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Case Report

Sacral Herpes Zoster in Immunocompetent Patient Followed by Dysfunction of Voiding Reflexes of the Bowel and Bladder. A Rare Elsberg Syndrome that Needs to be Differential Diagnosed

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Abstract

Herpes zoster (HZ) is a neurologic skin infection caused by reactivation of varicella-zoster virus (VZV) latent in the human body. VZV establishes an incubation period in ganglia and when activated it may influence the micturition and the defecation.

Keywords: Herpes Zoster (HZ); Urinary Retention

Introduction

Urinary retention and constipation are probable related to the following pathophysiological mechanisms: Viral meningitis radiculitis, or both are regarded as causing sacral motor and sensory neuropathy with dysautonomia, inducing sphincter dysfunction. VZV invades the sacral dorsal root ganglia of the spinal cord, the nerve roots, and the peripheral nerves undergoing neuroinflammatory changes, which cause disruption of the voiding reflexes; VZV directly invades bladder inner wall nerves leading to inflammatory lesions of the bladder, which ultimately triggers low contractility of the detrusor muscle; the perineal nerves and pelvic plexus nerves are damaged due to the destruction of VZV, which breaks the coordination of the functions of the detrusor muscle, the internal bladder sphincter, and the urethral sphincter, and affects the urinary excretion. [1,2]. According to the literature, VZV of the sacral roots causes a consistent syndrome which it may cause retention with sensory loss of both bladder and rectum and flaccid paralysis of the detrusor [3].

Voiding dysfunction caused by herpes zoster was first described in 1890 [4], and since then more than 200 cases have been reported in the literature. The herpes zoster infections in the sacral region accounting for 8% of all herpes zoster infections, and it was found that 4.02% of these voiding dysfunctions were associated with VZV infection [2,5]. In 1931, Elsberg described a syndrome characterized by rapidly developing sacral root deficits with sphincter dysfunction. In cerebrospinal fluid (CSF), pleocytosis and elevated protein are often observed [4]. Most probably, the basis of this condition is localized radiculitis of the sacral roots. Elsberg syndrome (ES) is an unidentified cause of acute lumbosacral neuritis and is related to recent herpes virus infection. ES often presents with urinary retention and/ or bowel dysfunction. However, there are very few case reports of this condition, and it remains understudied. It is commoner to immunocompromised patients [6–8]. However, existing diagnostic criteria help to identify probable cases (see Table 1).

We present the case of a76 year-old immunocompetent female patient, who suffered a recent VZV infection of sacral distribution with micturition and the defecation dysfunction.

Case Report

A 76-year-old female patient of Caucasian origin, immunocompetent, was hospitalized In Euromedica General Clinic of Thessaloniki due to urinary and stool retention starting 10 days ago.

According to her previous medical history, she has hypothyroidism and hyperlipidemia, and she is treated accordingly with Levothyroxine 112μgr and simvastatin/ezetimibe.

Diagnostic criteria for Elsberg syndrome (Savoldi's criteria) and assessment in the present case [3]		
Categories 1. Laboratory-supported definite 2. Clinically definite 3. Clinically probable 4. Clinically possible 5. Excluded	Criteria (A1 OR A2) AND B5 A1 OR A2; B1 AND two of B2–B4; B1 and B2 (if concomitant) A1 OR A2; B1 AND one of B2–B4 A1 OR A2; one of B1–B4 Neither of A1 nor of A2; any of D1–D3	Assessment in the present case ✓
A. Required		
A1. Clinical symptoms and signs of cauda equina involvement: urinary hesitancy or retention; bowel incontinence, or severe constipation (erectile dysfunction insufficient on its own)		✓
A2. MRI or electrophysiologic evidence of cauda equina involvement: enhancement of cauda equina; EMG evidence of radiculopathy		
B. Supportive but not required		
B1. Time course: acute/subacute onset; no relapse; progression over <3 months		✓
B2. Coexisting or recently preceding symptoms of genital herpes infection OR other clinical symptoms of herpes virus infection.		✓
B3. Clinical (e.g., exaggerated reflexes and Babinski signs) or MRI evidence of myelitis in conus		
B4. CSF pleocytosis		N/A
B5. Documented herpes virus infection from CSF by PCR, culture, or detection of IgM serology		✓
C. Red flags		
C1. Relapses beyond 1 year from onset		N/A
D. Exclusionary		
D1. Myelitis extending rostral to T9		No
D2. Other neurologic symptoms suggestive of alternative etiology: optic neuritis, brain/brainstem syndrome		No
D3. Other etiology proven/more likely for syndrome: NMOSD, dural arteriovenous fistula, viral transverse myelitis, other causes of myelopathy		No

Table 1

Fifteen days ago, she felt excruciating pain on the left side of her lower waist to her left glute. During the shower, she sensed that her skin was not only sensitive, but it had also lesions that were later diagnosed as a herpes infection. She was prescribed 11 days of Brivudine 125mg QD. During her hospitalization, the patient was dermatologically and gynecologically investigated, with no infection of the genitalia. While the skin marks where still present (see Figure 1A) in S2-S5 distribution. Even though the skin lesions subsided, the neuralgic pain and the sphincter disturbance remained. The sense of need to urinate and defecate were totally diminished.

Her abdominal examination detected present bowel sounds but the abdominal CT revealed faces content in the colon.

She needed a urinary catheter placement for more than 15 days that was placed before her hospitalization. With the gradual symptom improvement, the catheter was removed almost 3 weeks post the symptoms initiation after ultrasound confirmation of no residue post-urination. However, the stool retention remained longer and still needed daily laxatives (macrogol), three months post the symptom initiation.

Her neurological examination was unremarkable, and no signs of myelitis were present. The only concerning finding was the sphincter dysfunction. However, an MRI of Lumbar and Sacral Spine was performed to investigate any active inflammatory lesion on the conus medullaris and affected roots according to the skin lesions (I1-I5) (see Figure 1B). No enhancement or swelling was detected.

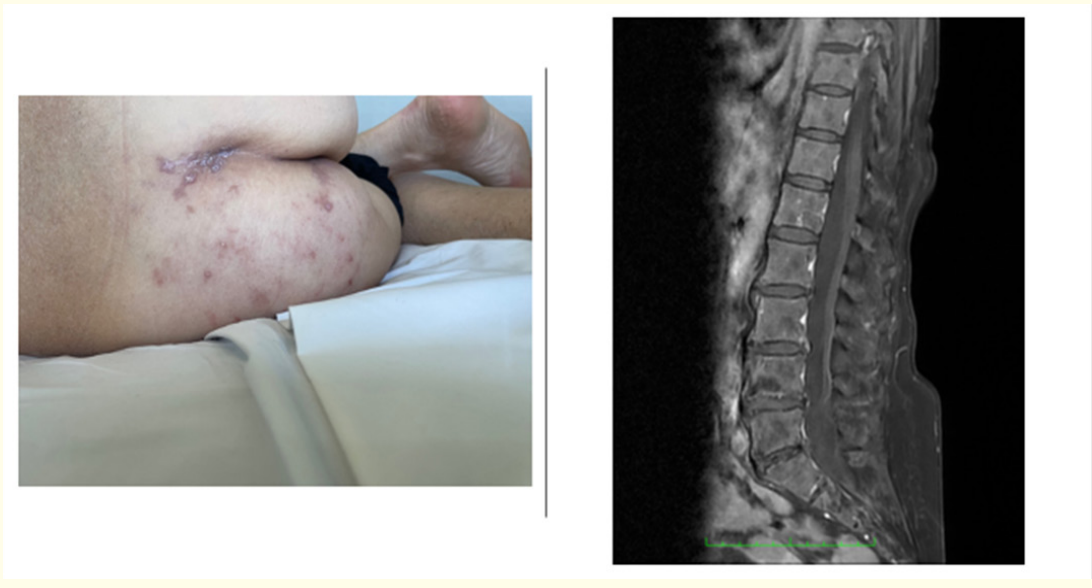

Figure 1: A. Skin affected area of S1-S5 distribution by VZV. B. Lumbar-sacral Sagittal MRI T1 FSE, Flex FS+ IV contrast without any enhancing lesions.

The serological testing for Herpes Simplex Virus (HSV)1&2 and Varicella zoster Virus (VZV), revealed that the patient was infected with VZV (IgG 2067 IU/ml, IgM 1,82 IU/ml with negative indexes <150 IU/ml and <1.0 IU/ml accordingly).

The pain was treated with pregabalin, with gradual titration starting from low doses and close monitoring for side effects. It was still present and intense at three-month follow-up.

Discussion

Urinary retention may precede the appearance of skin rash, and ES should be considered in the differential diagnosis. Differential diagnosis between herpes virus type 1/2 (HSV-1/2) and varicella zoster virus (VZV) is sometimes necessary because the recommended antiviral treatment doses differ between the herpes types [9–11]. ES is an infectious syndrome manifested by various signs of acute lumbosacral nerve root myelitis (e.g., urinary retention and lumbosacral sensory symptoms), and the viral etiology of the disease has been established [2]. Depending on the level of the infection the symptoms can vary. Most of patients present a complete recovery with or without sequelae over a mean interval of 6 to 10 weeks, except for severe myelitis cases. VZV mostly present in immunocompromised patients [6–8] however, there are cases in immunocompetent people as well. In our case, the patient was not immunocompromised, the pain and the rash showed first, and the urine retention and the abdominal distention followed causing her hospitalization ten days after the symptom's onset.

Despite the antiviral treatment she received early in the disease process, only the rash subsided. The neurological dysfunction of bladder and bowel had slower improvement, and the neuralgic pain remained intense at three-month follow up, despite the pharmacological treatment. Acupuncture has been used as a safe and promising complementary treatment option for ES [2]. The diagnosis of Elsberg syndrome of our case was clinically, and laboratory supported definite. However, the syndrome is not well-recognized among clinicians and often is misdiagnosed.

Conclusion

Special challenge is the timing of the rash, which can be absent, or it can follow the neurological deficit of bladder and bowel. Serological viral examination should be performed in all patients with sudden urine retention and abdominal distention, especially in immunocompromised patients. Urinary retention is a rare complication of herpes zoster. It can occur in any site of infection but is more frequent in the case of lumbosacral lesions.

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