Longitudinally extensive transverse myelitis with encephalopathy following acute dengue infection

Mielite transversa longitudinalmente extensa com encefalopatia após dengue

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ABSTRACT

Dengue is a common arboviral infection in tropical/subtropical regions; neurologic complications occur in ~5% of cases, usually as encephalopathy from systemic illness. Viral neurotropism and immune-mediated injury are rare and mimic infectious/inflammatory etiologies. Although corticosteroids are not routinely recommended, case reports describe high-dose steroids usage with favorable outcomes.

We report the case of a previously healthy 25-year-old male patient from Rio de Janeiro who developed fever and retro-orbital headache followed by abdominal pain, hallucinations, upper limb ataxia, and areflexic paraparesis with T10 sensory level and urinary retention. CSF showed lymphocytic pleocytosis (140 cells/μL); infectious and autoimmune/demyelinating tests were negative. MRI revealed longitudinally extensive intramedullary hyperintensity from upper cervical cord to conus with leptomeningeal enhancement. Dengue IgM with IgM/IgG seroconversion confirmed acute infection. IV methylprednisolone (1 g/day) improved symptoms by day 5 and fully recovered within 1 year.

In endemic settings, neurodengue is a possibility when alternatives are excluded. Once infection is ruled out, immunotherapy should be considered.

Keywords: Dengue; Encephalopathy; Transverse Myelitis

RESUMO

A dengue é uma arbovirose frequente em regiões tropicais e subtropicais. Complicações neurológicas ocorrem em cerca de 5% dos casos, geralmente como encefalopatia secundária à doença sistêmica. Neurotropismo viral e lesão imunomediada são raros e podem mimetizar etiologias infecciosas e inflamatórias. Embora corticosteroides não sejam rotineiramente recomendados, relatos de caso descrevem seu uso em altas doses com desfechos favoráveis.

Relatamos o caso de um homem de 25 anos, previamente hígido, do Rio de Janeiro, que apresentou febre e cefaleia retro-orbitária, seguidas de dor abdominal, alucinações, ataxia de membros superiores e paraparesia arreflexa com nível sensitivo em T10 e retenção urinária. O líquor evidenciou pleocitose linfocitária (140 células/μL); suas investigações infecciosa, autoimune e desmielinizante foram negativas. A ressonância magnética mostrou hipersinal intramedular longitudinalmente extenso, da medula cervical alta ao cone medular, com realce leptomeníngeo. A positividade de IgM para dengue, com soroconversão IgM/IgG, confirmou infecção aguda. O tratamento com metilprednisolona intravenosa (1 g/dia) resultou em melhora até o 5º dia e recuperação completa em 1 ano.

Em áreas endêmicas, a neurodengue deve ser considerada após exclusão de diagnósticos alternativos. Uma vez afastada infecção, a imunoterapia deve ser considerada.

Palavras-chave: Dengue; Encefalopatia; Mielite Transversa

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INTRODUCTION

Dengue is a common arboviral infection in tropical and subtropical regions, with manifestations ranging from a self-limited febrile syndrome to severe disease with plasma leakage, hemorrhage, and organ dysfunction[1]. Neurological complications occur in up to 5% of cases most often as encephalopathy related to systemic involvement. Viral neurotropism and immune-mediated injury are rare and may ressemble infectious, inflammatory or demyelination disorders that can be severe and potentially fatal[2].

We report a rare case of acute dengue fever presenting with encephalopathy and longitudinally extensive transverse myelitis (LETM), with rapid improvement following high-dose corticosteroid therapy.

CASE REPORT

A previously healthy 25-year-old man from Rio de Janeiro presented with a one-week history of fever and retro-orbital headache, followed by hallucinations, abdominal pain, and urinary retention. On admission, he was febrile (38°C) and confused (Mini-Mental State Examination, 22/30; Glasgow Coma Scale, 14). Neurological examination revealed a T10 sensory level, upper-limb ataxia, and progressive areflexic paraparesis. Given the initial clinical presentation, dengue serologic testing was obtained.

Cerebrospinal fluid (CSF) analysis showed lymphocytic pleocytosis (140 cells/mm³) and mildly elevated protein levels, while infectious, autoimmune, and demyelinating tests were negative (Table 1).

TABLE 1 – Cerebrospinal fluid (CSF) and complementary diagnostic studies

Category	Test / Parameter	Result
CSF gross examination	Appearance	Colorless, clear
	Red blood cells	5 red blood cells per field
CSF routine analysis	White blood cells	140 cells/mm³
	Differential count	100% LMN cells
	Protein	67.2 mg/dL
	Glucose	43.6 mg/dL
	Adenosine deaminase	3 U/L
CSF microbiology	Bacterioscopy	Negative
	Direct mycological examination	Negative
	Bacterial cultures	Negative
	Mycobacterial culture	Negative
	GeneXpert (for *Mycobacterium tuberculosis*)	Negative
	Acid-fast bacilli smear	Negative
	India ink stain	Negative
CSF serologic / immunologic tests	VDRL	Negative
	TPHA	Negative
	CSF toxoplasmosis test	Negative
	Oligoclonal bands index	Negative
CSF multiplex PCR	*Escherichia coli* K1	Not detected
	Haemophilus influenzae	Not detected
	Listeria monocytogenes	Not detected
	Neisseria meningitidis	Not detected
	Streptococcus agalactiae	Not detected
	Streptococcus pneumoniae	Not detected
	Cytomegalovirus (CMV)	Not detected
	Enterovirus	Not detected
	Herpes simplex virus 1 (HSV-1)	Not detected
	Herpes simplex virus 2 (HSV-2)	Not detected
	Human herpesvirus 6 (HHV-6)	Not detected
	Human parechovirus	Not detected
	Varicella-zoster virus (VZV)	Not detected
	Cryptococcus neoformans/gattii	Not detected
Serum studies	Anti-aquaporin-4 antibody	Negative
	Anti- MOG antibody	Negative
	Antinuclear Antibody	Negative
	Rheumatic Factor	Negative
	HIV serology	Negative
	HTLV serology	Negative

Spinal magnetic ressonance imaging (MRI) demonstrated longitudinally extensive intramedullary T2 hyperintensity extending from the upper cervical cord to the conus medullaris, with associated leptomeningeal enhancement (Figures 1-3). Dengue IgM positivity, followed by subsequent IgM/IgG seroconversion, confirmed acute infection.

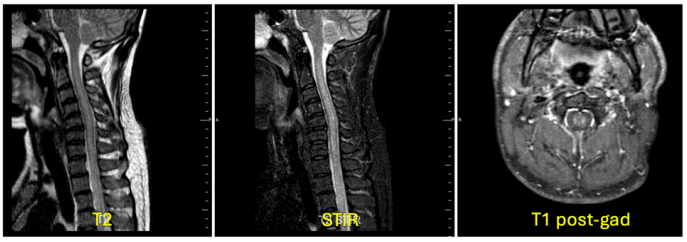

FIGURE 1 – MRI showing a predominantly T2/STIR hyperintense intramedullary signal. Post-contrast sequences demonstrate leptomeningeal enhancement.

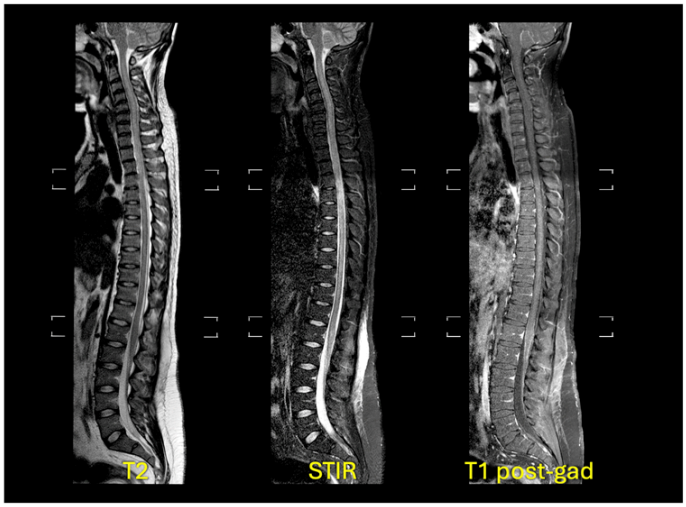

FIGURE 2 – Sagital view of spine and neuroaxis MRI showing extensive hipermedullary signal and leptomeningeal enhancement.

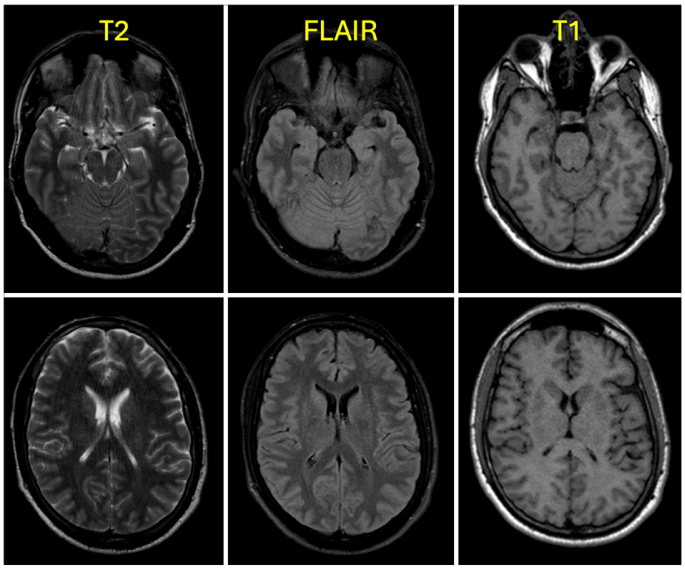

FIGURE 3 – Brain MRI with no major findings

High-dose intravenous corticosteroid therapy was initiated, with marked clinical improvement observed within three days. After five days of treatment, he was able to walk and had complete resolution of the ataxia. He was discharged after two weeks with residual neurogenic bladder, which had fully resolved by the 6-month follow-up.

DISCUSSION

Neurological manifestations of dengue are increasingly reported but remain underrecognized. Encephalopathy is the most common presentation and is usually related to systemic complications such as hepatic failure, hyponatremia, or shock. Direct viral neurotropism and immune-mediated mechanisms may lead to more severe syndromes, including encephalitis, demyelination, and transverse myelitis[3]. Our patient fulfilled WHO criteria[4] for dengue with neurological involvement, supported by seroconversion and CSF lymphocytic pleocytosis with mild hyperproteinorrachia. The absence of systemic complications supports a neurotropic mechanism.

Acute disseminated encephalomyelitis (ADEM) has classically been associated with dengue, particularly in younger patients[5]. However, despite the lack of a formal adult dianostic criteria, diffuse demyelinating brain lesions are expected[5] and are included in the International Pediatric Multiple Sclerosis Study Group (IPMSSG) consensus criteria for the pediatric population[6]. Their absence in our patient makes ADEM less likely and instead favors a dengue-related neurological syndrome mediated by direct viral neurotropism and/or immune mechanisms.

Although the true incidence of dengue-associated LETM is unknown, it is generally considered an exceedingly rare complication. While one Brazilian outbreak reported myelitis in more than half of neurological dengue cases[7], most studies estimate it accounts for only 1-5% of neurological manifestations[8]. In our review, we identified less than 40 reported cases of dengue-associated transverse myelitis, only a minority of which were longitudinally extensive[8,9], and none combined LETM with such a prominent encephalopathic presentation.

Most reported cases occurred during the acute or para-infectious phase, although delayed presentations have been described. Outcomes were heterogeneous, ranging from full recovery to permanent disability and death. Still, about half of patients achieved complete recovery over weeks to months, and overall mortality appeared to be low[10].

Early administration of high-dose corticosteroids has been proposed as a potentially beneficial strategy in dengue-associated demyelinating syndromes, possibly by modulating the neuroinflammatory response and favoring rapid clinical improvement and complete recovery[3,8]. The prompt and marked response observed in our patient is consistent with this hypothesis. Nevertheless, the evidence is still limited to observational reports, and spontaneous recovery cannot be ruled out.

CONCLUSION

Dengue-associated LETM with encephalopathy is a rare but potentially life-threatening neurological syndrome. Although the evidence is still limited, published reports suggest that dengue-related neurological complications may be reversible with early recognition and timely immunomodulatory treatment. This report underscores the importance of maintaining a high index of suspicion for dengue-related neurological involvement in endemic settings and of considering corticosteroids when a severe neuroinflammatory presentation is suspected and alternative diagnoses have been carefully excluded.

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