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Case Report

Case report: a rare case of Herpes zoster Ophthalmicus with simultaneous impairment of lumbar and sacral dermatomes

Herpes Zoster Oftálmico e Lombossacral: relato de caso raro de acometimento simultâneo

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ABSTRACT: Herpes zoster (HZ) occurs as a result of the reactivation of varicella-zoster virus and is characterized by neuralgic pain, constitutive symptoms, pruritus and dysesthesia along nerve pathways. Vesicles on an erythematous base in a dermatome can be identified, and vision loss may occur in cases of ocular involvement. HZ is clinically diagnosed and treated with antivirals. This study aims to report a rare case of HZ simultaneously affecting the ophthalmic branch of the trigeminal nerve and the lumbar and sacral dermatomes. The patient was clinically cured after treatment with acyclovir for 7 days and did not present other complications.

KEY WORDS: Herpes Zoster; Human Herpesvirus 3; Immunocompromised host; Shingles; Varicella Zoster; Herpes Zoster Ophthalmicus.

RESUMO: O Herpes Zoster (HZ) ocorre como resultado da reativação do vírus varicela-zóster e é caracterizado por dor neuropática, sintomas constitutivos, prurido e disestesia ao longo das vias nervosas. Vesículas em base eritematosa em um dermátomo podem ser identificadas, e a perda de visão pode ocorrer em casos de envolvimento ocular. HZ é diagnosticado clinicamente e tratado com antivirais. Este estudo tem como objetivo relatar um caso raro de HZ acometendo simultaneamente o ramo oftálmico do nervo trigêmeo e os dermátomos lombares e sacrais. O paciente apresentou cura clínica após tratamento com aciclovir por 7 dias e não manifestou outras complicações.

PALAVRAS-CHAVE: Herpes Zoster; Herpesvirus Humano 3; Hospedeiro imunocomprometido; Infecção pelo Vírus da Varicela-Zoster; Herpes Zoster Oftálmico.

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INTRODUCTION

Herpes Zoster (HZ) occurs following the reactivation of varicella-zoster virus (VZV), also known as human herpesvirus type 3 (HHV3), which typically remains latent for decades in sensory ganglia cells after primary infection[1,2]. Its lifetime incidence is 10% to 20% and the risk increases with age: it almost doubles every decade from the age of 50[3], probably because of immunosenescence[2]. The average incidence of HZ in immunocompromised adults exceeds the expected HZ incidence in healthy adults ≥60 years [4]. The varicella-zoster virus is transmitted to individuals by direct contact with contents of skin vesicles and respiratory tract secretions [3,5,6], by inhalation of aerosol particles. The disease manifests as prodromal neuropathic pain in a unilateral dermatomal distribution, which typically occurs at least two days before the onset of skin lesions and constitutive symptoms as headaches, general malaise, and photophobia. The acute eruptive phase is characterized by the maintenance of these nonspecific symptoms and dermatological involvement: painful vesicles that follow a dermatome and appear in a centripetal pattern. Vesicles usually involute within 2-4 weeks; however, the pain may continue with an average duration of 45 days. The persistence of pain is more common in elderly and immunocompromised people[6,7].

Special clinical presentations can occur[8]. Herpes Zoster Ophthalmicus (HZO) typically happens due to the involvement of the ophthalmic branch of the trigeminal nerve. The Hutchinson's sign, described as vesicles on the nose tip, is a predictor of this ocular involvement[5]. HZO can lead to vision loss, debilitating pain and acute retinal necrosis[6,8]. Ramsay Hunt syndrome occurs if the virus affects the geniculate ganglion and facial nerve. Typical manifestations include vesicles in the auditory canal, unilateral facial paralysis, and earache[6,8]. Simultaneous affection of the vestibulocochlear nerve (Herpes Zoster Oticus) or trigeminal nerve is frequently observed. This can lead to manifestations such as facial pain, dizziness, tinnitus or hearing impairment (HZ Oticus)[8]. The central nervous system can also be involved resulting in nerve palsies, muscular weakness, diaphragmatic paralysis, neurogenic bladder, Guillain Barré syndrome and myelitis[6].

Postherpetic neuralgia (PHN) is the commonest complication seen in elderly. Other reported complications include renal, gastrointestinal, and vasculitis, which are associated with increased morbidity and mortality[6,8].

The diagnosis is based on clinical history and physical examination and can be confirmed by laboratory tests in case of uncertainty[8]. Systemic antivirals, including acyclovir and valacyclovir are considered first-line treatment. Other nucleoside analogues such as penciclovir, its prodrug famciclovir, and brivudine can also be used [9].

We report a rare case of HZO confirmed by ophthalmologic evaluation, with concomitant involvement of lumbar (L1-L3) and sacral (S1-S2) dermatomes identified by clinical examination. This case is unusual due to the concurrent impairment of multiple non-contiguous dermatomes.

CASE REPORT

A 60-year-old man presented with palpebral edema, erythema, and crusts in the periocular region, on the left of his forehead, and in the upper third of the left hemiface without crossing the midline (Figure 1).

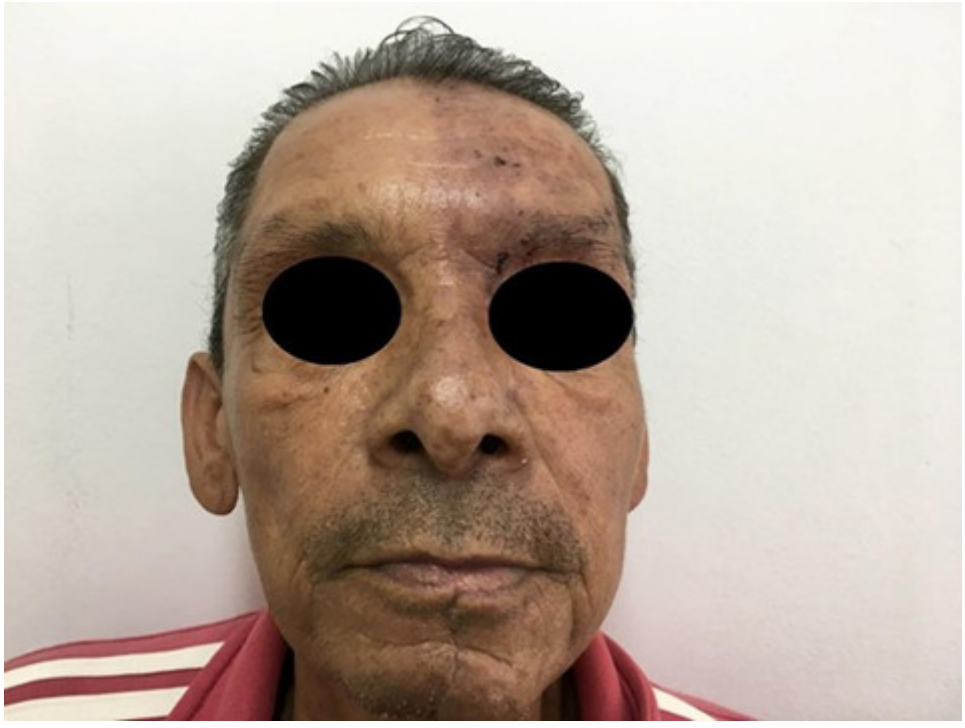

Figure 1 - Herpes zoster Ophtalmicus before treatment.

He also had vesicles and pain in the left lower limb (Figures 2) and flank (Figure 3).

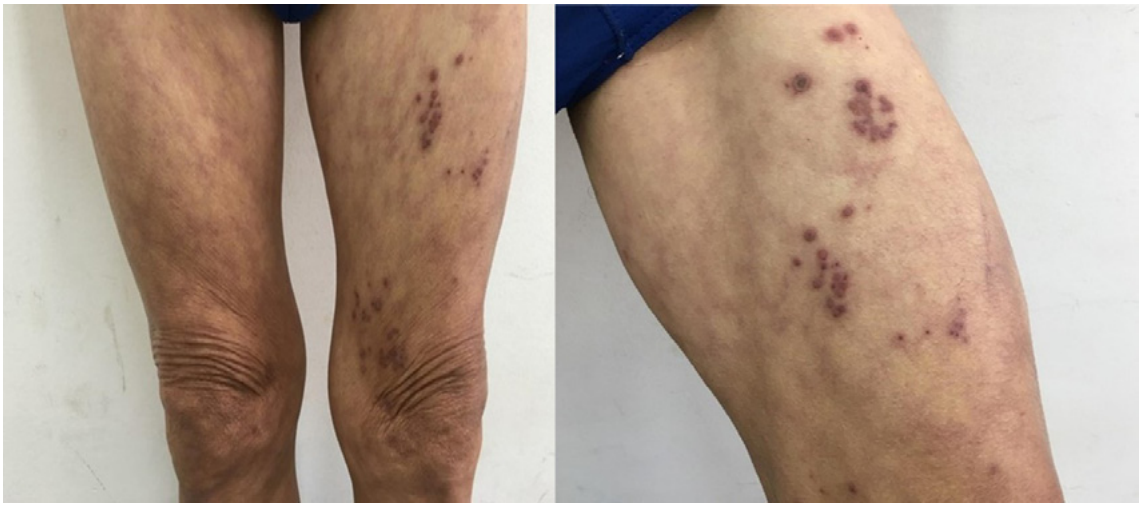

Figure 2 - Herpes zoster affected dermatome unilaterally, along the lumbar nerves (L1–L3).

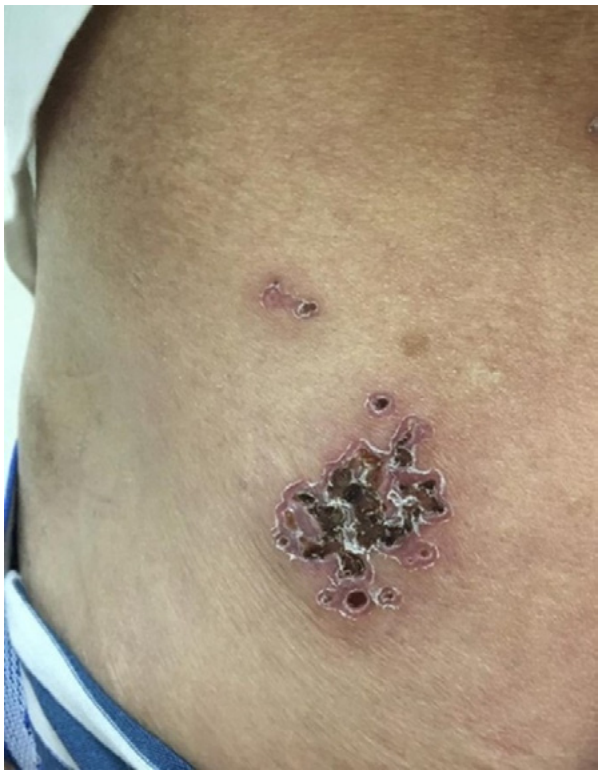

Figure 3 - Herpes zoster in the sacral nerves (S1–S2) during the healing phase.

Changes in facial mimetics, vesicles on the nose tip or ear canal and pupillary alterations were not observed. In another service he was mistreated with topical ketoconazole and betamethasone and did not exhibit clinical improvement. HZ in the lumbar (L1-L3) and sacral dermatomes (S1-S2) and HZ Ophthalmicus were clinically diagnosed. Upon reviewing the patient's medical history, it was ascertained that five years prior, he had been diagnosed with well-differentiated squamous cell carcinoma of the palate, previously treated with surgical excision, 20 sessions of radiotherapy and 6 cycles of chemotherapy in 2012. He has been considered cured since then and the tumor did not recur. He also had a history of alcohol abuse and smoking for 20 years, nonetheless, he has ceased both.

The serology for hepatitis B, hepatitis C and HIV was negative. Analyses of the blood cells count (hemoglobin 18.7 g/dL, mean corpuscular volume: 118 μm^3, white blood cells: 9,000/mm3, platelet: 438,000/mm3) and clinical chemistry of alanine aminotransferase (ALT = 46 mU/mL), aspartate aminotransferase (AST = 69 mU/mL), urea (14 mg/dL) and creatinine (0.4 mg/dL) were performed. Haemoglobin and AST levels were elevated, probably due to smoking and alcohol consumption or hepatitis due to VZV. Treatment was initiated with acyclovir (800 mg five times a day, during 10 days), opioid analgesia, neuromodulation of pain with amitriptyline, and the symptoms vanished. He did not report any adverse effect due to the use of acyclovir. The patient was referred for ophthalmologic evaluation and presented normal tests.

DISCUSSION

In childhood infection with VZV often results in Varicella (Chickenpox). Afterwards, the virus reactivation of endogenous latent VZV infection within the sensory ganglia leads to HZ or Shingles. The VZV is predominantly regulated by cell-mediated immunity, and reactivation is viewed as an outcome of the breakdown of this immune surveillance[8]. The elevated incidence of Herpes Zoster (HZ) in older individuals is believed to result from this decline in cell-mediated immunity, a natural age-related process[3]. It manifests as neuralgic pain, and as vesicles grouped on an erythematous base in a sensory dermatome corresponding to the affected nerve. The vesicles commonly rupture, subsequently ulcerate, and eventually form a crust. Patients remain highly contagious during this stage until the lesions have fully dried out[6]. Typically, HZ is unilateral[7] and the

dermatomes most frequently affected are those of the thoracic region, followed by the face, and the cervical, and lumbosacral regions[10]. Disseminated disease, defined as more than twenty skin lesions developing outside the primarily affected area or dermatomes directly adjacent to it, consists in 1% of the cases[10]. The Systematic review performed by Susannah L Mckay et al. (2020) analyzed the risk of HZ and associated complications in immunocompromised patients, including for analysis adult patients with hematopoietic cell transplant (HCT), cancer, human immunodeficiency virus (HIV), and solid organ transplant. It indicated that most common disseminated HZ were described in adult patients with HCT[4].

When the shingles appear between the T2 and L1 segments, the incidence of clinically detectable segmental paresis is 0.3% in the myotome corresponding to the dermatome distribution of the rash[1]. The patient presented simultaneous damage of the ophthalmic branch of the trigeminal nerve and also non-contiguous dermatomes: lumbar (L1-L3) and sacral (S1-S2); this condition is considered rare[10]. Ayisi-Boateng et al. (2021) cited the involvement of two distant dermatomes, the ophthalmic branch of the trigeminal and L1, in a woman living with HIV. She presented Orbital Apex Syndrome, a serious sequelae of the disease, that is a condition characterized by the impairment of cranial nerves II, III, IV, V, and VI, leading to complete internal and external ophthalmoplegia[11].

VZV reactivation at more than one site may occur because of the presence of factors such as previous tumors and chemotherapy/radiotherapy[4], which in the present case constituted an immunosuppressive environment in an elderly patient with a history of alcohol abuse and smoking. In 2020 Steinberg *et al.* reported a case of an elder man with a past of radiation therapy presenting disseminated HZ predominantly along the L3-S2 dermatomes with lumbosacral plexopathy[10]. Herpes Zoster Ophthalmicus is a cause for concern because of the risk of retinal vascular occlusion, ulceration, necrosis and vision loss[12]. Disseminated HZ may complicate in secondary bacterial infection, hepatitis or central nervous system involvement resulting in meningitis, encephalitis with a risk of death, Guillain Barré Syndrome and acute retinal necrosis[6].

The diagnosis is based on clinical history and physical examination, and laboratory tests such as Polymerase Chain Reaction (PCR), direct fluorescent antibody (DFA) and viral cultures[5,6].

Treatment should be performed with antivirals, preferably within 72 h of the onset of cutaneous eruptions and for all immunocompromised patients[10]. Treatment lessens the severity and duration of pain associated with acute neuritis, promotes more rapid healing of skin lesions, prevents new lesion formation, decreases viral shedding to reduce the risk of transmission, and prevents postherpetic neuralgia (PHN)[3,6] The drugs of choice are acyclovir, famciclovir and valacyclovir[4]. These three drugs reduce the duration of pain, presumably by limiting sensory nerve damage resulting from viral replication[5,9]. Valacyclovir demonstrated superiority over acyclovir in the management of postherpetic neuralgia in various clinical studies. Other drugs are brivudine and amenamevir[9]. Valnivudine and Valomaciclovir are new antiviral drugs under development for treatment of HZ. Both had shown to be effective, well-tolerated, with a once-daily therapy regimen[9]. The patient in this study had no signs of clinical deterioration despite the simultaneous impairment of more than one dermatome.

Treatment may include anticonvulsants (gabapentin) and tricyclic antidepressants in cases involving moderate to severe pain[6]. Treatment for acute neuritis may include corticosteroids[5]. The association of opioid and tricyclic antidepressant was effective in the patient in this study.

Postherpetic neuralgia is the main complication of HZ[9] and it is defined as persistent pain for more than 3 months after rash onset, with an estimated incidence between 9% and 14% one month after the HZ eruption. The most firmly established risk factor for this complication is advancing age. As individuals age, both the risk and duration of postherpetic neuralgia increase. Also, an immunocompromised state is considered a risk factor for the development of PHN[3]. It is palliatively treated with topical capsaicin and tricyclic antidepressants. A physical alternative is transcutaneous neural electrostimulation (TENS)[8,9]. A Cochrane review carried out in 2013 has not shown evidence corroborating the use of corticosteroids to prevent the onset of postherpetic neuralgia[3].

Special antiviral prophylaxis with valacyclovir or acyclovir is important to prevent VZV reactivation in some immunocompromised patients, as HCT recipients and patients undergoing T-Cell suppressive therapies. The duration of antiviral prophylaxis has appeared to modify the risk of HZ: the longer it lasts, the better the outcomes are[4].

A live-attenuated Herpes Zoster vaccine and a recombinant adjuvanted vaccine are available for immunocompetent adults older than 50 years old and both had proven to prevent HZ and the development of PHN[7,8]. The recombinant HZV vaccine is non-replicative, making it a safe option for individuals with compromised immune systems as well[8]. The patient in the present study did not present postherpetic neuralgia and had not been vaccinated.

Cases of coronavirus disease 2019 (COVID-19) and HZ co-infections are being currently reported. The acute disease, the usage of corticosteroids and COVID-19 vaccination are believed to reactivate the virus[8,9]. The Centers for Disease Control and Prevention (CDC) recommend vaccination against HZ for individuals at high risk, in particular for patients ≥ 50 years old and immunosuppressed patients ≥ 18 years old[9].

CONCLUSION

We describe a rare case of HZO confirmed by ophthalmologic evaluation, with concomitant involvement of lumbar (L1-L3) and sacral (S1-S2) dermatomes identified via clinical examination. This case is rare because of the simultaneous impairment of more than one dermatome, and the affected dermatomes were non-contiguous. The medical history of the patient, including alcohol abuse, old age, and previous tumor, may have contributed to this rare manifestation of HZ. The patient was clinically cured following treatment with acyclovir for 10 days and did not present further complications.

Contribution: Mônica de Albuquerque Costa, Maria Cristina Vieira de Andrade, Erika Santos Freire: Case report Writing. Isabella Boa Sorte Costa: Literature review. text review, formatting. Daniela de Oliveira Werneck Rodrigues, Nádia Rezende Barbosa Raposo: Approval of the final version of the manuscript.

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