness and weakness in her left arm. A follow-up MRI study of the cervical spine revealed a high signal intensity lesion with diffuse swelling, eccentric to the left, throughout the C_{2-5} segments of the cervical cord; a primary right-sided lesion was inconspicuous (Fig. 2). She was again treated with intravenous acyclovir.

She was transferred to the Department of Neurology of Yonsei Medical Center 11 days after her second admission when she complained of chest tightness and dyspnea. On admission, her vital signs were stable. Multiple postherpetic scars were present on her left paramedian posterior neck area. She was alert but appeared to be anxious. Muscle power was decreased to 4+/5 in her left arm and leg. Deep tendon reflexes were brisk on her left knee and ankle, and her plantar response

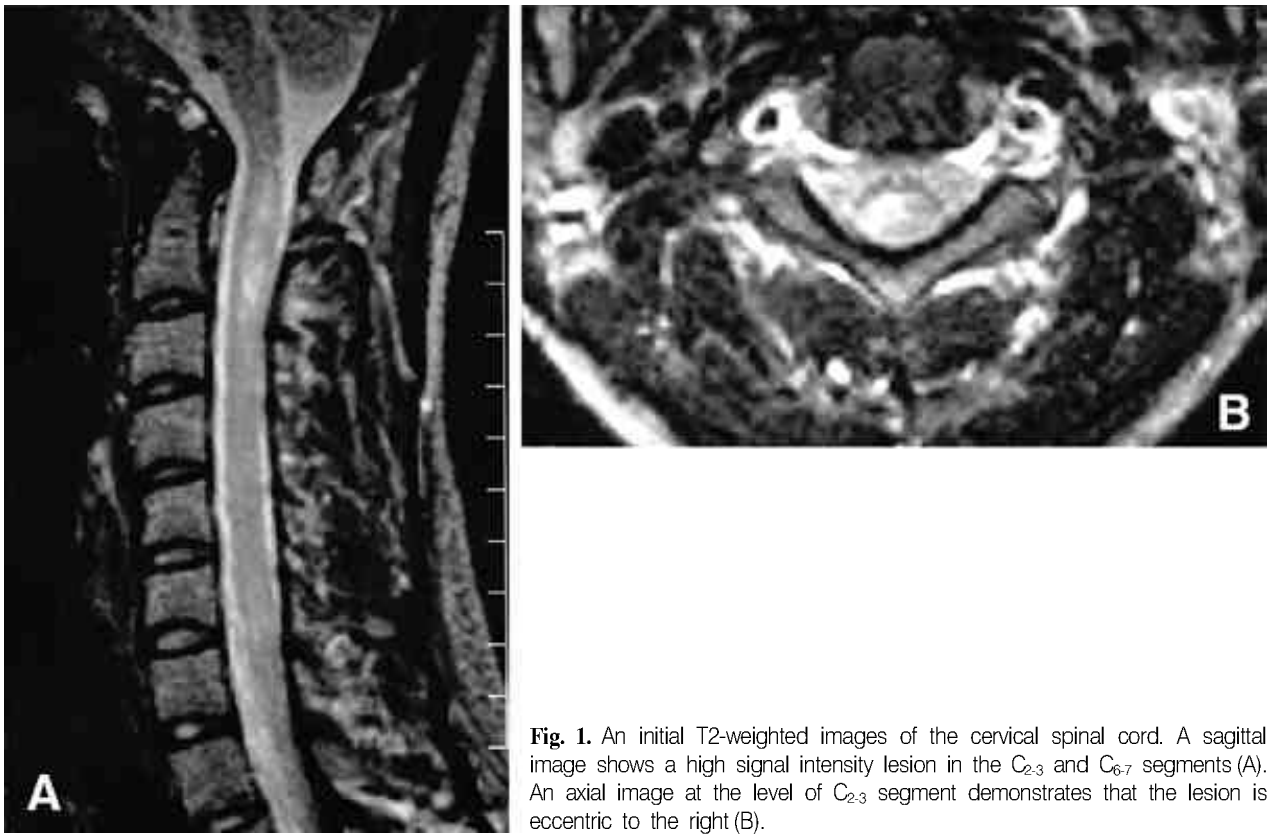

Fig. 1. An initial T2-weighted images of the cervical spinal cord. A sagittal image shows a high signal intensity lesion in the C_{2-3} and C_{6-7} segments (A). An axial image at the level of C_{2-3} segment demonstrates that the lesion is eccentric to the right (B).

was extensor in the left foot. Pain and light touch sensations were moderately decreased on the bilateral C_{3-5} dermatomal distributions, and mildly decreased below the right C_5 dermatomal distribution. Position and vibration sensations were decreased in the left hand. Chest roentgenography, electrocardiography, thyroid function tests, and arterial blood gas analysis were all normal, and psychiatric evaluation revealed that her chest tightness and dyspnea were from a panic attack. Laboratory data revealed a complete blood count, urinalysis, and blood chemistry to be normal. CSF contained 6 white blood cells, while the protein and glucose were normal. Serum and CSF immunoelectrophoreses were not remarkable. The titer of IgG Ab for VZV in serum was increased to 4400 IU/ml, while that in CSF and the titer of VZV IgM in serum and CSF were all negative. Although we performed additional laboratory tests including abdominal and pelvic ultrasound sonography, carcinoembryonic antigen, alpha fetoprotein, beta-2 microglobulin, beta-human chorionic gonadotrophin, anti-human immunodeficiency virus antibody, stool occult blood, and peripheral blood smear, we failed to find any predisposing disease to cause defective cellular immunity. She gradually improved. Three weeks later, we could find neither focal nor lateralizing neurologic deficits.

DISCUSSION

Myelitis is a rare neurologic complication of herpes zoster and is diagnosed based on the close temporal relationship between skin rash and the onset of myelitis (1). There was no doubt about the diagnosis of zoster myelitis in our patient because she developed the characteristic skin rashes of herpes zoster, the temporal relationship was clear in that each interval between the onset of the rash and that of myelitis was 4 days and 7 days respectively, and MRI findings were consistent with those of the previously reported patients (6, 7).

Recurrent cases are even more rare. To the best of our knowledge, three patients have been documented so far. McAlpine et al. (3) and O'Donnell et al. (5) reported patients with recurrent myelitis associated with encephalitis, and Nakano et al. (4) reported a patient with pure recurrent myelitis. Therefore, there has been only one reported patient with pure recurrent involvements of the spinal cord. Our patient is similar to the patient of Nakano et al. in that both developed pure recurrent myelitis, but is differentiated from their patient by the following points : 1) Our patient had a short interval between the development of skin rash and myelopathy in each attack (four and seven days respectively), while their patient had relatively long interval (two weeks) ; 2)

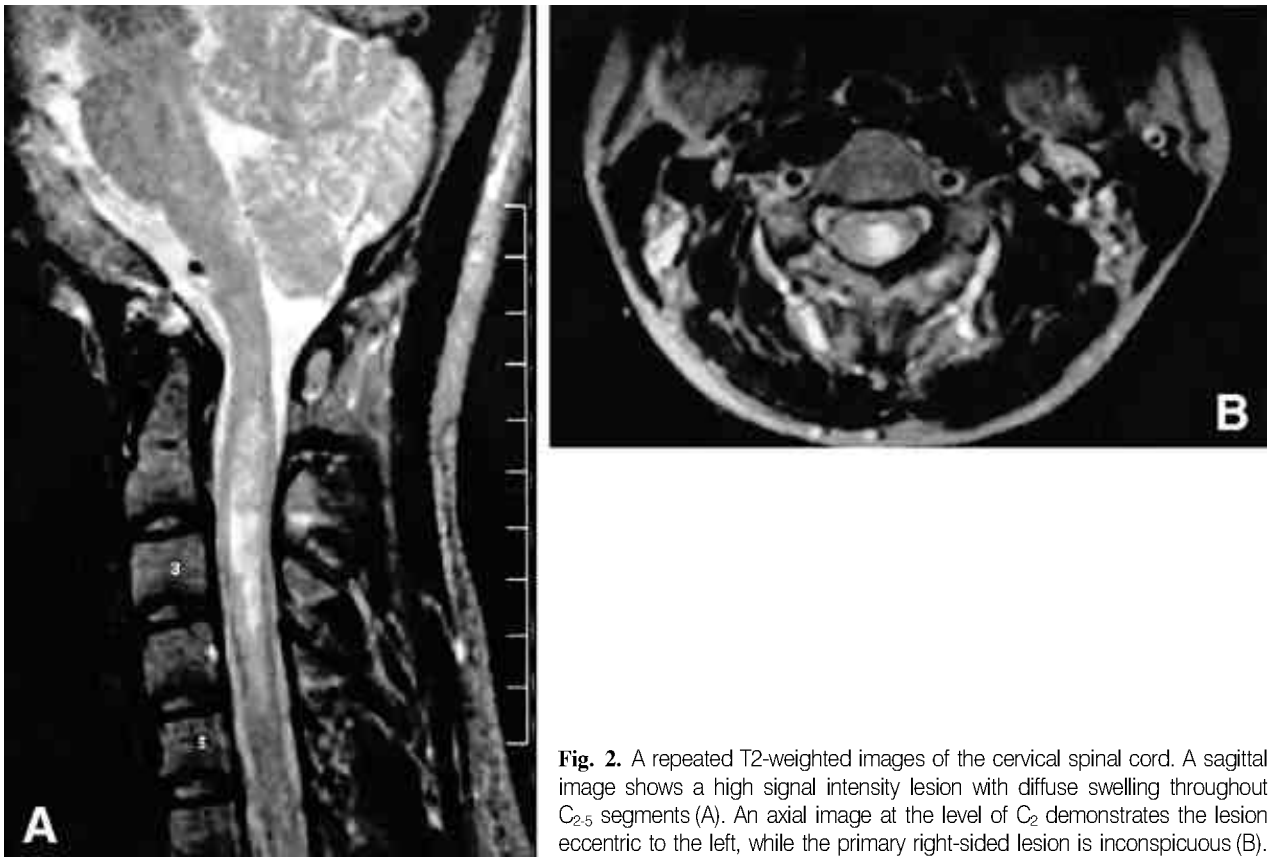

Fig. 2. A repeated T2-weighted images of the cervical spinal cord. A sagittal image shows a high signal intensity lesion with diffuse swelling throughout C_{2-5} segments (A). An axial image at the level of C_2 demonstrates the lesion eccentric to the left, while the primary right-sided lesion is inconspicuous (B).

Each lesion demonstrated with MRI developed at the same segments of the spinal cord as skin rashes in our patient, while a recurring lesion in their patient did not. The site of recurring spinal cord lesion, in their patient, was at least two segments above the recurring skin rash and was in the vicinity of the primarily involved spinal cord ; 3) Our patient responded well to acyclovir, while their patient did not. The exact mechanism of the recurrence in zoster myelopathy is uncertain, although several mechanisms of injury in zoster myelitis, such as direct infection, immune-mediated demyelination, and vasculitis with resultant cord ischemia, have been suggested throughout extensive pathologic studies (1). Nakano et al. suspected immunologic mechanism of a delayed-type hypersensitivity to be the cause of recurrent myelopathy in their patient because of the ineffectiveness of the acyclovir and a relatively long interval between developments of skin rashes and neurologic dysfunctions in their patient. However, our observations of the very close temporal and spatial relationship between the skin rash and myelopathy and the effectiveness of acyclovir strongly suggest that direct viral infection might be the causative factor in our patient.

The clinical course of zoster myelitis is quite variable such as acute, subacute, chronic, or remitting exacerbating myelopathy (1, 8). The prognosis of it is also variable from benign and self-limiting (9, 10) to progressive and fatal (1). This variability may be ascribed in part to the diverse pathologic mechanisms of injury which are mentioned above. Although a pathologic confirmation could not be obtained in our patient, complete functional recovery accompanied by radiological improvement of a primary right-sided lesion on a follow-up MRI strongly suggested that profound destruction or necrosis, a frequent pathologic finding in patients with progressive and fatal myelopathy (1), might be absent. Early therapeutic intervention with acyclovir in our patient appears to have been instrumental in her improvement by preventing multiplication or spread of the virus in the spinal cord (2, 11). Absence of a serious predisposing disease also may have contributed to her excellent prognosis because most of the previously reported patients with poor prognosis had some kind of serious predisposing immune-competent disease (1,8,12).

Defective cellular immunity may increase the likelihood of VZV reactivation (13). It has been described that many patients with zoster myelitis had lymphoma, leukemia, the acquired immune deficiency syndrome, metastatic carcinoma, or systemic lupus erythematosus (1, 8, 12). Although we have failed to find such pre-

disposing diseases, which may have had an effect on our patients cellular immunity during her admission and a six month follow-up course in the outpatient clinic, we do not rule out the possibility of the presence of some kind of defective cellular immunity.

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