Neuromeliodosis—A Tropical Illness Presenting as Meningoencephalitis with Unusual Brain Imaging

Sir,

A 56-year-old lady, hailing from a coastal area in Western India, a known case of diabetes mellitus, presented with fever and dry cough for 1 week. She was given a short course of antibiotics and symptomatic treatment at a local hospital, following which her symptoms resolved. A week later, she once again began spiking fever. She also noticed the acute weakness of the right upper and lower limb which affected her ambulation, and she was referred to our tertiary care center.

On initial evaluation, she was febrile, pale with tachycardia. Her neurological examination revealed the power of 3 (MRC Grading) in the right upper and lower limb with diminished reflexes on the right side with an extensor plantar response. The respiratory exam revealed basal crepitations. Musculoskeletal examination revealed swelling of the right knee that was erythematous and tender.

Her routine laboratory investigations revealed hemoglobin of 8.1 g, raised total leukocyte count of 21,000 with a neutrophilic predominance and adequate platelets. Peripheral smear showed a microcytic hypochromic picture. Blood glucose levels were raised with a fasting value of 196 mg/dL and HbA1C of 11. Blood and Urine cultures were negative. X-ray chest was normal. Contrast-enhanced CT Brain revealed hypodensity in the left parieto-occipital region.

Over the next week, her sensorium deteriorated and she was only able to obey simple commands. She also developed the right focal seizures. As a result, her clinical examination and imaging findings were revisited. Her antibiotics were escalated, and Levetiracetam was prescribed to control seizures. CSF analysis showed mildly raised proteins but no cells. CSF culture was negative. MRI of the brain [Figure 1] with contrast imaging showed cortical T2 FLAIR hyperintensity in the left parieto-occipital region and subcortical white matter with diffusion restriction and pachymeningeal enhancement suggestive of focal encephalitis. MRI brain angiogram was normal.

At this point, the swollen knee joint was reexamined and ultrasound-guided synovial fluid aspiration was done. The synovial fluid analysis showed a leukocyte count of 1,660 with neutrophilic predominance and proteins of 1 g/100 mL and occasional Gram-negative bacilli. The synovial fluid culture showed a growth of *Burkholderia pseudomallei* which was sensitive to Meropenem. Considering these findings, a diagnosis of NeuroMeliodosis was made.

She was started on a 6-week course of Meropenem at a dose of 1 g every 8 h, with a plan to maintain on Trimethoprim-Sulfamethoxazole for a year. Over the course of the next 6 weeks, she showed gradual clinical improvement and improved sensorium. At discharge, she was conscious, oriented, with the power of 4 (MRC Grading) on the right side. Her joint swelling resolved and there was satisfactory glycemic control. Imaging findings also showed a resolution.

B. pseudomallei is a Gram-negative, facultative, soil saprophyte responsible for the disease. Melioidosis is contracted by inoculated soil and water through wounds or inhalation and is particularly common during and after the wet season.[1] The disease is endemic to areas of the Indian subcontinent, Southeast Asia, and territories of Australia.[2] Comprising approximately 4% of all cases of melioidosis, neurologic melioidosis has a mortality rate of approximately 25% and survivors have significant morbidity.

The pathogenesis of CNS melioidosis includes the hematogenous spreading of microorganisms to the CNS via following routes: Transcellular, paracellular, Trojan horse method via L-selectin (CD62L)-mediated migration,[3] direct brainstem invasion, and percutaneous inoculation.

NeuroMelioidosis can present as encephalomyelitis, brain abscess, Guillain-Barre syndrome, seizures, isolated meningitis or the isolated extra-axial collection, and focal neurological deficits especially cranial nerve palsies.[4] Presentation with fever is more prevalent in melioidosis (74%).

The CSF profile of CNS melioidosis commonly shows pleocytosis with mononuclear cell predominance. Contrast-enhanced MRI shows brainstem lesions in the encephalomyelitis type. One study shows a propensity to involve and spread along white matter tracts across the commissural or longitudinal fibers.[5] Three patients in this study showed supratentorial lesions similar to our patients.

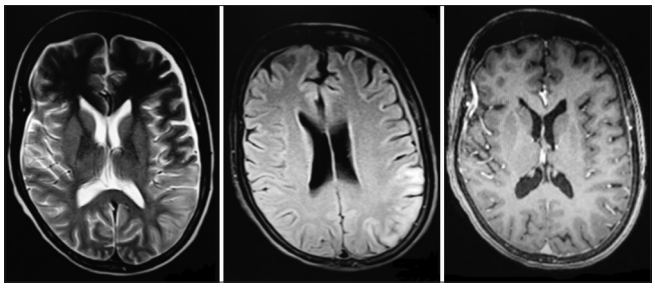

Figure 1: Left—cortical T2-weighted hyperintensity is seen in the left parieto-occipital region with mild hyperintensity seen in underlying subcortical white matter. Center—cortical FLAIR hyperintensity is seen in the left parieto-occipital region with mild hyperintensity seen in underlying subcortical white matter. Right—diffuse pachymeningeal contrast enhancement seen

Culture is essential for definitive diagnosis. As brain biopsy may not be feasible, the diagnosis can be made by presentation of compatible CNS diseases supported by positive cultures from other body fluid specimens.

According to the 2010 consensus recommendations, ceftazidime and meropenem are the drugs of choice for intensive-phase therapy, while trimethoprim/sulfamethoxazole is the first-line drug for eradication-phase therapy.[6] Eight weeks and six months are the minimum duration for intensive- and eradication-phase therapy, respectively.

There are very few case reports of NeuroMeliodosis in India,[7] with some authors describing its presentation as cerebral abscesses.[8,9] This case proves that in tropical countries like India, especially in patients residing in coastal areas, a high index of suspicion along with knowledge of varied presentations and unique imaging findings are key to the management of NeuroMeliodosis. Educating patients about footwear and sanitation would be crucial in the prevention and spread of the disease.

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.

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