

Subacute measles encephalitis in a case of late presenting congenital HIV with epilepsy partialis continua as the first manifestation: A case report

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ABSTRACT

We report a case of a 14-year-old non-immunised girl with a prior history of measles infection presenting with afebrile seizures progressing to epilepsy partialis continua (EPC), quadriparesis and headache. Further evaluation revealed Human immunodeficiency virus (HIV) seropositivity with elevated anti-measles antibody titres in Cerebrospinal fluid (CSF). Electroencephalography showed focal epileptiform activity and Magnetic resonance imaging (MRI) of the brain revealed bilateral, asymmetrical long repetition time MRI (TR) hyperintensities involving juxtacortical white matter in both parietal lobes, left temporal and also in the left basal ganglia without any contrast enhancement or Diffusion weighted imaging (DWI) restriction. We describe the intriguing association of EPC with subacute measles encephalitis/measles inclusion body encephalitis (SME/MIBE) in the backdrop of immunocompromised state (HIV seropositivity), thought to have been acquired by vertical transmission. Also, prolonged asymptomatic HIV infection, first unmasked by measles infection, followed by rapidly deteriorating neurological illness makes this index case worthy to be reported.

Keywords: Epilepsia partialis continua, HIV, measles encephalitis, MIBE

Introduction

Measles is associated with diverse complications, e.g., pneumonia, otitis media, hepatitis, and central nervous system (CNS) involvement. The most frequent neurological complication is encephalitis^[1] which includes acute disseminated encephalomyelitis (ADEM), subacute measles encephalitis or measles inclusion body encephalitis (SME/MIBE) and subacute sclerosing panencephalitis (SSPE). Immunocompromised patients are particularly prone to develop SME/MIBE due

to incomplete clearance of the measles virus after primary Central nervous system (CNS) infection.^[2] Here, we report a rare case of subacute measles encephalitis in a 14-year-old immunocompromised girl, highlighting late presenting congenital HIV and various manifestations of CNS measles infection. This echoes the importance of detection and prevention of mother to child transmission (PMTCT) and vaccination to internists and primary care physicians.

Case Report

Ethical and institutional clearance was taken for this report. A 14-year-old female apparently well before the present illness, presented with a history of fever and maculopapular rash 3 months ago, diagnosed as measles at the primary care level. Her immunisation status was incomplete as per the mother. She

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was treated conservatively at home and recovered within the next 10 days. One and half months later, she developed recurrent episodes of tonic-clonic seizures involving the left half of the body and face followed by progressive left hemiparesis. Subsequently, the episodic attack of the left focal seizure became continuous along with a dull-aching holocranial headache. Ten days preceding admission, she started having a progressive right hemiparesis. Throughout this course, she remained alert and conscious.

Her premorbid mental state including age-adjusted cognitive maturity was normal. A general survey revealed normal vitals, generalised wasting and oral thrush. She had a Mini-Mental State Examination score of 27, without any behavioural abnormality. Deep tendon reflexes were present in the upper limbs and absent in the lower limbs. upper motor neuron (UMN)-type right facial palsy, left-sided epilepsy partialis continua (EPC) and quadriplegia were detected.

The total blood count showed pancytopenia (haemoglobin 10.1 g%, total leukocyte count: 2400/ μ L, platelet count: 1 lakh/cc). The serum electrolytes, hepatic, renal and thyroid function tests, including anti-thyroid peroxidase antibody levels, chest X-ray and ultrasonography of the whole abdomen were within normal limits. Serology for HIV-1 was positive with a CD4 T-cell count of 97 cells/ μ L. The patient's mother also tested HIV positive. (The patient's father died of uncertain causes 10 years ago.) The CSF study including a cartridge based nucleic acid amplification test (CBNAAT) test was unremarkable. The autoimmune encephalitis panel, antinuclear antibody (ANA) profile (using Hep-2 cells) were negative. CSF culture, polymerase chain reaction assay for JC virus and Epstein-Barr virus and serology for (Toxoplasma, Rubella, Cytomegalovirus, Herpes) TORCH organisms, were normal. An elevated CSF anti-measles Immunoglobulin gamma (IgG) antibody quotient was detected. Electroencephalography noted focal epileptiform activity. MRI of the brain revealed bilateral, asymmetrical long

TR hyperintensities involving juxtacortical white matter in both parietal lobes, left temporal and also in the left basal ganglia without any contrast enhancement or DWI restriction [Figure 1].

Her EPC were refractory to antiepileptic drugs, antiretroviral therapy; with a rapid downhill course, and ultimately, the patient succumbed to death within 2 weeks of hospital stay.

Discussion

Differentials of EPC in an immunocompromised patient can be opportunistic CNS infections, mainly viral, tubercular, fungal etiologies, CNS lymphoma, autoimmune (especially Rasmussen's encephalitis), CNS vasculitis and structural causes. A history of measles 3 months back made us consider measles-related encephalitis^[1] initially. The absence of characteristic findings of other infective pathology on MRI, absence of infarcts or haemorrhages, a normal brain MR angiography, poor response to intravenous immunoglobulin (IVIG) and a normal CSF study weighed against the other differentials. Measles-related encephalitides exists in four variations^[1]: primary measles encephalitis, acute post-measles encephalitis, MIBE and SSPE. Though brain biopsy is confirmatory for diagnosis; a history of measles within 7 months in an unimmunised, immunocompromised patient with refractory seizures with or without generalisation, absence of fever and a normal CSF study is highly suggestive of MIBE.^[3] An elevated CSF measles IgG antibody quotient along with negative Epstein Barr virus deoxyribonucleic acid (EBV DNA), JC virus polymerase chain reaction (PCR) and negative TORCH screening and autoimmune encephalitis profile helped strengthen our diagnosis.

Immunocompromised patients who fail to clear the measles virus after a primary CNS infection mainly suffer from MIBE.^[2] A wide array of clinical features like afebrile focal seizures (97%), hemiplegia (36%), aphasia (24%), ataxia (24%), altered mental status (100%)^[2,3] are found in MIBE. The CSF is usually normal with the detection of the measles virus antibody in 50% of the cases.^[3] The MRI findings are usually normal in the beginning,^[2] but, similar to a case of MIBE in an immunocompromised boy reported previously,^[4] our patient had multiple, non-enhancing, bilateral, asymmetric T2 hyperintensities in the brain involving both white and grey matter.

MIBE and SSPE can be argued to be the part of a spectrum where typically immunocompetent patients develop SSPE and the immunodeficients develop MIBE. The recent measles outbreak in Ukraine (2017–2019) showed transformation into MIBE in a child with measles and acute leukaemia.^[5] Pathologically, inclusion bodies have been found to be deposited in the diffuse cerebral cortex, brainstem, paraventricular area and mammillary bodies in MIBE.^[6] The incubation period of SSPE is generally much longer than MIBE. In an index patient, the relatively short time frame of the disease, retained cognitive function, presence of the immunocompromised state in the

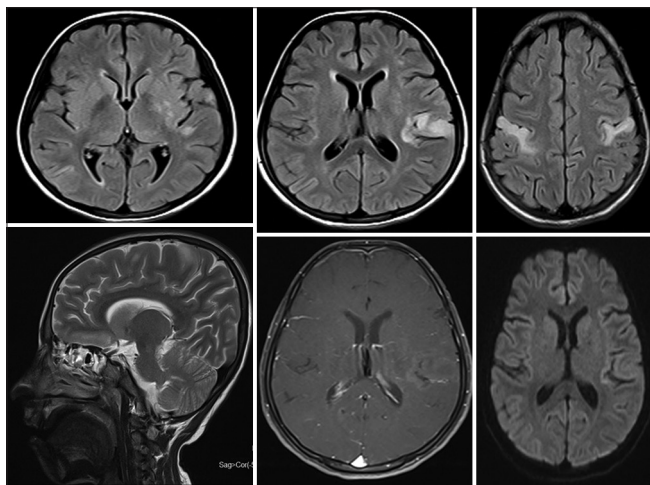


Figure 1: Bilateral, asymmetrical long TR hyperintensities involving juxtacortical white matter in both parietal lobes, left temporal and also in the left basal ganglia without any contrast enhancement or DWI restriction.

form of HIV seropositivity, aforesaid MRI findings and lack of electroencephalogram (EEG) changes of SSPE pointed towards the former entity in the spectrum.

The intriguing phenomenon about this case is the sudden clinical decompensation of congenital HIV, which the patient harboured asymptotically for 14 years, unmasked by measles. Though, the incidence of congenital HIV has reduced significantly due to effective PMTCT, most patients harbouring vertical infection present by 3–5 years of age.^[7] It is rare for congenital HIV to present this late,^[8] with the first manifestation being EPC followed by a deteriorating neurological illness. Earlier, there were just a handful of case reports notably of cytomegalovirus encephalitis^[9] and progressive multifocal leukoencephalopathy^[10] in HIV presenting with EPC.

To conclude, EPC can be a presenting feature of HIV acquired immunodeficiency syndrome (AIDS). Measles in an immunocompromised patient can have devastating consequences. This case also heralds the importance of ensuring universal vaccine coverage and early HIV detection in children to prevent further complications.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

References

1. Fisher DL, Defres S, Solomon T. Measles-induced encephalitis. *QJM* 2015;108:177-82.
2. Buchanan R, Bonthius DJ. Measles virus and associated central nervous system sequelae. *Semin Pediatr Neurol* 2012;19:107-14.
3. Mustafa MM, Weitman SD, Winick NJ, Bellini WJ, Timmons CF, Siegel JD. Subacute measles encephalitis in the young immunocompromised host: Report of two cases diagnosed by polymerase chain reaction and treated with ribavirin and review of the literature. *Clin Infect Dis* 1993;16:654-60.
4. Freeman AF, Jacobsohn DA, Shulman ST, Bellini WJ, Jaggi P, de Leon G, *et al.* A new complication of stem cell transplantation: Measles inclusion body encephalitis. *Pediatrics* 2004;114:e657-60.
5. Lytvyn H, Basa N, Stasiv M, Troyanovska O, Dorosh O. Difficulties in diagnosing of measles inclusion body encephalitis in a child with acute lymphoblastic leukaemia. *IDCases* 2020;21:e00877.
6. Aldecoa I, Archilla I, Herrero L, Garcia F, Torres B, Gaig C, *et al.* Measles inclusion body encephalitis. *Clin Neuropathol* 2020;39:148-51.
7. Abbas M, Bakhtyar A, Bazzi R. Neonatal HIV. [Updated 2021 May 4]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2021. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK565879/>.
8. Meyers A. Natural history of congenital HIV infection. *J Sch Health* 1994;64:9-10.
9. Ramanujam B, Dash D, Dabla S, Tripathi M, Srivastava MV. Epilepsia partialis continua as presenting manifestation of AIDS: A rarity. *J Int Assoc Provid AIDS Care* 2016;15:19-22.
10. Ferrari S, Monaco S, Morbin M, Zanusso G, Bertolasi L, Cerini R, *et al.* HIV-associated PML presenting as Epilepsia Partialis Continua. *J Neurol Sci* 1998;161:180-4.