# Unique presentation and clinical course of shingles: A case report

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## Abstract

We report the unique presentation of a Varicella Zoster Virus reoccurrence, commonly referred to as shingles, in a patient who developed three concurrent but different manifestations: 1. A cutaneous rash in a cervical dermatome; 2. A rash-free reoccurrence of a previous Post Herpetic Neuralgia in a different dermatome, and; 3. A rash-free, new-onset of Bell's palsy. Literature searches did not produce any reported cases with this triad of concomitant events. The patient is a 58-year-old immunocompetent Hispanic man whose only risk factor is age greater than 50 years. He had been treated with immunosuppressants in the past for Inflammatory Bowel Disease. Immunosuppression was stopped eight years ago after undergoing a total colectomy as definitive treatment. Now immunocompetent, he develops shingles for the second time in his life, with a very unusual presentation and disease course. The presentation and course of his disease promoted the use of empiric pharmacologic treatment courses, including three courses of oral corticosteroids, for which no current guidelines exist. It is hoped that the observations drawn from the clinical course of this patient may shed light into the better understanding and treatment of Varicella Zoster Virus and Post Herpetic Neuralgia. We also discuss the possibility that each of these individual manifestations may constitute separate disease processes, requiring individual and unique evaluation and treatment. Keywords: Varicella Zoster Virus, Shingles, Post-Herpetic Neuralgia, Zoster Sine Herpete, Bell's palsy

Varicella Zoster Virus (VZV) reoccurrences, also known as shingles, and Post-Herpetic Neuralgia (PHN), are conditions that remain poorly understood and difficult to treat, even by qualified experts in pain management. Treatment options remain inadequate, particularly for PHN, and the disease can have devastating effects on patients' quality of life. About one in four people will develop a VZV outbreak in their lifetime, with this risk increasing after the age of 50. Up to 20% of patients with VZV develop PHN, manifested as moderate-tosevere chronic dermatomal pain persisting for months or years after the acute phase, often causing depression, anxiety and sleep disturbances. Some may even become suicidal [1]. VZV outbreaks in immunocompetent hosts, while uncommon, are known to recur [2].

Reactivation of shingles in a previously affected dermatome after a traumatic injury has been reported [3]. There are also cases of VZV dermatomal pain developing without a prior skin rash, referred to as Zoster Sine Herpete (ZSH) [4]. Reactivation of latent VZV usually affects one dermatome, at times affecting adjacent ones, while disseminated herpes may affect large portions of the body [5]. Risk factors for reactivation include advanced age, diabetes mellitus, malignancy, immunodeficiency, and immunosuppression [5]. The role played by distant immunosuppression, if any, remains poorly understood. The main predictive risk factor for developing PHN after VZV is advanced age, although the reason remains poorly understood. Other risk factors include pain preceding the rash, severe pain in the acute phase of infection, severe and

prolonged blistering, sensory loss, skin ulceration and necrosis [6, 7, 8]. Three phases of pain associated with herpes zoster have been described, although their precise duration varies by author. According to most authors, the acute phase begins with the onset of the prodrome and resolves within 30 days. In the sub-acute phase, pain persists up to 120-160 days. In the chronic phase, pain persists over three months after the rash heals [7, 8]. While PHN pain usually resolves within six to twelve months after onset of the rash, it may last longer [7]. In some cases, it may never resolve. PHN rarely has been reported to occur months to years after resolution of the VZV episode. Recurrent PHN episodes in the same distribution as the initial rash are often preceded by a specific event affecting that dermatome (e.g., a surgical procedures, tooth abscesses, trauma, etc.) [3].

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### **Case Report**

A written HIPAA authorization to use/disclose existing protected health information and consent to publish this report were obtained. The patient is a 58year-old man with a history of Varicella

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infection at age six. He underwent immunosuppression with continuous 6mercaptopurine, sulfasalazine, and frequent, periodic prednisone tapers starting at age 37 for refractory Inflammatory Bowel Disease (IBD). At age 40, while immunosuppressed, he developed a non-disseminated, exquisitely painful vesicular rash along the right T3 dermatome, consistent with VZV. The rash resolved in six weeks, but he subsequently developed PHN with severe allodynia. The PHN subsided after a year of treatment with a tricyclic antidepressant, with mild residual allodynia, which did not require treatment. At age 50 he underwent a total colectomy as definitive treatment for IBD and his immunosuppression was tapered off. Now, at age 58, immunocompetent for eight years, he developed a second episode of shingles, initially presenting as deep neck pain near the right C4 transverse process, not preceded by any injury. Five days later, he developed a dermatomal vesicular rash consistent with VZV, which was promptly treated with a 5-day course of valacyclovir 1000 mg 3 times/day, plus a prednisone taper starting at 50 mg/day, consistent with currently accepted guidelines. Simultaneously, the same T3 dermatomal PHN pain that he had experienced 18 years earlier reoccurred without a vesicular rash. On day 13, duloxetine 30 mg/day (ramping to 60 mg/day), and gabapentin 300 mg 3 times/day were prescribed due to severe neuropathic pain. On day 18, gabapentin was substituted by pregabalin 100 mg 3 times/day, with improved effectiveness. The rash eventually resolved, but he developed severe cutaneous burning pain along both dermatomes. On day 18, he developed moderate but incomplete Bell's Palsy not preceded by a vesicular rash. Despite having undergone treatment with prednisone and valacyclovir for VZV a week earlier, a decision was made to administer a second course of the above, in concordance with the guidelines from the American Academy of Otolaryngology-Head and Neck Surgery Foundation for treatment of Bell's palsy when VZV is suspected as the causative agent [9]. His Bell's palsy improved moderately. Interestingly, the C4 and T3 pain completely subsided while he was taking prednisone, only to return after completing the corticosteroid taper. On day 42, now in the sub-acute phase of herpetic neuralgia, and having responded to and

improved during the prior course of corticosteroids, a decision was made to empirically administer a third 5-day course of prednisone. No guidelines exist for this. His Bell's palsy improved significantly while taking prednisone. As before, the C4 and T3 pain completely subsided, this time returning at a much lower intensity after completion of the taper. By day 75, his Bell's palsy was completely resolved, and there was only minimal residual pain along the C4 and T3 dermatomes. The T3 allodynia returned to its baseline. Pregabalin and duloxetine were continued as above. On day 188, the patient reported that he had been pain-free for perhaps three to four weeks. The pregabalin and duloxetine were tapered off over 2 weeks and he has remained painfree for over two months.

#### Discussion

This case involves presentation of a VZV reoccurrence with a unique triad of concurrent but different conditions: (1) Severe allodynia without sensory loss along a dermatome preceded by a rash; (2) Reoccurrence of severe PHN along a distant dermatome without a rash, and; (3) Bell's Palsy without a rash. Literature searches did not produce any reported cases with this triad of concomitant events. The course of the disease promoted the use of empiric pharmacologic treatment with three courses of oral prednisone for which no current guidelines exist. This patient did not meet any of the predictive risk factors for reactivation of VZV at the time of this second shingles outbreak, although he was immunosuppressed during his first episode. He did, however, meet two of the predictive risk factors for development of PHN, namely, pain beginning before the blisters appeared, and severe pain in the acute phase of the infection. Inexplicably, he also developed reoccurrence of pain in the right T3 dermatome 18 years after resolution of the first episode of PHN, with no precipitating traumatic event. The occurrence of ZSH has been widely described, sometimes in a dermatome distant from a dermatome with a rash. However, most of these reports involved VZV reactivation in the setting of other concurrent immunological deficiencies or other conditions causing depression of cellmediated immunity to VZV [5]. Reoccurrence of VZV in a previously affected dermatome has also been

documented after a traumatic episode in that dermatome. In contrast, this patient's immunological system was presumably intact after cessation of years-long immunosuppression eight years earlier, and he did not suffer any previous injury. This patient had isolated facial droop and muscle weakness consistent with Bell's palsy [9]. Bell's palsy was not the initial presentation of VZV, but the third isolated event. The most widely accepted treatment for Bell's palsy is oral corticosteroids, endorsed by The American Academy of Neurology guidelines for treatment of new-onset Bell's palsy, rating them as "highly likely to be effective" [10]. Evidence suggests that a combination of antivirals with corticosteroids might be more effective than corticosteroids alone. The American Academy of Otolaryngology-Head and Neck Surgery Foundation guidelines also support the use of corticosteroids and the optional use of antivirals [9]. Although the treatment guidelines cited above constitute the recommended initial course of treatment, this patient eventually was treated with a second course of steroids for the late development of Bell's palsy, serendipitously relieving the pain in other dermatomes and guiding ensuing clinical decisions. Based on the effectiveness of the second course of steroids, a third course was tried empirically on day 42 (for which no guidelines exist) which turned out to be the most effective treatment for this patient.

#### Conclusion

This case describes three VZV-related conditions occurring concurrently: (1) VZV in a dermatome preceded by deep pain and a vesicular rash, (2) Reactivation of an 18-year-old PHN in a different dermatome without a rash (ZSH); (3) Delayed development of isolated Bell's palsy without a rash. These all occurred in an immunocompetent patient without disseminated VZV. A literature search did not reveal any prior cases involving this triad of VZV-related concurrent manifestations, presumptive correlation between the presentation of symptoms and clinical course of this case, or the remote long-term immunosuppressive treatment that this patient had undergone years earlier. Further, the patient ended up receiving not one, but three, courses of corticosteroids over less than 2 months, for which there are no guidelines, which turned out to be key in

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resolution of his symptoms. No prior cases of an episode of VZV triggering recurrence of a previous case of PHN in a different distant dermatome were found either. VZV and PHN disease processes remain poorly

understood. It has been postulated that PHN constitutes more than one single disorder. The clinical presentation of PHN may manifest as several distinct patterns responding to different therapeutic

interventions [11]. Symptom presentation, disease course, and treatment response in this patient may support this theory.

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