Hemi-diaphragmatic paralysis and herpes zoster infection in a renal transplant recipient; a case report

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Abstract

Herpes zoster or shingles is a common condition caused by the varicella zoster virus (VZV). Although it may cause numerous complications, diaphragmatic paralysis is an extremely uncommon event. It usually occurs after involvement of the C3, 4 and 5 dermatomes which give rise to the phrenic nerve. No case has ever been reported in renal transplant recipients. We report a case of 60-year-old-female patient from Punjab, Pakistan, who was a renal transplant recipient on immune suppressant therapy. She presented with worsening shortness of breath 3 weeks after the onset of a painful vesicular rash involving her neck, shoulder and axilla. Serial chest radiographs revealed a persistently elevated right hemi-diaphragm. Suspecting diaphragmatic paralysis, an ultrasound sniff test was performed which revealed absent contractions of the right hemi diaphragm clinching the diagnosis. She was started on anti-viral therapy and managed conservatively. Six months later a repeat ultrasound sniff test revealed normal contractions and movements of the paralyzed diaphragmatic muscle. Herpes zoster may cause diaphragmatic paralysis in renal transplant recipients. It should be considered in patients with respiratory distress and a typical vesicular rash involving the C3, 4 and 5 dermatomes. Diagnosis is supported by chest radiography and may be confirmed with ultrasonography. Treatment is usually supportive and acyclovir may be of benefit.

Introduction

Herpes zoster also known as shingles, is a common infection caused by reactivation of the varicella zoster virus (VZV). It produces a painful vesicular segmental rash in a dermatomal distribution. Neuralgic pain is thus the most important complication which may last for months and even years (1). Motor weakness is rare with few case reports on limb paresis post infection. Diaphragmatic paralysis is an even rarer complication that may occur with involvement of the C3, C4 and C5 dermatomes, thus affecting the phrenic nerve supply to the muscle (2). We performed a thorough literature review and found 18 case records. We report a case of a middle aged lady who was a renal transplant recipient and presented with worsening shortness of breath 3 weeks after the onset of a painful rash that involved her neck, shoulder and axilla. Work up revealed a paralyzed right hemidiaphragm. She responded to treatment with acyclovir and 6 months later was found to have normal movements of the muscle. To the best of our knowledge, this is the first case of herpes zoster induced diaphragmatic paralysis in a renal transplant recipient. We also discuss important clinical, diagnostic and therapeutic aspects of the disease (1-3).

Case Presentation

A 60-year-old female patient of Punjabi descent, presented to our hospital with progressively worsening shortness of breath for one week. She also complained of moderate to severe pain involving the skin of her neck, shoulder and upper chest for the...
past 10 days. She described the eruption of a painful rash in this region about four weeks ago which had started to resolve when the pain appeared. Her past medical history was significant for type II diabetes mellitus, hypertension and end stage renal disease for which she received a living-related renal transplantation in 2005. She did not smoke, had no addictions or allergies. Her home medications included nifedipine 30 mg once daily, carvedilol 12.5 mg twice daily, mycophenolate mofetil 360 mg twice daily, cyclosporine 50 mg twice daily and prednisolone 5 mg once daily. On examination, she was an elderly woman in significant respiratory distress. The pulse rate was 105 beats per minute, blood pressure 140/80 mm Hg, respiratory rate 30 per minute and she was a febrile with an oxygen saturation of 96% on supplemental oxygen of 2 liters/minute. There was a healing rash on the shoulder and in the axilla. Examination of the chest revealed reduced chest movements and expansion on the right side with decreased air entry at the right lung base.

Investigations performed in the emergency revealed hemoglobin of 10.4 g/dL, White cell count of 9.7×10⁹/µL, platelets ×10¹²/µL and serum creatinine of 1.3 mg/dL. A chest x-ray was performed which revealed an elevated right hemidiaphragm as shown in Figure 1A. She was managed with oxygen and bilevel positive airway pressure (BiPAP) support. This persistent respiratory distress, history of herpes zoster involving dermatomes C3, 4, 5 and the persistently elevated right hemidiaphragm lead us to consider herpes zoster induced diaphragmatic paralysis as a possible culprit. To confirm this, an ultrasound sniff test of the diaphragm was performed. This revealed absent excursions of the right hemidiaphragm confirming the diagnosis.

She was subsequently managed with pulmonary rehabilitation and pain management for post herpetic neuralgia. Acyclovir 800 mg five times a day was started for day 14 days. Patient remained hospitalized for another week during which she became oxygen independent and was discharged home with frequent follow ups and home BiPAP advice if she developed difficult breathing. Six months later, a repeat ultrasound sniff test revealed improved excursions and movements of the diaphragm thus re confirming the diagnosis. To date, she is well without any respiratory distress. A repeat chest radiograph revealed normal position of the previously paralyzed hemidiaphragm as shown in Figure 1B.

Discussion

Herpes zoster also called shingles is caused by the VZV of the herpes family. It produces a painful vesicular rash in a dermatomal distribution classically appearing on only one side of the body (1,2). The virus causes chicken pox earlier in life and then remains dormant in the dorsal or cranial root ganglia. It may reactivate any time later, producing the rash of shingles (3). Risk factors for reactivation include malignancies, HIV virus infection, immune suppressant use and a fall in the cell mediated immunity (4).

A frequent complication associated with shingles is post herpetic neuralgia that occurs in 8%-70% of patients. This may persist for months and sometimes for years and is more frequent in older individuals (1). Other complications include myelitis, meningoencephalitis and motor weakness. The latter may involve the limbs, the facial nerve, the bladder or bowel function and very rarely the diaphragm (5). Particularly interested about the later complication we performed a literature search on PubMed for herpes zoster induced diaphragmatic paralysis. We found a total of 18 cases.

The mechanism postulated is a neuritis or nerve inflammation with resultant weakness of the muscle. The phrenic nerve is affected in this case which carries motor supply to the diaphragm via C3, 4 and 5 nerve roots. The time period between appearance of the rash and development of diaphragmatic weakness is variable with reported intervals between one week and two months (6-8). Our patient had a rash affecting the neck, shoulder and axilla in a similar dermatomal distribution and probably suffered diaphragmatic weakness three weeks after the onset of rash. Very rarely the paralysis may be bilateral, though our patient had unilateral involvement.

The most common symptom for presentation is respiratory distress due to paresis of the diaphragm. Our patient had a similar presentation. The possible pathogenetic mechanisms include either the direct damage of the anterior horn cells by the virus or diffusion of the viruses into anterior nerve 0-= roots from the dorsal root ganglion, the usual location of the latent virus (8,9).

Herpes zoster induced diaphragmatic paralysis has been reported in conjunction with HIV-tuberculosis co infection and after chemotherapy for adeno squamous carcinoma of the lung. No case has previously been documented in a patient with a solid organ transplant. In our case, the patient was a renal transplant recipient and had been on immune suppressants for the past nine years. Modalities used to diagnose diaphragmatic dysfunction include ultrasound, fluoroscopy, electromyography and magnetic resonance imaging (MRI). We diagnosed the entity in our patient with ultrasonography which has a sensitivity of 93% and is 100% specific.
Treatment involves pulmonary rehabilitation and supportive care along with acyclovir or valacyclovir for one week. A past study has shown improvement of idiopathic diaphragmatic paralysis with these antivirals (9). We administered acyclovir for two weeks. Six months later an ultrasound sniff test revealed completely normal excursions of the diaphragm. Past cases report a variable time for recovery of diaphragmatic function, though it usually takes 4-6 weeks for recovery (9).

Conclusion
Herpes zoster may cause diaphragmatic paralysis in renal transplant recipients. It should be considered in patients with respiratory distress and a typical vesicular rash involving the C3, 4 and 5 dermatomes. Diagnosis is supported by chest radiography and confirmed with ultrasonography. Treatment is usually supportive, though acyclovir may be helpful.

Authors’ contribution
ZUA performed the literature review, drafted the entire manuscript, reviewed it and approved the final version. AA diagnosed the entity in the patient and was involved in patient management. He also reviewed and approved the article before being sent to the journal. SNM was the primary care physician and was involved in the management of this patient. He also helped in drafting the manuscript, reviewing it and approved the final version. AR was involved in performing the literature review and drafting the manuscript and also helped in reviewing the article and approved it for final submission. USM was involved in drafting the manuscript and reviewing subsequent drafts. He also approved the final version for submission. All others read and approved the final manuscript.

Conflicts of interest
The authors declared no competing interests.

Ethical considerations
Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors. Also, written informed consent was obtained from the patient for publication of this case report and accompanying images.

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