CASE REPORT

Brachial plexitis preceding encephalomyelitis in a patient with West Nile virus infection

Sonja Scholz, Bonnie Kaas, Alexis Simpkins, Jennifer Lyons, Arun Venkatesan, John Probasco

Department of Neurology,
Johns Hopkins University,
Baltimore, Maryland, USA

Correspondence to
Dr Sonja Scholz;
sscholz5@jhmi.edu

SUMMARY

We describe the case of a 54-year-old woman with West Nile virus infection presenting with painful brachial plexitis and radiculitis that preceded the more typically associated symptoms of meningoencephalitis. Physicians should be aware that West Nile virus infection can present with painful brachial plexitis.

BACKGROUND

West Nile virus (WNV) meningoencephalitis is an increasingly recognised flavivirus infection transmitted to humans primarily through infected mosquito bites.¹ Although the majority of infections are asymptomatic, a small percentage of patients present with neurological deficits and even death.² Last summer several US cities declared public health emergencies due to the rising numbers of WNV. Targeted therapies, including ribavirin, interferon α and WNV-specific immunoglobulin, have been reported to be beneficial in early stages of infection.³ However, diagnosis and treatment can be delayed by the protean manifestations.

Here, we document a case of WNV infection presenting with painful brachial plexitis and radiculitis. The patient only developed symptoms of meningoencephalitis, more typically associated with WNV infection, at a later stage of her illness. This unusual presentation of WNV infection led to a delay in establishing the diagnosis and institution of antiviral therapy.

CASE PRESENTATION

A 54-year-old woman presented with a 3-week history of generalised muscle weakness and severe, bilateral shoulder pain. She was a veterinary nurse who had undergone a dental procedure 3 weeks prior to presentation. After completion of postprocedure prophylactic antibiotics, she developed fevers of 102°F, and a diffuse, non-pruritic, maculopapular rash on her anterior chest that spread to her arms and legs. Her clinical picture was dominated by severe, sharp pain involving her shoulders and interscapular region. The pain did not radiate, was constant and required high doses of opiates for relief. Over the next week, she developed weakness of the upper and lower limbs resulting in difficulty climbing stairs and raising her arms above shoulder level.

An examination demonstrated a facial and torso erythematous rash and mild encephalopathy manifesting as altered behaviour and irritability. Strength testing revealed a marked weakness of bilateral shoulder abductors (MRC grade=3/5), left external

rotator muscles (3/5), as well as bilateral hip flexion (3/5), knee flexion (4/5) and knee extension (4/5) weakness. All other muscle groups had normal strength. Deep tendon reflexes were initially normal (2+) and symmetrical in the upper and lower limbs and toes were down going on Babinski testing. The rest of the neurological examination was within normal limits. In particular, cranial nerves II to XII were intact, and there was no evidence of ataxia or sensory loss (light touch, pinprick, proprioception and vibration) in either the upper or lower limbs. The rest of her medical examination was unremarkable.

INVESTIGATIONS

Cerebrospinal fluid (CSF) analysis revealed a lymphocytic pleocytosis (119 white cell count/uL, normal range 0–4/uL; protein 207 mg/dL, normal range 15–45 mg/dL) and normal glucose (61 mg/dL, normal range 50–80 mg/dL). Bacterial and fungal cultures from CSF were without growth. Nerve conduction studies and electromyography (EMG) were initially normal. Brain MRI with and without gadolinium was unremarkable. Spine MRI demonstrated diffuse abnormal enhancement of the anterior nerve roots of the cauda equina at the level of T12 and below. There was no evidence of spinal cord compression or mass. Shoulder MR neurogram showed hyperintense signal in the left brachial plexus consistent with brachial plexitis (see figure 1). An extensive initial laboratory workup was unremarkable aside from mildly elevated lactate dehydrogenase (268 U/L; normal range: 122–220 U/L). Creatine kinase and aldolase were normal, and autoantibodies including antinuclear antibody, rheumatoid factor, anti-DNA, anti-Ro, anti-La, anti-RNP and anti-Jo1 were negative. An initial infectious evaluation was negative (PCR was negative for herpes simplex, cytomegalovirus, varicella zoster, Epstein-Barr virus and enterovirus; negative monospot test and normal Lyme antibody serum titres). Several days later, WNV IgM and IgG titres from plasma returned positive (4.19 and 1.95, respectively; repeat titres 2 weeks later were 4.14 and 3.35, reference: IgM<1.30 and IgG<0.90) and WNV IgG titre from CSF was 4.63 (reference <0.90). WNV RNA from CSF was not detected.

DIFFERENTIAL DIAGNOSIS

The patient was diagnosed with brachial plexitis, radiculitis and meningoencephalitis (based on CSF leucocytosis and mild encephalopathy manifested by the patient) secondary to WNV infection.

CrossMark

To cite: Scholz S, Kaas B, Simpkins A, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-200833

Figure 1 MR neurography demonstrating left brachial plexitis. Figure (left panel) showing hyperintense signals (arrow) at the left brachial plexus consistent with brachial plexitis. Arrowheads (right panel) indicating denervation oedema in the supraspinatus and infraspinatus muscles. These changes were detected, while electromyography was still normal.

TREATMENT

During her hospital stay, the patient received high doses of intravenous opiates to relieve her shoulder/upper back neuropathic pain. The patient also received a 5-day course of intravenous methylprednisolone (1000 mg/day), followed by an oral steroid taper. With this treatment regimen, her pain symptoms gradually improved, allowing a transition to oral opiates and gabapentin (300 mg thrice daily). The patient was discharged to rehabilitation after a 7-day admission.

OUTCOME AND FOLLOW-UP

Follow-up 2 months after symptom onset revealed moderate improvement in her muscle strength to the point that she was able to climb stairs and remain standing for over 2 hours at a social event. Her shoulder pain had resolved and she no longer required analgesia. Follow-up EMG at that time demonstrated positive sharp waves in the left C5–T1 myotomes.

DISCUSSION

Our report highlights that WNV infection can present as brachial plexitis in addition to classical meningoencephalitis. Although it is possible that the brachial plexitis arose as a complication of her dental procedure, the associated CSF lymphocytic pleocytosis and positive WNV antibody titres strongly support an infectious aetiology for our patient's condition. Indeed, brachial plexopathy has been previously described in the setting of WNV infection.⁴ Infection with this particular virus is frequently associated with diffuse centrifugal maculopapular rash and mildly elevated lactate dehydrogenase levels, as was observed in our case. Despite positive viral antibody titres, WNV RNA was not detected. However, viral nucleic acid is typically detected only during the first 48 h postinfection, rapidly becoming negative thereafter.

Early diagnosis of brachial plexitis is based on the clinical picture of severe shoulder and back pain. Clinical weakness and associated neurophysiological findings are typically delayed. In our patient, the clinical diagnosis of brachial plexitis was confirmed by MR neurography, supporting the utility of this new imaging modality in the workup of suspected brachial plexitis. As the interval between symptom onset and diagnosis was protracted, and data were lacking to justify the risks of targeted antiviral therapies, she was continued on steroids alone with gradual clinical improvement.

WNV infection may be under-recognised due to its protean presentations, which complicates accurate assessment of the

frequency of neurological involvement. Understanding its diverse manifestations is imperative for early institution of supportive care and/or targeted therapies on a case-by-case basis. This case highlights the need for improved recognition of the neurological complications of WNV and for concentrated efforts to identify more effective antiviral treatments.

Learning points

- ▶ West Nile virus (WNV) infection can present with painful brachial plexitis.
- ▶ Recognition of the protean manifestations of WNV infection will facilitate early diagnosis and the institution of appropriate therapeutic measures.
- ▶ Diffuse, centrifugal, maculopapular rash and mildly elevated lactate dehydrogenase levels are frequently observed in patients with WNV infections.
- ▶ MR neurogram is a new imaging modality that is helpful in the diagnosis of brachial plexitis.

Contributors SS, BK, AS, JL, Arun Venkatesan and John Probasco were involved in the conception and design of this manuscript. Each of the above listed authors critically reviewed and revised the article and gave final approval of the current version.

Competing interests Dr Scholz received research support from the Michael J Fox Foundation. She received a honorarium from WebMD.com for an online publication. Dr Venkatesan receives research support from the Howard Hughes Medical Institute and the Maryland Stem Cell Research Foundation. He receives consulting fees as a medical expert for the US Government Vaccine Injury Compensation Program. Ms Kaas, Dr Simpkins, Dr Lyons, and Dr Probasco report no disclosures.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 CDC Fact Sheet. West Nile Virus: what you need to know. http://www.CDC.gov/ncidod/Dvbid/westnile/WNV_factsheet.htm
- 2 Sejvar JJ, Haddad MB, Tierney BC, *et al.* Neurologic manifestations and outcome of West Nile virus infection. *JAMA* 2003;290:511–15.
- 3 Hayes EB, Sejvar JJ, Zaki SR, *et al.* Virology, pathology, and clinical manifestations of West Nile virus disease. *Emerg Infect Dis* 2005;11:1174–9.
- 4 Almhanna K, Palanichamy N, Sharma M, *et al.* Unilateral brachial plexopathy associated with West Nile virus meningoencephalitis. *Clin Infect Dis* 2003;36:1629–30.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow