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Facial Diplegia Associated with a Reversible Splenial Lesion due to Influenza B Viral Infection

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Abstract

Introduction— Influenza-associated encephalitis/encephalopathy includes a wide spectrum of syndromes reported often in children. A rare form presents with mild encephalitis and reversible splenial lesion(s). In this report, we present a rare case of facial diplegia as the main manifestation of this clinical disorder.

Case Report— A 23-year-old man presented with acute onset of speech difficulty and mild upper and lower facial asymmetric weakness and eye-closure difficulty bilaterally. Motor examination revealed a pronator drift bilaterally. Cerebrospinal fluid analysis revealed mild lymphocytic pleocytosis. Magnetic resonance imaging of the brain revealed areas of hyperintensities and diffusion restriction in the corpus callosum and bilateral centrum semiovale. Serologic testing via Reverse Transcriptase - Polymerase chain reaction revealed influenza B infection. The patient was treated with intravenous methylprednisolone with significant clinical improvement. Magnetic resonance imaging of the brain 4 weeks after admission revealed complete resolution of the previously seen hyperintensities.

Conclusion— We present the first case of mild encephalitis and reversible splenial lesion triggered by influenza B infection with facial diplegia as the main clinical manifestation.

Keywords

Influenza B, Influenza-associated encephalitis, Influenza-associated encephalopathy, reversible splenial lesion..

INTRODUCTION

Influenza-associated encephalitis/encephalopathy includes a series of heterogeneous clinic-radiologic syndromes reported frequently in children from East Asia.[1] Mild encephalitis/encephalopathy with reversible splenial lesion is a rare form of influenza-associated encephalitis/encephalopathy that has been reported in association with a variety of infections and metabolic disorders and carries an excellent prognosis.[2] This variant is characterized by a monophasic course of mild encephalopathy and neurological symptoms, minimal pleocytosis in the cerebrospinal fluid, and magnetic resonance imaging finding of a reversible lesion in the splenium of the corpus callosum.[3] Mild encephalitis/encephalopathy with reversible splenial lesion is classified as type I or type II, depending on the extent of central nervous system involvement.[4] Type I typically involves a singular lesion in the midline of the splenium of the corpus callosum and is the more common form. Type II typically presents with symmetrical lesions in the cerebral white matter or anterior aspect of the corpus callosum with similar signal manifestations as type I and is more frequently reported with influenza type A infection.[5] Trigeminal neuralgia, unilateral facial palsy, and olfactory disturbance have been reported in patients with mild encephalitis/encephalopathy with reversible splenial lesion.[6,7] We report a case of a young adult who developed acute bilateral facial diplegia as the chief complaint following influenza B infection and was diagnosed with type II of mild encephalitis/encephalopathy with reversible splenial lesion.

CASE REPORT

A 23-year-old right-handed Asian man with no significant medical history presented to the Emergency Department with acute onset of speech difficulty. Review of systems revealed a productive cough for 3 days, fatigue, generalized weakness, and difficulty swallowing. One day prior, the patient was treated with a local anesthetic before a root canal procedure.

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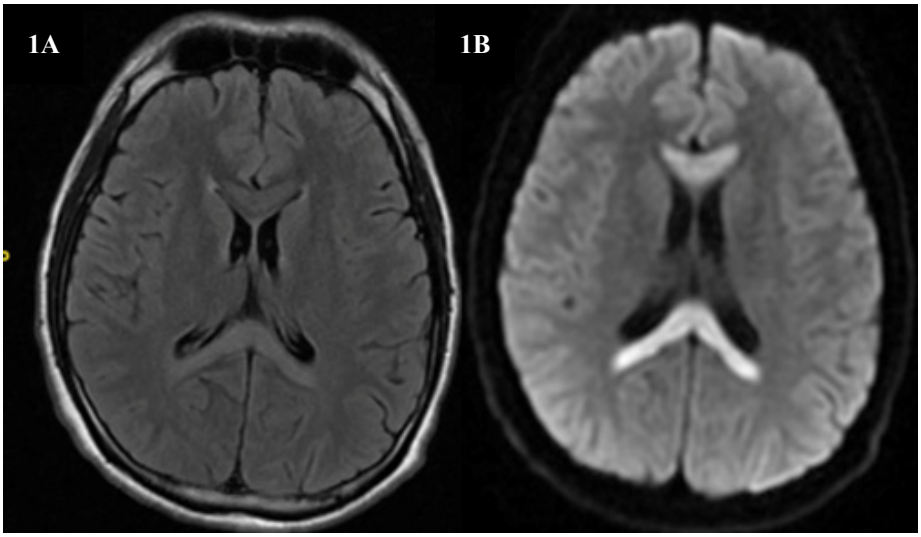

FIGURE 1: Hyperintensity on T2- fluid-attenuated inversion recovery sequence (A) and restricted diffusion (B) in the corpus callosum.

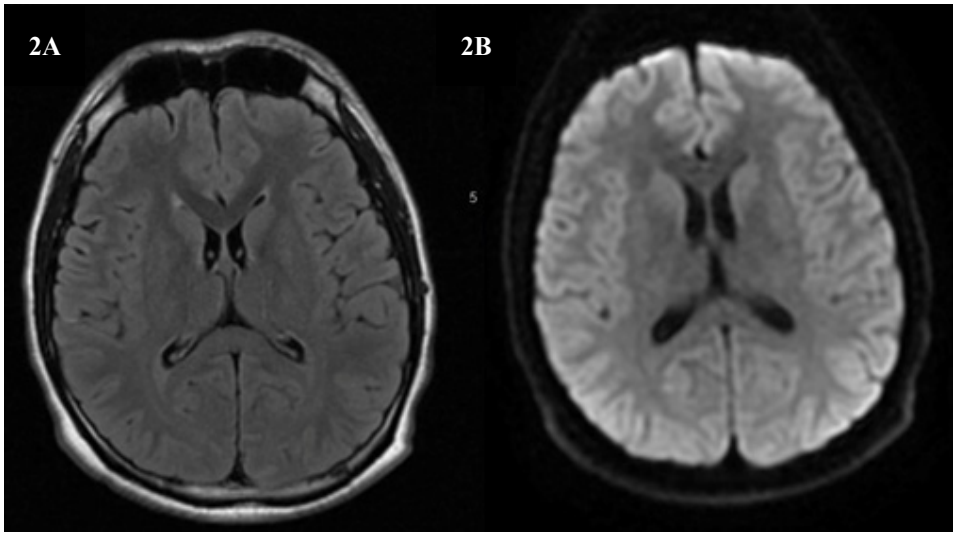

FIGURE 2: Complete resolution of T2- fluid-attenuated inversion recovery hyperintensity (A) and restricted diffusion (B) in the corpus callosum on repeat imaging four weeks after initial presentation.

Five hours prior to arrival to the Emergency Department, he had an acute onset of difficulty speaking and was unable to chew or swallow food.

Upon arrival, the patient was afebrile with a temperature of 98.5°F. Initial neurologic examination revealed unremarkable cranial nerve examination except for mild upper and lower facial asymmetric weakness and eye-closure difficulty bilaterally. Intermittent drooling was observed. Palatal movement was mildly reduced. The patient was unable to speak but was able to phonate some sounds with mild dysarthria. Speech repetition and comprehension were intact, and he was able to follow one- and two-step commands. Motor examination revealed a subtle pronator drift and mild weakness in grip strength bilaterally. Sensory, coordination, and gait examination was unremarkable.

Laboratory work-up included a complete blood count with a normal white blood count of 3.5 x 10^9/L. Comprehensive metabolic panel and urine drug screen were unremarkable. Further work-up revealed influenza B infection via RT-PCR. Cerebrospinal fluid analysis revealed mild lymphocytic pleocytosis with a white blood cell count of 5/mm^3 (lymphocytes 64%, monocytes 36%), red blood cell count of 4/mm^3, protein 35 mg/dl, and glucose 90 mg/dl. Additional studies performed on cerebrospinal fluid including cultures and gram stain were unrevealing.

Computed tomography of the head was unremarkable. One day after admission, the patient developed worsening of facial diplegia and difficulty with eyes closure. He was unable to perform single breath counts due to inability to maintain inspiratory position. Examination also revealed mild bilateral hyperreflexia and a transitory subtle finger-to-nose ataxia bilaterally. Magnetic resonance imaging of the brain with and without gadolinium showed hyperintensities on fluid-attenuated inversion recovery sequence and focal restriction on diffusion-weighted imaging in the corpus callosum (Figures 1A and 1B) and bilateral centrum semiovale (images not shown), with no evidence of pathological enhancement. Magnetic resonance imaging of the cervical spine was unremarkable.

The patient was treated with 1 gram of intravenous methylprednisolone daily for 3 days. Within 72 hours of admission, the facial diplegia completely resolved.

Upon discharge on day 7 of hospitalization, neurological examination was unrevealing except for residual subtle hyperreflexia bilaterally. Magnetic resonance imaging of the brain four weeks after admission revealed complete resolution of the corpus callosum (Figure 2A & 2B) and centrum semiovale (images not shown) hyperintensities and restricted diffusion.

Outpatient evaluation 6 weeks after discharge revealed no change in the neurological examination and no residual facial weakness.

DISCUSSION

The clinical spectrum of influenza-associated central nervous system involvement includes Reye Syndrome, acute necrotizing encephalopathy, and other types of influenza-associated encephalitis/encephalopathy.[8] Since the first report by Tada et al, the clinical presentation of mild encephalitis/encephalopathy with reversible splenial lesion expanded from encephalopathy, seizures, hallucinations, ataxia, and headache to include focal weakness, dysarthria, gastrointestinal symptoms, ophthalmoplegia, and olfactory and trigeminal dysfunction.[3,6] In this case report, we present the first upper motor neuron syndrome of facial diplegia as the chief clinical manifestation in an adult patient with this condition. Mild encephalitis/encephalopathy with reversible splenial lesion has been associated with multiple infectious agents other than the influenza virus, including the rotavirus, measles, mumps, O-157 Escherichia coli, legionella, mycoplasma pneumonia, and salmonella enteritidis.[7,9]

Serial diffusion tensor imaging in our patient showed normal fractional anisotropy values upon resolution of the splenial lesion, indicating that mild encephalitis/encephalopathy with reversible splenial lesion was most likely due to transient interstitial edema with preservation of white matter tracts.[10] The lack of detection of the influenza virus in the central nervous system favors an inflammatory cytokine-mediated response rather than direct virus invasion leading to brain–blood barrier dysfunction causing cerebral edema of varying intensity.[11] Among these cytokines, serum interleukin-6 levels are well-correlated with clinical severity and prognosis, making it a good predictive marker for influenza-associated encephalitis/encephalopathy.[12] Kawada et al reported significant up-regulation of the transcription level of interleukin-6 and tumor necrosis factor-α in patients with influenza-associated encephalitis/encephalopathy.[13] Other anti-inflammatory cytokines titers have been identified including IL-10 during the early stages of viral-induced mild encephalitis/encephalopathy with reversible splenial lesion that may contribute to localized anatomical damage and prevent sequela.[14] Nevertheless, this is not a common trend in all reported cases.[8]

In conclusion, we present the first case of a patient with mild encephalitis/encephalopathy with reversible splenial lesion triggered by influenza B viral infection who presented with facial diplegia as the main manifestation. This case expands the clinical spectrum of influenza-associated encephalitis/encephalopathy with transient pure motor syndrome as the chief clinical presentation paralleled by reversible splenial and extracallosal involvement. Further investigation is imperative for a better understanding of the pathophysiology and management of this entity in order to avoid unnecessary invasive diagnostic and therapeutic interventions.

FINANCIAL DISCLOSURES

None.

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