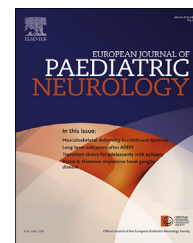




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Case Study

Longitudinal extensive transverse myelitis with cervical epidural haematoma following dengue virus infection



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ABSTRACT

Background: Longitudinal extensive transverse myelitis associated with dengue infection is rare with no reported paediatric cases.

Methods: We report a 12-year-old girl who presented with flaccid quadriplegia 8 days after onset of acute dengue fever. MRI spine showed T2 hyperintensity associated with epidural hematoma at C3–C6 level of the spinal cord. Transcranial magnetic brain stimulation revealed absent motor evoked potentials bilaterally. We also summarise and compare the reported cases of transverse myelitis associated with dengue infection.

Results: Immunomodulatory treatment was given which included pulse methylprednisolone, intravenous immunoglobulin and plasmapheresis. Six months post-admission, there was a good (near-complete) clinical recovery with the repeat MRI showing mild residual hyperintensity at C4 level and complete resolution of epidural haematoma.

Conclusion: This is the first reported paediatric case of longitudinal extensive transverse myelitis following dengue infection. It is also the first to illustrate that in patients with concomitant epidural haematoma a good outcome is possible despite not having surgical decompression. Clinicians should be aware of parainfectious dengue-related longitudinal extensive transverse myelitis in children and consider prompt immunomodulatory treatment.

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1. Introduction

Dengue is the most common mosquito borne disease in the world with an estimated 50 million people affected by dengue

each year.¹ Dengue virus infection is known to be associated with neurological manifestations including encephalopathy, acute disseminated encephalomyelitis (ADEM), encephalitis, Guillain-Barre syndrome and transverse myelitis (TM).^{2,3}

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Acute TM is a spinal cord syndrome with a relatively abrupt onset of motor, sensory, and sphincter disturbances that is usually attributed to an inflammatory demyelinating lesion. Approximately 20% of acute TM cases occur in children.⁴ Longitudinally extensive transverse myelitis (LETM) refers to a spinal lesion that extends at least three vertebral segments.⁵ TM is classified broadly as: (i) demyelination – including monofocal clinical isolated syndrome or part of multifocal demyelinating disease including ADEM or multiple sclerosis; (ii) associated with systemic connective tissue disease; (iii) infectious and (iv) idiopathic of which the majority are presumably parainfectious in aetiology.⁵

Spinal magnetic resonance imaging (MRI) plays a crucial role in making the diagnosis of TM. In addition, transcranial magnetic brain stimulation (TMS) used to measure central motor conduction time (CMCT) is also a useful non-invasive investigation to assess the integrity of the corticospinal tract. Prolonged CMCT has been demonstrated in patients with TM.⁷ There has only been one published report to date showing the utility of CMCT in a paediatric TM patient.⁸

TM associated with dengue infection is uncommon with only 10 published case reports to date of which only one was in a paediatric patient aged 14 years.^{9–17} Of these 10 published cases, LETM was even rarer with only 5 adult cases of LETM reported (age range 31–45-years old) and 2 of these having concomitant spinal epidural haematoma.^{9–12} There were no reported paediatric cases of LETM or spinal epidural haematoma associated with dengue infection.

We present a 12-year-old girl with LETM and concomitant cervical epidural hematoma following dengue infection. We also show the utility of serial non-invasive TMS in this patient and summarise the reported cases of TM associated with dengue infection.

2. Case report

A previously healthy 12-year-old girl was transferred to our tertiary unit from a local hospital on day 9 of illness with acute lower limb paraparesis and dysesthesia of her chest wall. She was diagnosed as acute dengue fever at the local hospital after a 6-day history of pyrexia and myalgia. Her full blood count at day 6 showed haemoglobin of 18 g/dL (elevated haematocrit of 46.8), leucopenia ($2.7 \times 10^9/L$), thrombocytopenia ($32 \times 10^9/L$) and positive dengue IgM serology. There was no evidence of severe plasma leakage or organ impairment. She was managed with intravenous (IV) fluid rehydration and entered the defervescence phase at day 7 associated with a defervescence rash and normalisation of her blood counts from day 8. At day 8 of illness; she developed bilateral lower limb weakness of grade 1/5 Medical Research Council (MRC), impaired sensation from C6 dermatome downwards and incontinence of both bowel and bladder. Her weakness progressed and reached the nadir at 24 h resulting in a complete flaccid quadriplegia with power of grade 0/5 MRC in all 4 limbs. Proprioception and vibration sense were absent bilaterally and she had autonomic instability with a fluctuating heart rate. She remained fully conscious without features of encephalopathy; however she was ventilated for an impending respiratory failure.

MRI spine at day 1 of weakness (day 8 of illness) showed a non-enhancing intramedullary lesion from C3 to C6 (hyperintensity on T2-weighted images) associated with marked cord oedema and cervical epidural haematoma (Fig. 1). Her brain MRI was normal. Cerebrospinal fluid (CSF) biochemistry, microscopy and culture were normal. Repeat CSF examination at day 17 of illness was negative for oligoclonal bands, dengue virus PCR, dengue IgM and Japanese encephalitis IgM. Ophthalmologic examination was normal with no optic neuritis. Repeat dengue serology testing at day 9 of illness were both IgM and IgG positive and dengue NS1 antigen was negative. Serum for N-methyl D Aspartate Receptor antibody, anti-voltage-gated potassium channel antibody and aquaporin 4 (AQP4) antibody were also negative. The rest of her baseline haematological, renal, liver function and autoimmune tests were normal.

On day 2 of weakness she was treated with IV methylprednisolone 30 mg/kg/day for 3 days followed by oral prednisolone and IV Immunoglobulin 1 g/kg/day for 2 days. Her cervical epidural haematoma was managed conservatively as her repeat MRI showed a reduction in epidural haematoma on day 3 of weakness (Fig. 1). As there were still no signs of improvement by day 10 of weakness, she then underwent 6 cycles of plasma exchange. She was extubated on day 13 of weakness and an improvement in both upper limb muscle power of grade 1/5 MRC were seen on day 16 of weakness. From week 3 of weakness onwards, she regained bladder and bowel control and continued to improve in strength (upper limbs were grade 3/5 MRC and her lower limbs were grade 2/5 MRC).

Somatosensory evoked potential done at week 3 of her weakness showed an intact median nerve response at the Erb's point (left N9: 8.33 ms, right N9: 8.44 ms), the dorsal horn of the cervical spinal cord (left N13: 10.1 ms, right N13: 10.94 ms) but an absent response at the somatosensory area indicating conduction defect between the cortex and cervical spinal cord. A repeat spine MRI at month 1 following onset of weakness showed resolution of epidural hematoma with residual hyperintense cord lesion at the level of C4 which reduced in signal further at month 6 post-weakness onset (Fig. 1). To monitor her progress of recovery in addition to regular clinical assessment of power (MRC power grading), we also performed TMS to calculate CMCT. The summary of the serial results of both her CMCT and MRC power are shown in Table 1. Six months following her onset of weakness, she made good clinical recovery being able to walk independently with mild residual weakness of her right limbs (grade 4+/5 MRC) and her CMCTs had normalised.

3. Discussion

This is the first reported paediatric case of dengue virus associated LETM and expands the parainfectious causes of paediatric LETM. Our case also reiterates the clinical importance of differentiating between TM and LETM as LETM is associated with a number of specific conditions which has important diagnostic, prognostic and therapeutic implications.¹⁸ The most frequent cause of LETM is neuromyelitis optica (NMO) which requires long-term immunosuppression to prevent relapses.¹⁸ In cases with a negative NMO/AQP4 IgG, other causes including infectious and parainfectious causes

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