

## Neuromelioidosis Presenting as Bells Palsy in a Child

Sir,

Melioidosis is a tropical infection caused by *Burkholderia pseudomallei* and is endemic in South East Asia and Northern Australia.<sup>[1]</sup> Central nervous system melioidosis is rare in children and only a few cases have been reported in the literature.<sup>[2]</sup> It can present as meningoencephalitis, cerebral abscess, myelitis, cranial nerve palsies, and can mimic Guillain Barre Syndrome.<sup>[3]</sup> We report an interesting case of neuro melioidosis presenting as lower motor neuron type of facial palsy who proceeded to develop third nerve palsy, hemiparesis, and meningeal signs.

An 8-year-old healthy female child presented with a deviation of angle of mouth to the right side for 4 days. She had difficulty in closing her left eye in addition. She had a low-grade fever for 2 days, a week ago. There was no history of headache, nuchal pain, vomiting, seizures, altered sensorium, weakness of limbs, or unsteadiness while walking, and no history of trauma, ear discharge, recent exanthem either. She had a habit of playing in pooled water and soil near her house.

On examination, she had left LMN facial palsy with no other deficit. A diagnosis of Bell's palsy was made. She was prescribed oral steroids and advised physiotherapy. However, because of low-grade fever, neuroimaging was planned. Parents decided to undergo neuroimaging a week later due to financial reasons. Four days later, she developed a high-grade fever associated with headache and vomiting and got admitted to our institution.

Examination revealed a conscious child with left LMN facial palsy, normal tone and power in all four limbs, preserved deep tendon reflexes, and flexor plantars. Other cranial nerves were intact. There were no cerebellar signs. She had neck stiffness and a positive Kernig sign. Fundi were normal. She was started on a meningitic dose of ceftriaxone and hypertonic saline. Hematological workup revealed neutrophilic leucocytosis, high C reactive protein, and normal renal and liver function tests. Blood culture was sterile. CSF was clear, acellular, with normal biochemistry, negative gram staining, and sterile culture. CSF AFB stain and GeneXpert MTB/RIF were negative. HIV serology was negative. MRI brain revealed hyper-intense lesions in the left side of the medulla, pons, midbrain, and along the posterior limb of the left internal capsule [Figure 1a–c]. A diagnosis of neuro melioidosis was made based on classical imaging findings, and the antibiotic was switched over to meropenem. Oral trimethoprim/sulfamethoxazole (TMP-SMX) was added.

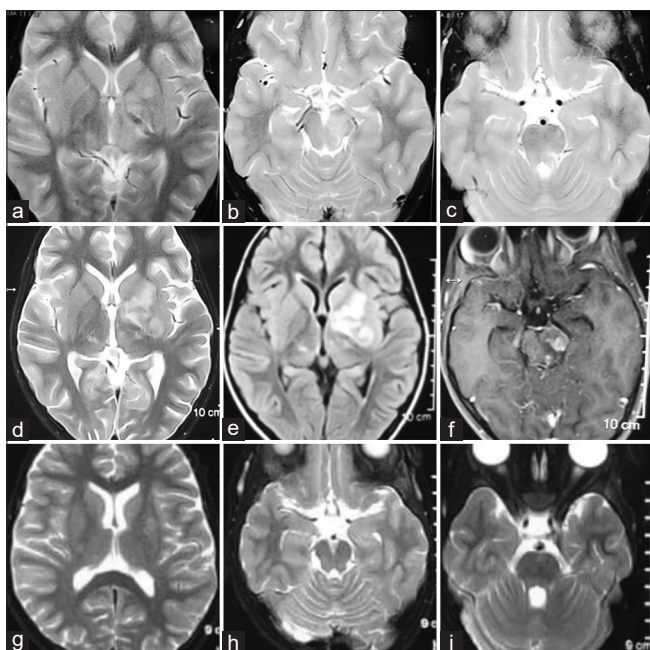
On the seventh day of hospital stay, she developed partial ptosis in the left eye and right hemiparesis in addition to left LMN facial palsy. Pupils were equal reacting to light and extraocular movements were full. Fever spikes continued despite meropenem. Blood culture was negative for *Burkholderia*.

Repeat imaging revealed extensive lesions in the brainstem, left basal ganglia, and internal capsule. The lesion in the midbrain showed intense contrast enhancement [Figure 1d–f]. Because of persistent fever and a new neurological deficit, ceftazidime was added. Repeat CSF analysis was not contributory, including culture for *Burkholderia* species. CSF fungal smear and culture were negative. The role of empirical antituberculous therapy was considered but deferred as the imaging findings were suggestive of neuro melioidosis. Fever subsided four days after the addition of ceftazidime. Ptosis, facial lag, and hemiparesis improved over a span of 2 weeks. Parenteral antibiotics were continued for 6 weeks. She was discharged with a plan of eradication therapy with oral TMP-SMX for 1 year. Repeat imaging at the end of 3 months showed complete resolution of the lesions [Figure 1g–i]. Six months post-discharge, she was asymptomatic and on oral TMP-SMX.

Melioidosis is a potentially fatal disease caused by a motile, non-acid-fast, gram-negative bacillus, *Burkholderia Pseudomallei*.<sup>[1]</sup> The organism is an environmental saprophyte, found predominantly in the soil and groundwater sources during the rainy season. Infection usually occurs via percutaneous inoculation, ingestion, or inhalation.<sup>[4]</sup> The incidence of central nervous system (CNS) involvement in melioidosis ranges from 3% to 10%.<sup>[5]</sup> The pathogenesis of neuro melioidosis is postulated to be due to direct bacterial invasion or due to hematogenous spread.<sup>[6]</sup>

Pediatric neuro melioidosis is very rare and meningoencephalitis is the most common presentation. Cranial nerve palsies and fever are prominent presenting features,<sup>[7]</sup> the commonest cranial nerve affected being the facial nerve.<sup>[8]</sup> Our patient initially presented as unilateral facial palsy along with low-grade fever. She developed high-grade fever, partial third nerve palsy, and hemiparesis in a week. Neuroimaging provided a clue to the diagnosis. Brainstem neurotropism and propensity for spread along the white matter tracts across the commissural or longitudinal fibers is the hallmark of neuro melioidosis, especially the encephalomyelitis type.<sup>[9,10]</sup> MRI brain of our patient showed hyper-intense lesions in the medulla, pons, midbrain, and along the posterior limb of the internal capsule.

Culture from the infected tissue is the gold standard for the diagnosis of melioidosis. Although it is 100% specific, sensitivity may be as low as 60%. The blood culture yields a positive result in the highest sensitivity with 52%.<sup>[11]</sup> The peculiarity of neuro melioidosis is the absence of CSF spillage of the organism as the organism is largely confined to the white matter fiber tracts; thus, isolation from CSF may not always be possible.<sup>[12]</sup> Blood and CSF cultures were sterile in our patient, probably due to low sensitivity and prior antibiotic therapy.



**Figure 1:** First row: a, Axial T2W MR image showing hyperintensity along posterior limb of left internal capsule. b, hyperintensity in left midbrain. c, in the pons. Second row: d, MRI brain repeated 10 days after the first imaging shows extensive involvement of left basal ganglia. e, Axial FLAIR image showing involvement of basal ganglia. f, Contrast MR showing enhancement in midbrain. Third row: Repeat MRI two months later showing resolution of lesions in g, basal ganglia. h, in the midbrain. i, in the pons.

The indirect hemagglutinin assay (IHA) is not reliable for diagnosis because of its problematic false positive and false negative results<sup>[13]</sup> and hence not done in our patient.

Neuro melioidosis needs a prolonged treatment for a complete cure of the disease. *B. pseudomallei* are resistant to aminoglycosides, penicillin, first and second-generation cephalosporins, ampicillin, and polymyxin. Treatment consists of an intensive phase with ceftazidime or carbapenem plus TMP/SMX for a minimum of 4 weeks, followed by an oral eradication phase with TMP/SMX plus/or doxycycline for a minimum of 6 to 12 months.<sup>[3]</sup> Neuro melioidosis is associated with high mortality and morbidity if not treated properly. In their review article on CNS melioidosis in the pediatric age group, Prasad *et al.*<sup>[7]</sup> quoted an overall mortality rate of 18.5% and incomplete recovery of 30%. Timely institution of appropriate antibiotics would help in reducing mortality and eradication therapy for recommended duration would help in reducing relapses.

In conclusion, neuro melioidosis, the mimicker of maladies can masquerade as Bell's palsy. The propensity for spread along the white matter tracts and brainstem neurotropism can help to establish the diagnosis. A high index of suspicion and prompt institution of appropriate antibiotics would improve the overall outcome.

### 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.

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

**Leema P. Cornelius, Neeraj E, Venkateswaran KJ, Gopinathan K<sup>1</sup>**

Department of Paediatric Neurology, Institute of Child Health and Hospital for Children, Madras Medical College, <sup>1</sup>Department of Radiology, Govt. Royapettah Hospital, Kilpauk Medical College, Chennai, Tamil Nadu, India

**Address for correspondence:** Dr. Leema P. Cornelius, Department of Paediatric Neurology, Institute of Child Health and Hospital for Children, Egmore, Chennai, Tamil Nadu, India.  
E-mail: leema\_pauline@rediffmail.com

### REFERENCES

- Currie BJ, Fisher DA, Howard DM, Burrow JN. Neurological melioidosis. *Acta Trop* 2000;74:145-51.
- Muthusamy KA, Waran V, Puthucherry SD. Spectra of central nervous system melioidosis. *J Clin Neurosci* 2007;14:1213-5.
- Limmathurotsakul D, Chaowagul W, Wongsrikaew P, Narmwong A, Day NP, Peacock SJ. Variable presentation of neurological melioidosis in Northeast Thailand. *Am J Trop Med Hyg* 2007;77:118-20.
- Limmathurotsakul D, Peacock SJ. Melioidosis: A clinical overview. *Bri Med Bull* 2011;99:125-39.
- Bommakanti K, Ankathi P, Uma P, Malladi S, Laxmi V. Cerebral abscess and calvarial osteomyelitis due to *Burkholderia pseudomallei*. *Neurol India* 2010;58:801-2.
- Currie BJ, Ward L, Cheng AC. The epidemiology and clinical spectrum of melioidosis: 540 cases from the 20 years Darwin prospective study. *PLoS Negl Trop Dis* 2010;4:e900.
- Prasad GL, Kini P, Divya S. Central nervous system melioidosis in the pediatric age group: A review. *Childs Nerv Syst* 2017;33:1-6.
- Wongwadee M, Linasmita P. Central nervous system melioidosis: A systematic review of individual participant data of case reports and case series. *PLoS Negl Trop Dis* 2019;13:e0007320.
- Hsu CC, Singh D, Kwan G, Deuble M, Aquilina C, Korah I, *et al.* Neuro-melioidosis: Craniospinal MRI findings in *Burkholderia pseudomallei* infection. *J Neuroimaging* 2016;26:75-82.
- Maulik K, Shaikh GN, Kasinathan A, Chandrasekaran V, Parameswaran N, Biswal N. MRI findings in a child with neuro melioidosis. *Neurology* 2020;95:836-7.
- Hoffmaster AR, AuCoin D, Baccam P, Baggett HC, Baird R, Bhengisri S, *et al.* Melioidosis diagnostic workshop, 2013. *Emerg Infect Dis* 2015;21:e141045.
- Baby N, George M, Malini P, Divakar A, Bhanu DK, Saji M, *et al.* Melioidosis presenting as rhombencephalitis with antiganglioside antibody positivity. *J Neurol Exp Neurosci* 2021;7:9-14.
- Appassakij H, Silpajajakul KR, Wansit R, Pornpatkul M. Diagnostic value of the indirect hemagglutination test for melioidosis in an endemic area. *Am J Trop Med Hyg* 1990;42:248-53.

**Submitted:** 11-Apr-2021 **Revised:** 16-Jul-2021 **Accepted:** 06-Aug-2021

**Published:** 22-Oct-2021

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

**DOI:** 10.4103/aian.aian\_316\_21