CASE REPORT

West Nile virus-associated brachial plexopathy

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SUMMARY

West Nile virus (WNV) is the most frequent cause of arbovirus infection in the USA. Only 20% of infected individuals are symptomatic. Less than 1% of symptomatic individuals display West Nile neuroinvasive disease. We report a rare case of WNV-associated brachial plexopathy in a young immunocompetent individual, without cerebrospinal fluid pleocytosis or encephalitis. Additionally, there was subjective and objective improvement after high-dose corticosteroids. This case adds to the clinical spectrum of WNV neuroinvasive disease. The literature regarding immunomodulatory treatment and WNV is reviewed.

BACKGROUND

West Nile virus (WNV) is a mosquito-transmitted flavivirus, which is the most frequent cause of arboviral disease in the USA.[1] Less than 1% of infected, symptomatic patients present with WNV neuroinvasive disease (WNND), commonly manifesting as acute flaccid paralysis, meningitis and encephalitis. Here, we present an atypical case of WNND associated with brachial plexopathy, near normal cerebrospinal fluid (CSF) profile and possible response to steroids. This case illustrates a rare presentation of WNND.

CASE PRESENTATION

A 21-year-old man presented with at least 6 months of severe right shoulder pain, progressive right upper extremity weakness and paraesthesias. He denied intravenous drug use, antecedent infection, neck or shoulder injury. He denied known tick or mosquito bites. Examination revealed significant right shoulder girdle atrophy with scapular winging. Using the Medical Research Council (MRC) scale, right trapezius, supraspinatus, infraspinatus and rhomboid muscle were 4/5. Right deltoid and biceps brachii muscles were 4+/5. Reduced sensation to all modalities was noted in the right radial, lateral antebrachial and posterior cutaneous nerve distributions. Right brachioradialis and biceps reflexes were hypoactive. Motor and sensory examination was otherwise normal. Plantar responses were flexor with absence of frontal release signs.

INVESTIGATIONS

Nerve conduction studies of the bilateral median, ulnar and radial nerves showed normal sensory responses. The right lateral antebrachial cutaneous sensory nerve testing showed an amplitude reduction of greater than 50% when compared to the left. Needle electromyography showed fibrillations, positive sharp waves and reduced recruitment without signs of reinnervation in the right biceps, deltoid, infraspinatus and serratus anterior. These findings were interpreted as a brachial plexopathy, rather than polyradiculopathy, given the predilection for serratus anterior, clinical course and reduced right lateral antebrachial cutaneous sensory nerve action potential.

Serum methylmalonic acid, complete blood count, inflammatory studies (erythrocyte sedimentation rate, antinuclear antibody), syphilis and Lyme antibody testing were all normal. Contrasted MRI of the cervical spine and brachial plexus was normal.

Patient's serum WNV immunoglobulin M (IgM) titre was elevated at 1.57 (normal <0.90) with serum immunoglobulin G (IgG) antibody titre showing a four-fold increase at 4.64 (normal <1.30). CSF analysis revealed an elevated WNV IgG antibody titre with a negative IgM antibody titre. CSF protein concentration was mildly elevated at 48 mg/dL (normal 15–45 mg/dL) with zero white cell count on CSF.

TREATMENT

Clinical examination 1 month later revealed progression of weakness on the right deltoid MRC 4+ to 4 with correlating subjective difficulty with activities of daily living (ADL). The diagnosis of WNND was discussed with the patient. The unproven treatments including steroids, intravenous immunoglobulin, ribavirin and interferon were discussed with the patient.[2] Since the patient was only 21 and reported difficulty with ADL and progression of weakness, we discussed the risk and benefit profile of immunomodulatory treatment. The patient opted for a treatment trial of scheduled high-dose intravenous methylprednisolone 1000 mg weekly for 3 months.

OUTCOME AND FOLLOW-UP

At follow-up testing, the patient reported that he no longer had difficulty with his activities of daily living due to right arm weakness. His objective deltoid strength measurement by the same examiner improved from MRC 4 to 4+. His repeat electrodiagnostic study showed normal bilateral upper extremity sensory responses and no further signs of active denervation in the previously affected muscles.

DISCUSSION

WNND most commonly presents as an acute flaccid paralysis.[2 3] This case illustrates a rarely reported clinical phenotype of brachial plexopathy associated with WNV.[4] Additionally, the lack of

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CSF pleocytosis is rarely reported in only 3% of patients with WNV.[5]

The presence of elevated WNV serum IgM/IgG and CSF IgG antibodies established that our patient had neuroinvasive WNV disease at some point in the past year, which occurs in <1% of patients infected with WNV.[5] Occasionally WNV CSF IgG may be a false positive due to other flavivirus infections; however, this is unlikely when WNV serum IgM and IgG studies both show increased titres. Unfortunately, WNV PCR was not performed. Parsonage turner's syndrome, idiopathic brachial neuritis, was considered in our patient. However, the majority of patients with Parsonage turner's syndrome will reach their nadir within 6 months, as this is considered a monophasic illness. The persistent progression of our patient's symptoms for more than 6 months, with progression established by objective clinical examinations 1 month apart, provided an indication for evaluating further aetiologies. We believe that the temporal profile of the WNV serum and CSF studies is also consistent with our patient's temporal profile of symptoms and progression. Although there are a few case reports of WNND affecting the brachial plexus, peripheral nerve involvement is unusual.[4 6 7] Objective sensory abnormalities are usually absent and this is attributed to WNV predominantly attacking the anterior horn motor neurons of the spinal cord, as can be seen in pathological studies.[1 2 7] It is possible that previously described case reports of the Guillain-Barre syndrome and brachial plexopathies in relation to WNND may be due to the virus leading to a secondary immune-mediated phenomenon.[6–8] This may explain the subacute presentation of our patient as opposed to the acute findings in the more common manifestions of WNND: meningitis, encephalitis or acute flaccid paralysis. Therefore, peripheral nerve manifestations could theoretically be more susceptible to treatments such as methylprednisolone.

Our patient improved both subjectively and objectively to a treatment trial of intravenous methylprednisolone. However, it would be difficult to ascertain causality as physiotherapy was also initiated in this time frame. Additionally, resolution of active denervation in electrodiagnostic studies may be due to the expected evolution of nerve injury. We speculate that if corticosteroids played a role in our patient's recovery, it would be due to an anti-inflammatory mechanism in improving secondary immune-mediated injury.[6] However, further large-scale randomised studies should be performed to evaluate safety, risks and benefits of immunomodulatory treatment of WNND. There is anectodal evidence to suggest efficacy of immunomodulatory treatment. In Israel, 12 patients with WNND were treated with intravenous immunoglobulin (IVIG) with partial or complete recovery observed in 9 of the 12 patients.[8] However, IVIG in Israel had higher titres of anti-WNV than was found in IVIG in the USA.[7] Further studies of IVIG treatment in patients with WNV have had mixed results.[9]

We present an atypical case of subacute, progressive brachial plexopathy in a patient with WNV, expanding the known neuromuscular manifestations. Further studies should be performed to evaluate efficacy of immunomodulatory treatments in patients with WNND.

Learning points

- West Nile virus neuroinvasive disease (WNND) can present without pleocytosis in cerebrospinal fluid.
- WNND can include rare manifestations such as brachial plexus, rather than the more common acute flaccid paralysis, meningitis or meningoencephalitis.
- Supportive care is considered a mainstay of treatment.
- Some literature suggests that patients may benefit from immunomodulatory treatment.

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Competing interests None declared.

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