Bilateral asymmetrical herpes-zoster with Ramsay hunt syndrome in an immunocompetent adult

Siqi Dai†, Xiaowen Huang†, Yuxiang Chen, Menglei Wang, Huanxin Zheng, Kang Zeng* and Li Li*

Abstract

Background: Bilateral herpes zoster (BHZ) is an atypical presentation of herpes zoster (HZ), with few cases reported before. Ramsay Hunt syndrome (RHS) is an uncommon complication of VZV infection. Cases of BHZ with RHS in immunocompetent adults have been reported rarely.

Case presentation: We described an immunocompetent adult who suffered from left-sided thoracic herpes zoster and contralateral RHS simultaneously, and summarizes the characteristics of BHZ.

Conclusions: Cases of BHZ with RHS in immunocompetent adults have not been reported previously. Antivirus - glucocorticoid combination therapy showed a good effect in this case.

Keywords: Antiviral therapy, Bilateral herpes zoster (BHZ), Glucocorticoid, Ramsay hunt syndrome (RHS), Varicella-zoster virus (VZV)

Introduction

Herpes zoster (HZ) is a common infection caused by the varicella-zoster virus (VZV), usually happened in patients in hypoimmunity. VZV remains dormant in nerve tissue until activated. And then it can move along the nerve fibers, lurking in the posterior root ganglion of the spinal cord. Patients with HZ present as erythema, pinhead-sized blister, exudate, neuralgia, which usually do not cross the midline of the body [1]. When bilateral dermatomes are involved, called bilateral herpes zoster (BHZ), Ramsay Hunt syndrome (RHS) is an infrequent, severe presentation of VZV reactivation in the geniculate ganglion. Patients with RHS often appear as herpes of external auditory meatus or tympanic membrane, earache, and facial numbness because of viral invasions to the facial nerve and auditory nerve [2]. Herein, we report an immunocompetent adult suffered from BHZ and RHS simultaneously. As far as we know, there are no other known cases like this patient.

Case report

A 55 year-old-male presented to the dermatology clinic with diffused erythema and clustered vesicles affecting the left chest and right ear (Fig. 1a, b). He complained of severe pain in the affected region. One week before, some vesicles appeared after taking alcohol. The typically neuropathic pain, such as burning sensation, Shock-like pain, stabbing pain, and feeling of numbness, has been accompanied by the rash. He took some anodyne in an attempt to relieve the pain, but it does not affect reducing symptoms. In the following days, there was a facial asymmetry that occurred in this patient, and the patient developed exudating in his right ear canal.

In the physical examination, grouped blisters, even hemorrhagicbulls with an erythematous base, appeared on his left chest and back along T4-T6 dermatomes. Some blisters had been ruptured and scabbed. The patient’s face was asymmetrical with the droopy corner of
the right mouth, and his right nasolabial fold became flattened, and the right eyelid could not be completely closed. The tympanic membrane was integral, but yellow to white exudation was observed on the surface of the external auditory canal.

Laboratory investigations, pure tone audiometry tests, and ear examination were routine. He did not have any chronic disease, medical history, recent weight loss, or exposure to any infectious diseases. The patient was not performed virology tests because the disease could be diagnosed based on typical clinical manifestations. He received the treatment of penciclovir 250 mg twice by dripping and methylprednisolone 40 mg once daily. Pain relief with oral gabapentin and super laser irradiation. By using this therapy for 1 week, vesicles over the right ear and left chest had been absorbed and crusted. The patient could perceive the pain relieving effect. Besides, the right facial palsy with lagophthalmus had slightly improved during this period. Continuation of hospitalization has been advised to the patient, but he denied and left the hospital voluntarily. After being discharged, he took oral valaciclovir 500 mg twice daily, methylprednisolone 24 mg per day, and mecobalamin for 7 days. Besides, acupuncture therapy was conducted on the patient once a week in a traditional Chinese medicine hospital. Two weeks after being discharged, the patient could almost close his right eyelid, and his feeling of pain was entirely resolved. And then, the dose of methylprednisolone had been reduced gradually and discontinued within 1 month. After 2 months follow-up, this patient could close his right eyelid completely, flattened nasolabial fold, and droopy corner of the mouth on his right side has also been improved. (Fig. 2).
Table 1: Overview of reported cases of bilateral herpes zoster (BHZ)

| No. | Age (years) | Sex | Dermatomes | Symmetry | Underlying disease | Treatment | Reference (see supplementary materials for details) |
|-----|-------------|-----|------------|----------|--------------------|----------|---------------------------------------------------|
| 1   | 61          | M   | R: T2–3 L: C5-T1 | Asymmetry | /                  | VCV oral 1 g/q8h 7 days | 1 |
| 2   | 24          | F   | R: L1–2 L: maxillary dermatome | Asymmetry | /                  | ACV oral 800 mg x 5/d 10 days | 2 |
| 3   | 7           | M   | R: C4 L: T3–4, L2 | Asymmetry | /                  | ACV oral 800 mg x 5/d 7 days | 3 |
| 4   | 16          | M   | R: trigeminal nerve dermatome L: T4–7 | Asymmetry | /                  | ACV oral 500 mg x 3/d 7 days | 4 |
| 5   | 45          | M   | R: T9 L: trigeminal nerve dermatome | Asymmetry | /                  | ACV IV 10 mg/kg x 3/d | 5 |
| 6   | 26          | M   | R: T8 L: T9 | Asymmetry | /                  | Isoprinosine 1,000 mg x 4/d | 6 |
| 7   | 73          | F   | R: L1–2 L: T9–10 | Asymmetry | /                  | Prednisolone oral 40 mg/d topical ACV and steroids | 7 |
| 8   | 60          | M   | R: trigeminal nerve dermatome, forearm L: back | Asymmetry | /                  | Quinine, iron and sulphate of magnesium oral | 8 |
| 9   | 40          | F   | R: neck and ear L: neck and shoulder | Asymmetry | /                  | ACV oral 800 mg x 5/d 7 days | 9 |
| 10  | 28          | M   | R: T8–9 L: T12, L1–2 | Asymmetry | /                  | ACV IV 1500 mg/m²/d Clindamycin IV | 10 |
| 11  | 14          | M   | R: forehead L: L1 | Asymmetry | /                  | ? | 11 |
| 12  | 21          | M   | Trigeminal nerve dermatome | Symmetry | /                  | ? | 12 |
| 13  | 3           | M   | Face, nose, chin and ear | Symmetry | /                  | ? | 13 |
| 14  | 41          | F   | Neck | Symmetry | /                  | ? | 14 |
| 15  | 15          | M   | T7–9 | Symmetry | /                  | ? | 15 |
| 16  | 33          | F   | Upper sacral areas, hips, and upper part of the buttocks bilaterally | Symmetry | /                  | ? | 16 |
| 17  | 24          | M   | Chest | Symmetry | /                  | ? | 17 |
| 18  | 18          | M   | Face and head | Symmetry | /                  | ? | 18 |
| 19  | 54          | M   | Neck | Symmetry | /                  | ACV IV 21 days | 19 |
| 20  | 23          | M   | Forehead and temporal areas | Symmetry | /                  | ? | 20 |
| 21  | 55          | M   | T4 | Symmetry | /                  | ACV oral 800 mg x 5/d 7 days | 21 |
| 22  | 75          | M   | Trigeminal nerve dermatome | Symmetry | Prostate carcinoma | ACV | 22 |
| 23  | 70          | M   | R: C4, T2 L: L1–2 | Asymmetry | CLL | ACV IV 10 mg/kg/8 h | 23 |
| 24  | 70          | M   | R: C4, T4 L: T9–10 | Asymmetry | Diabetic, CKD and MM | ACV 375 mg/d 10 days | 24 |
| 25  | 39          | F   | T8 | Symmetry | After thoracoscopic splanchinectomy | ACV oral 800 mg x 5/d 5 days | 25 |
| 26  | 31          | M   | Eyes | Symmetry | AIDS | ACV oral 800 mg x 5/d | 26 |
| 27  | 66          | F   | R: C4–5 L: facial and the posterior auricular nerves | Asymmetry | Rheumatism and heart disease | ? | 27 |
| 28  | 63          | M   | T11 | Symmetry | ESRD | VCV oral 250 mg/d | 28 |
| 29  | 54          | F   | R: T5–7 L: T10 | Asymmetry | MM | FCV | 29 |
Table 1 Overview of reported cases of bilateral herpes zoster (BHZ) (Continued)

| NO. | Age (years) | Sex | Dermatomes | Symmetry | Underlying disease | Treatment | Reference (see supplementary materials for details) |
|-----|-------------|-----|------------|----------|--------------------|----------|--------------------------------------------------|
| 30  | 52          | F   | Face and neck | Symmetry | SLE, TB            | ?        | 27                                               |
| 31  | 21          | M   | R: T9–10 L: T9 | Asymmetry | UC                | Antiviral IV | 28                                               |
| 32  | 47          | F   | L4–5, S1    | Symmetry | Renal transplantation | VCV oral 1 g tid 7 days | 29                                               |
| 33  | 27          | M   | R: T9 L: T6–8 | Asymmetry | Pharyngotonsillitis | Oseltamivir oral | 30                                               |
| 34  | 49          | F   | T4          | Symmetry | Breast cancer      | FCV 700 mg/d 7 days | 31                                               |
| 35  | 68          | F   | R: T8–9 L: C4 | Asymmetry | MM                | ACV 750 mg/d 6 days | 32                                               |
| 36  | 30          | M   | T10         | Symmetry | AIDS               | ACV 10 mg/kg tid | 33                                               |
| 37  | 64          | F   | L: T10      | Asymmetry | PAAS and diabetes  | FCV 750 mg/d 7 days | 34                                               |
| 38  | 67          | F   | L4–5 L: T7–8 | Asymmetry | Hypertension       | FCV 750 mg/d 7 days | 35                                               |
| 39  | 69          | M   | R: T5–7,10,12; L3–4 | Asymmetry | ESRD and SCCs | ACV IV 800 mg/d 7 days | 36                                               |
| 40  | 91          | F   | L2–5        | Symmetry | CKD               | Antiviral therapy | 37                                               |

M male; F female; L left; R right; C cervical; T thoracic; L lumbar; S sacral; IV intravenous; ACV aciclovir; FCV famiclovir; CLL chronic lymphocytic leukaemia; CKD chronic kidney disease; ESRD end-stage renal disease; MM multiple myeloma; AIDS acquired immune deficiency syndrome; SLE systemic lupus erythematosus; TB tuberculosis; UC ulcerative colitis; PAAS polymyositis associated antisynthetase syndrome; SCCs multiple squamous cell carcinomas

Discussion
Herpes Zoster is a viral disease caused by VZV, characterized as unilateral erythema, blisters, and pain. It usually affected one limited side of the body. When bilateral dermatomes are involved, called bilateral herpes zoster (BHZ), which is an atypical presentation of HZ, although it has an incidence rate under 0.1% and is usually found in immunosuppressed or senile patients [3], it also happened when VZV escapes unexpectedly from cellular immunity in healthy people. We have reviewed literature and found 40 cases of BHZ have been reported, as illustrated in Table 1. (References can be found in the Supplement 1), The data of those cases was organized with attributes of age, gender, involving dermatomes, underlying diseases, and treatments. It showed that the age of patients with BHZ ranged widely from 3 to 91, with an average of 43.35. Among them, 25 are males. Besides, the thoracic dermatome was mostly involved in BHZ, which was consistent with the previous reports [4]. Furthermore, 21 of the patients were immunocompetent, which includinte10 symmetrical and 11 asymmetrical lesions. The remaining 19 patients were immunocompromised, 7 of them have cancer, and 2 of them have acquired immune deficiency syndrome (AIDS) as underlying diseases. For most patients, the symptoms were relieved after the treatment of acyclovir or famciclovir.

In the case here, it is curious BHZ and RHS simultaneously happened in an immune-competent patient [5]. In our knowledge, there is no comparable cases have been reported. T cells are critical in the process of VZV delivery, especially for the reactivation of VZV [6]. By reviewing the medical history of this patient, we did not find he experienced any chronic illness nor received any immune suppressant medication. However, it is worth noting that the patient drank in 1 week before the onset of illness as mentioned in some reports that alcohol exposure weakens the body’s defense against virus and even leads to more severe or faster disease progression [7]. Thus, we hypothesize that heavy drinking may be one factor contributing to the reactivation of VZV in the two separate ganglia.

Antivirus therapy is necessary for treating HZ, and early application of glucocorticoid might be useful in reducing swelling and easing inflammation of the nerves [1]. A retrospective study suggested that antivirus - glucocorticoid combination therapy could improve the recovery rate of facial paralysis [8]. Our case confirmed the efficacy of the combination. After admission, the patient received penciclovir 250 mg twice by dripping and methylprednisolone 40 mg once daily immediately. The vesicles in the body had been absorbed, and the pain was alleviated. He also received acupuncture therapy after discharged from the hospital. The facial nerve function of this patient had improved gradually in the months’ follow-up.

Conclusions
In conclusion, effective antivirus treatment is the key to treat HZ. And antivirus - glucocorticoid combination therapy is necessary for patients who with RHS. Acupuncture therapy may be helpful to the reparation of injury nerves in advanced stages. However, more research is needed to confirm its effectiveness and security.
Supplementary information
Supplementary information accompanies this paper at https://doi.org/10.1186/s12985-020-01392-0.

Additional file 1. Supplement 1. References for 40 cases in the Table 1.

Abbreviations
AIDS: Acquired immune deficiency syndrome; BHZ: Bilateral herpes zoster; HZ: Herpes zoster; RHS: Ramsay Hunt syndrome; VZV: Varicella-zoster virus

Acknowledgments
Not applicable.

Authors’ contributions
DSQ and HXW analyzed the patient data and were a major contributor in writing the manuscript, CYX and ZHX followed up the patient, WML, LL, and ZK made clinical treatment decisions of this patient. The author(s) read and approved the final manuscript.

Funding
This work was supported by Guangdong Natural Science Foundation (2018A030313773).

Availability of data and materials
Not applicable.

Ethics approval and consent to participate
Not applicable.

Consent for publication
Consent for publication has been obtained from the patient.

Competing interests
The authors declare that they have no competing interests.

Received: 28 May 2020 Accepted: 30 July 2020
Published online: 15 August 2020

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