

CASE REPORT

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Clinical investigation of segmental zoster paresis: A case of rare motor complications of herpes zoster

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ABSTRACT

Introduction: Motor involvement is a rare complication of herpes zoster (HZ) infection, which is referred to as segmental zoster paresis (SZP).

Case Report: We evaluated the clinical course of a rare case of motor complications of HZ. A 77-year-old man showed limb paresis, considered as SZP of the limb, of which one also had hemidiaphragmatic paralysis. The time between onset and start of antiviral drug administration was 29 days. An almost complete recovery from motor complications was achieved nine months after onset.

Conclusion: Segmental zoster paresis was found in an old patient aged ≥ 75 years. In addition to age, it was suggested that a delay in antiviral drug initiation may have caused SZP. However, further large prospective cohort studies are required. The patient has recovered from motor paralysis slowly over half a year, which is consistent with the prognosis of conventional SZP.

Keywords: Hemidiaphragmatic paralysis, Herpes zoster, Prognosis, Segmental zoster paresis

How to cite this article

Shiraiwa N, Terada M, Tamaoka A, Ohkoshi N. Clinical investigation of segmental zoster paresis: A case of rare motor complications of herpes zoster. J Case Rep Images Med 2023;9(2):1–4.

Article ID: 100075Z09NS2023

doi: 10.5348/100075Z09NS2023CR

INTRODUCTION

Herpes zoster (HZ) is caused by the reactivation of the varicella-zoster virus (VZV) latent in the dorsal root ganglia. It usually occurs when the immune system does not function properly, particularly in older adults. It is characterized by vesicular rash and burning pain. Although post-herpetic neuralgia (PHN) is the most common neurological syndrome, motor involvement is only observed in 0.5–5% of patients with HZ [1–5].

The exact mechanism of motor complications of HZ, referred to as segmental zoster paresis (SZP), is unclear. However, this mechanism has been proposed to be due to the spread of the virus along the nerve [6–8]. Herein, we evaluated the clinical course of a rare case of motor complications of HZ and the possible factors influencing the prognosis.

CASE REPORT

A 77-year-old man had pain in the right side of his neck and found it difficult to raise his right upper limb. After several days, a rash extending from the shoulder to the right forearm appeared. In addition, weakness of the upper limbs occurred and progressed (Figure 1).

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Received: 13 June 2023

Accepted: 26 August 2023

Published: 14 September 2023

As the neck pain preceded the rash for several days, at first he applied a pain-relieving compress for his neck pain every day. That's why he thought a poultrice rash appeared. But he got finally worry about his symptoms and decided to see a doctor because the weakness of his arm was progressed. Approximately one month after onset, the patient visited the neurology department of our clinic. The patient experienced pain and dysesthesia extending from the right shoulder to the upper limbs. Furthermore, an HZ-like rash was observed at the same site. Enzyme immunoassay revealed VZV immunoglobulin G (IgG) antibody levels of >128 (titer <0.2 [-]). Muscle weakness was present in the right deltoid (2-/5), biceps brachii (4/5), and triceps brachii (4/5), according to the Medical Research Council Scale. Muscle atrophy was also observed. An antiviral drug (valaciclovir hydrochloride 3000 mg/day) was started 29 days after onset for seven days. Pregabalin was also administered to manage pain. However, paralysis of the right upper limb worsened: the right deltoid (2/5), biceps brachii (3/5), and triceps brachii (3/5) muscles. Pain and dysesthesia persisted. After an additional seven days of antiviral drug treatment (valaciclovir hydrochloride 3000 mg/day), pain and rash improved. However, weakness worsened to 1+/5 in the right deltoid muscle. Following rehabilitation, weakness was slightly improved in the right deltoid muscle (1+/5), biceps brachii (4-/5), and triceps brachii (4-/5).

Since the patient still had motor complications 2.5 months after onset, he was admitted to the neurology department at a university hospital for examination and treatment. Right upper limb weakness of the C5–C6 innervating muscles and decreased thermal pain sensation were noted. In addition, right phrenic nerve palsy was observed on a chest radiograph (Figure 2A). The patient reported pain extending from the back of the head to the back of the neck. Cerebrospinal fluid analysis showed a protein concentration of 46 mg/dL, cell count of 1/μL, sugar concentration of 58 mg/dL (blood glucose, 105 mg/dL), and negative polymerase chain reaction results for VZV DNA. Cerebrospinal fluid VZV-IgG was weakly positive. Serum VZV-IgG and VZV-IgM were >128.0 and <0.80, respectively. Anti-ganglioside antibody was negative. Motor and sensitive conduction studies showed no peripheral neuropathy distal from the brachial plexus level. However, needle electromyography showed long-duration polyphasic motor unit potentials at nerve root C3–C8 levels, suggesting neurogenic changes. Cervical contrast-enhanced magnetic resonance imaging (MRI) showed mild swelling of the right C6 nerve root, which was considered to be radiculopathy due to VZV.

After a course of steroid pulse therapy (methylprednisolone 1 g/day for 3 days), prednisolone was started at 30 mg/day (tapered over 2.5 months). Rehabilitation was performed at our clinic. During follow-up, muscle weakness in the right upper limb gradually improved. Approximately six months after onset, abduction of the shoulder joint was possible. Approximately two months later, pain and dysesthesia

improved, and pregabalin was discontinued. Although slight asphyxia due to right phrenic nerve palsy remained, it gradually improved. Approximately nine months after onset, chest radiography showed no elevation of the right diaphragm (Figure 2B).

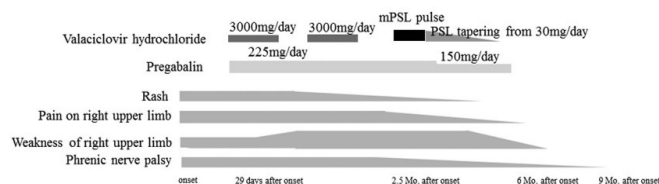


Figure 1: The patient's clinical course.

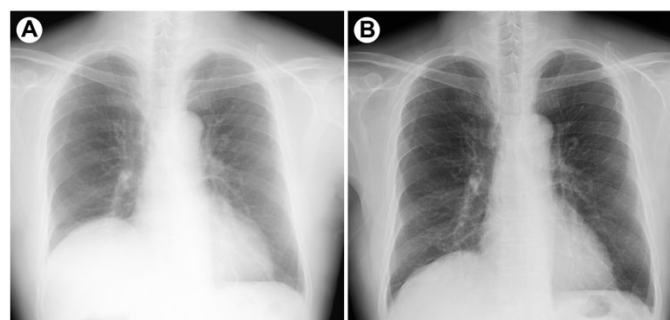


Figure 2: Clinical course of hemidiaphragmatic paralysis in the patient. (A) Chest X-ray during the examination at six months after the onset showed an elevated right diaphragm. (B) Chest X-ray at nine months after the onset showed no elevation of the right diaphragm.

DISCUSSION

In a study of 1393 patients with HZ, Liu et al. [3] reported that SZP occurred in 0.57% of patients with an average age of onset of 69 years. Akiyama [4] and Hung et al. [5] have also evaluated limb weakness in patients with HZ. Akiyama [4] showed that SZP was present in 0.84% of patients with HZ (12/1432 patients). Almost all affected patients were over 60 years of age except for one 43-year-old man with myelodysplastic syndrome. Hung et al. [5] reviewed data of five patients from their hospital and 26 patients from the literature regarding myelitis with HZ from 1980 to 2012. They concluded that 54.8% of patients were immunocompromised. Jones et al. [6] reviewed data of 49 patients with SZP of the limb from Mayo Clinic and reported a mean age of onset of 71 years. They concluded that SZP of the limb was caused by radiculopathy, plexopathy, or peripheral neuropathy. Moreover, paresis typically lasted for at least several months and was associated with high rates of PHN.

As reported previously, SZP of the limbs is a relatively rare complication in mainly older adults or immunocompromised patients. Therefore, it seems necessary to assess the detailed clinical history of each patient.

Herein, we identified a rare case of SZP, an uncommon motor complication caused by HZ. The patient was

determined to have cervical radiculopathy associated with PHN in the same dermatome. He appeared to have recurrent herpes infection for a month without receiving antiviral therapy, as the rash was still present when he visited our clinic 29 days after onset. The patient showed C3–C8 radiculopathy with upper limb paresis and phrenic nerve palsy on the same side.

Diaphragmatic paralysis is commonly caused by surgical or traumatic injuries, malignant neoplasms, and neurodegenerative disorders. However, there have been reports of diaphragmatic paralysis due to HZ virus infection. Oike et al. [8] described a case of an 85-year-old woman in whom hemidiaphragmatic paralysis developed within 19 days after appearance of a typical HZ rash involving the C4–C5 dermatome on the same side. Lin et al. [9] reported a case of upper limb paresis and dyspnea who recovered after antiviral administration three months after onset. In our patient, dyspnea due to hemidiaphragmatic paralysis disappeared approximately nine months after onset.

Our patient showed slow recovery from motor complications for over half a year. Gupta et al. [1] assessed the prognosis of SZP and indicated that 66% of patients achieved complete or almost full recovery within a year. However, 17% of patients experienced permanent weakness of the extremities, usually occurring in muscles, such as the diaphragm, anterior tibial, and hand intrinsic muscles. Liu et al. [3] reported different prognoses in eight cases of SZP: one patient recovered completely three months after onset, two recovered partially within a year, and the other five did not recover after six months and one year of follow-up.

This case showed that SZP appeared commonly in older adults (aged ≥ 75 years), suggesting that aging is a risk factor for SZP. Moreover, there were delays in antiviral therapy initiation. Early administration of antiviral drugs may reduce the incidence and severity of SZP. Mondelli et al. [10] reported that oral treatment with acyclovir reduced peripheral sensory axonopathy due to ganglion damage and reduced the risk of spread to the anterior roots and spinal motoneurons. Currently, the predominant opinion is that regular antiviral therapy within 72 hours after skin rash appearance could substantially reduce pain over time and the risk of other related complications [11]. Given this opinion, we note that our patient was not administered antiviral drugs within 72 hours after onset. Therefore, we consider that delayed treatment initiation may have been related to the comorbidity of SZP. However, further research is warranted.

CONCLUSION

Segmental zoster paresis was found in an old patient (aged ≥ 75 years). The patient slowly recovered almost completely within over half a year, indicating that the prognosis of motor complications was relatively

better than that of PHN. The results also suggested that complications of motor paralysis may be related to delay in antiviral drug initiation. However, further large prospective cohort studies are required.

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Acknowledgments

We would like to thank Dr. Kusunoki S in the Department of Neurology, Faculty of Medicine, Kindai University, for kindly providing the measurement of anti-ganglioside antibodies. We would also like to thank Editage (www.editage.jp) for English language editing.

Author Contributions

Nobuko Shiraiwa – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related

to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Makoto Terada – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Norio Ohkoshi – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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