CASE REPORT

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Postherpetic Neuralgia Mimicking Renal Colic: Zoster Sine Herpete with Segmental Thoracic Motor Paresis

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We describe a case of zoster sine herpete with a motor paresis mimicking a urolithiasis. A 54 year-old woman admitted for her flank pain in an emergency room. The computer tomography (CT) scans and her symptoms were similar to those caused by renal colic. A urology surgeon operated on her for a urolithiasis. However, her flank pain did not improve. She felt the allodynia and burning pain in the left T10-12 dermatomes. She also complained abdominal wall bulging in same lesion. She had no history of skin eruption and no scar of herpes zoster. The abdominal CT scan did not find any pathologic findings. Electromyelography showed denervation potential on some abdominal muscles. Serologic studies indicated that varicella-zoster virus IgG titers were positive. We concluded that this condition was zoster sine herpete. Fortunately, the patient responded well to our empirical therapy, which included gabapentin and epidural steroid injections.

Key Words: herpes zoster, postherpetic neuralgia, segmental paresis, zoster sine herpete.

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Varicella zoster virus (VZV) is a herpes virus that causes chickenpox in childhood. The virus migrates to the dorsal root and then remains dormant in the ganglia. In some patients, VZV may reactivate and spread to the sensory pathways. It can typically produce neuritis with prodromal pain, unilateral rash and vesicular eruptions in a restricted dermatomal distribution [1]. In most patients, a clinical diagnosis of herpes zoster is readily made when a rash appears. However, some patients have herpes zosters without the zosteriform rash (zoster sine herpete, ZSH). ZSH is uncommon and the incidence of ZSH is truly unknown. Moreover, the zoster sine herpete with visible thoracic motor paralysis in segment and involved sensory dermatomes is rare. A few cases of zoster sine herpete with segmental motor paralysis were reported [2].

Besides, it is difficult to accurately diagnose the ZSH and even it can be seriously misdiagnosed as a urolithiasis, appendicitis, biliary or renal colic, cholecystitis, duodenal ulcer. We report a case of ZSH with abdominal wall protrusion misdiagnosed as a ureter stone.

CASE REPORT

A 54-year-old woman presented with squeezing pain, no suspicious-appearing herpes zoster skin lesions. The pain began approximately one month earlier. At that time, she visited an emergency room in other hospital. The computed tomography (CT) scan showed a ureter stone and her pain site were similar to those caused by renal colic. A urology surgeon operated on her, but

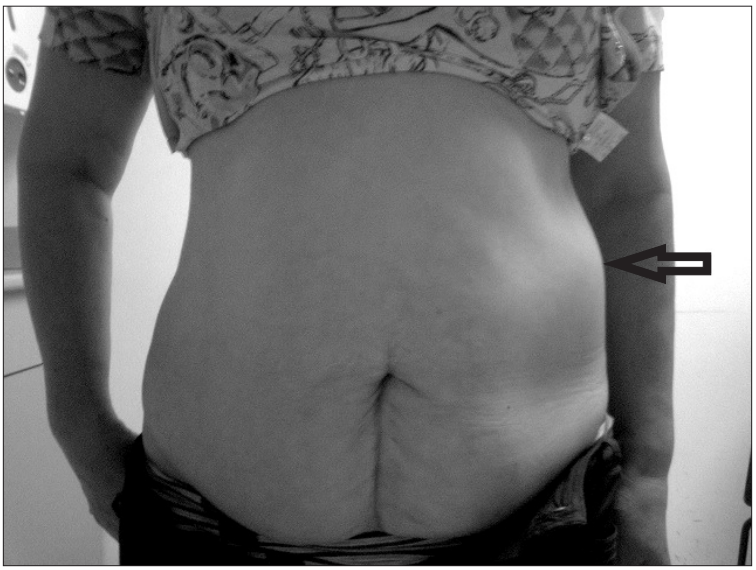

Fig. 1. Frontal view showing left unilateral abdominal swelling.

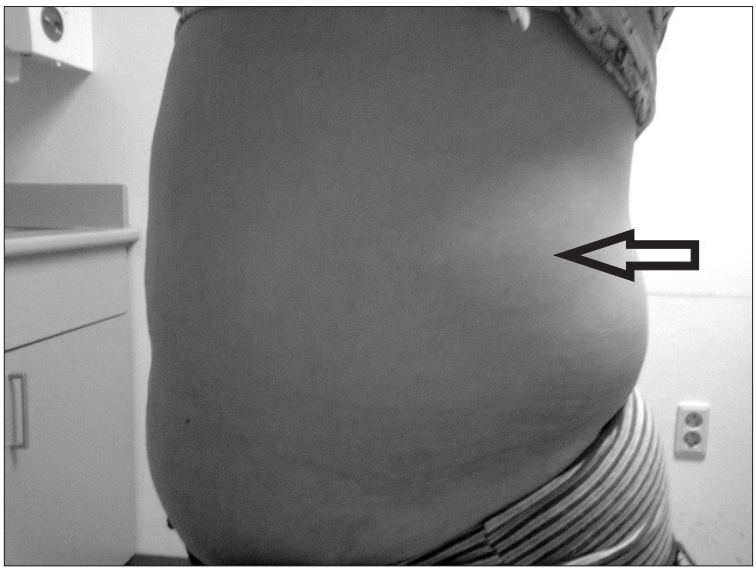

Fig. 2. Lateral view showing abdominal wall without rash.

no stone was found in her ureter, and her symptoms did not improve. An orthopedic surgeon, in local clinic, examined the patient and concluded that the symptoms were caused by intercostal neuralgia and prescribed medicines for the pain. However, when the patient still complained about a burning pain and allodynia, she was referred to our pain clinic one month later.

At the first visit to our clinic, her visual analogue scale (VAS) score was 10. On physical examination, the abdominal wall was showed asymmetric contour, the bulging was noted unilaterally in the left T10-12 dermatomes. No affected site tenderness, rash, vesicles were detected (Figs. 1 and 2). Hyperesthesia and allodynia were noted in the left T10-12 dermatomes.

A follow-up abdominal CT scan did not find any pathologic findings, such as hematomas, herniation or other masses. One month after first visit, we were able to confirm the serologic test. The serologic studies showed that anti-VZV immunoglobulin (Ig) G titers were positive but IgM titers were negative. On electromyelography (EMG), we found mild denervation potential on the left

paraspinal muscles at the T10-T12 levels and moderate denervation potential on the left obliquus externus abdominis muscle.

The pain in a dermatomal distribution was often appeared in DM truncal neuropathy. However, we could exclude DM neuropathy since she had normal values of serum glucose and HbA1C. Our patient was provisionally diagnosed with ZSH with segmental thoracic motor paresis. She was treated with gabapentin and thoracic epidural steroid injections. She was injected 0.5% lidocaine 10 ml with triamcinolone 40mg at the T10-11 level through interlaminar approach. At the follow-up visit, her VAS score decreased to 7. She was injected epidural blocks with 10ml of 0.5% lidocaine at T10-11 level twice, and underwent transforaminal epidural block with 2ml of 0.5% lidocaine with dexamethasone 1.6 mg at T11 level. Approximately two months later, her pain almost resolved. She reported that VAS score became 3 and the remaining pain was easily tolerable. A physical examination a month later revealed that her thoracic motor paresis still remained.

DISCUSSION

VZV reactivation can typically produce neuritis with prodromal pain, unilateral rash, and vesicular eruptions in a restricted dermatomal distribution [1]. Unilateral, band-like skin lesions are unique clinical findings of herpes zoster infection. A clinical diagnosis of herpes zoster is readily made when a rash appears; however, some patients have herpes zosters without zosteriform rash, termed "ZSH". Lewis [3] reviewed 120 patients who were misdiagnosed and inappropriately treated for ZSH, and reported that there were more patients suffering from ZSH than were recorded. A few cases of ZSH have been confirmed by tests. Gilden et al. [4] previously reported on two men who experienced years of radicular pain without rash. Therein, PCR-amplifiable VZV DNA was found in cerebrospinal fluid 5 and 8 months after onset of radicular pain. Subsequently, both patients were treated with IV acyclovir and the radicular pain was improved. Yeo et al. [5] examined 10 patients who presented with segmental intercostal neuralgia, but had no zosteriform skin lesions. After assaying anti-VZV IgM and IgG, a positive result was discovered for both VZV antibody tests. Treatments with amitriptyline, gabapentin, and epidural steroid injection alleviated their intercostal pain.

Motor neuropathy is rare (5% to 15%) and difficult to detect [1]. The pathogenesis of motor paresis is still controversial. Hanakawa et al. [6] reported that the inflammatory response is caused by direct spread of VZV from sensory nerves to motor compartments. Haanpaa et al. [7] discovered that widespread subclinical motor involvement is relatively more common than previously reported; abnormal EMG reactions were detected in 53% of patients with acute herpes zoster.

Approximately 52% of cases involve the thoracic dermatomes, 20% involve the cervical region, 17% involve the trigeminal nerve, and 11% involve the lumbosacral region [8]. Because of rich innervation, the thoracic segments are most commonly affected. When herpetic rashes appear between the T2 and L1 dermatomes, the incidence of clinically manifested motor paresis is about 0.3% [9]. In contrast, 35% of patients affected at the thoracic segment exhibit positive findings on electromyographic evaluation [10]. Due to overlap in the innervation of the trunk muscles, detection of thoracic muscle paresis is difficult. Motor paresis usually develops within 2 weeks of a herpetic rash. The prognosis of herpetic motor paresis is good. Complete or nearly complete recoveries have been reported in 67% of patients. About 9% patients exhibit significant improvements in 1-2 years [11].

Laboratory diagnosis is not necessary for the clinical management of most VZV infections; nevertheless, rapid diagnostic techniques are useful in guiding decisions on antiviral treatment. In the convalescent period, VZV IgM antibody titers are generally negative. Furthermore, depending on the time, site of specimen collection, and assay method, IgM antibody results can differ. Assays for VZV IgG antibodies are most useful for determining the immune status of patients, but do not indicate whether the patient has a primary or recurrent VZV infection [12]. Furuta et al. [13] reported that PCR-amplifiable VZV DNA is a useful test for the diagnosis of ZSH; meanwhile, there were patients with chronic active VZV infection who had no PCR-amplifiable VZV DNA. Bulmental et al. [14] reported a patient who developed cervical radiculopathy without rash, and in whom ZSH was confirmed by detection of intrathecal synthesis of anti-VZV IgG antibody. In this case, PCR-amplifiable VZV DNA in CSF was negative. They found that the serum/CSF ratio

of anti-VZV IgG antibody was lower than the ratios for albumin and total IgG. Nagel et al. [15] reported that 6 of 14 patients who were diagnosed with VZV vasculopathy had no antecedent zoster rash. In the present study, all of the patients' serum/CSF ratios were reduced; however, 4/6 patients exhibited negative results of PCR-amplifiable VZV DNA in CSF. Thus, the diagnostic value of detecting intrathecal synthesis of anti-VZV IgG antibody was greater than PCR-amplifiable VZV DNA.

In our case, the initial diagnosis was a ureter stone, because her symptoms and pain site were similar to those of renal colic and the initial computed tomography scanning at other hospital showed urolithiasis. She was operated on, but the surgeon was not able to find a stone. Although rare, CT scans can be misleading. There is also the possibility that the stone was spontaneously passed prior to the surgery. However, the patient's symptoms did not improve after surgery. The second diagnosis was intercostal neuralgia. Accordingly, the patient was prescribed only analgesics instead of antiviral agents. Antiviral therapy is effective for suppression of viral replication; studies have shown reduced neural damage and fewer complications when therapy is started within 72 hours of the onset of rash and continued for 7-10 days [8]. If the patient described here had begun antiviral therapy immediately upon visitation to the emergency room for her flank pain, she may not have had any remaining pain and motor paresis. The diagnosis of herpes zoster infection tends to depend on symptoms and signs (vesicular rash and neuritis). However, prodromal pain without rash at the thoracic level can mimic the pain of nephrolithiasis, urolithiasis, appendicitis, biliary or renal colic, cholecystitis, duodenal ulcer, and myocardial infarction. Furthermore, cases of motor paresis without zosteriform rash are rare, and may be confused with other etiologies (abdominal mass, herniation). This condition can lead to serious misdiagnoses among physicians who are unused to diagnosing neuropathic pain. Although we did not confirm the VZV infection by serologic test, we thought that this negative result was due to a delay in the test. Because of her clinical symptoms and signs, we strongly suspected ZSH. DM truncal neuropathy can produce segmental neuralgia just like that of ZSH; however, we could exclude DM truncal neuropathy based on the patient's HbA1C and serum glucose levels. Nevertheless, we felt that anti-VZV IgG antibody testing of CSF was helpful in confirming VZV reactivation in our patient [15].

The true incidence of ZSH is not definitively known, and we suspect that the incidence of ZSH is greater than what has been reported. ZSH should be considered in patients with a history of segmental neuropathy without zosteriform skin lesion. Early detection of ZSH is very important since rapid apply of antiviral therapy reduces neuropathic pain and complications. PCR for VZV DNA and anti-VZV IgG in CSF studies are helpful to establishing an appropriate diagnosis of ZSH.

REFERENCES

1. Scott M. Fishman JCB, James P. Rathmell: Bonica's management of pain. 4th ed. Philadelphia, Lippincott Williams & Wikins. 2010, pp 338.
2. McAllister RK, Borum SE, Mitchell DT, Bittenbinder TM: Thoracic motor paralysis secondary to zoster sine herpete. Anesthesiology 2002; 97: 1009-11.
3. Lewis GW: Zoster sine herpete. Br Med J 1958; 2: 418-21.
4. Gilden DH, Wright RR, Schneck SA, Gwaltney JM Jr, Mahalingam R: Zoster sine herpete, a clinical variant. Ann Neurol 1994; 35: 530-3.
5. Yeo JS, Sim WS, Kim YC: Herpes sine zoster: is the cause for the segmental intercostal neuralgia of unknown cause? Korean J Pain 2005; 18: 226-8.
6. Hanakawa T, Hashimoto S, Kawamura J, Nakamura M, Suenaga T, Matsuo M: Magnetic resonance imaging in a patient with segmental zoster paresis. Neurology 1997; 49: 631-2.
7. Haanpaa M, Hakkinen V, Nurmikko T: Motor involvement in acute herpes zoster. Muscle Nerve 1997; 20: 1433-8.
8. Waldman SD: Acute herpes zoster. 2nd ed. Philadelphia, WB Saunders. 2002, pp 171.

9. Healy C, McGreal G, Lenehan B, McDermott EW, Murphy JJ: Self-limiting abdominal wall herniation and constipation following herpes zoster infection. Q J Med 1998; 91: 788-9.
10. Cioni R, Giannini F, Passero S, Paradiso C, Rossi S, Fimiani M, et al: An electromyographic evaluation of motor complications in thoracic herpes zoster. Electromyogr Clin Neurophysiol 1994; 34: 125-8.
11. Gupta SK, Helal BH, Kiely P: The prognosis in zoster paralysis. J Bone Joint Surg Br 1969; 51: 593-603.
12. Krah DL: Assays for antibodies to varicella-zoster virus. Infect Dis Clin North Am 1996; 10: 507-27.
13. Furuta Y, Fukuda S, Suzuki S, Takasu T, Inuyama Y, Nagashima K: Detection of varicella-zoster virus DNA in patients with acute peripheral facial palsy by the polymerase chain reaction, and its use for early diagnosis of zoster sine herpete. J Med Virol 1997; 52: 316-9.
14. Blumenthal DT, Shacham-Shmueli E, Bokstein F, Schmid DS, Cohrs RJ, Nagel MA, et al: Zoster sine herpete: virologic verification by detection of anti-VZV IgG antibody in CSF. Neurology 2011; 76: 484-5.
15. Nagel MA, Forghani B, Mahalingam R, Wellish MC, Cohrs RJ, Russman AN, et al: The value of detecting anti-VZV IgG antibody in CSF to diagnose VZV vasculopathy. Neurology 2007; 68: 1069-73.