RESIDENT CASE REPORT

Paenibacillus dendritiformis Hemorrhagic Meningoencephalitis in an Infant: Case Report and Literature Review

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ABSTRACT

There is sparse literature on *Paenibacillus* species causing human infections, particularly in the pediatric population with only few case reports of neonatal sepsis with variable outcomes. To our knowledge, however, there are no existing reports of *Paenibacillus dendritiformis* species causing infection in humans. We present a case of a 31-day-old infant who presented with fever and was found to have hemorrhagic meningoencephalitis secondary to *Paenibacillus dendritiformis*.

ABBREVIATIONS

CSF: cerebrospinal fluid
ED: emergency department
EEG: electroencephalogram
EVD: extraventricular drain
IV: intravenous
MIC: minimum inhibitory concentration
MRI: magnetic resonance imaging
RBC: red blood cell
rRNA: ribosomal ribonucleic acid
WBC: white blood cell

INTRODUCTION

The Paenibacilli species are a recently classified but important bacteria that affect humans, animals, and plants. The species is well-known within the field of agriculture, often thought to promote crop growth for plants.[1] They are a facultatively anaerobic endospore-forming, rod-shaped bacteria thought to be Gram-positive to Gram-variable and frequently identified based on 16S rRNA.[2] Recent literature suggests that Paenibacilli have become a rare human pathogen with potentially fatal outcomes, particularly in immunocompromised adults and premature infants.[2-4]

CASE REPORT

A 31-day-old male infant, born prematurely after 33 weeks gestation and 5 days as a dichorionic diamniotic twin, presented to a local emergency department (ED) with fever, irritability, and decreased oral intake. Perinatal maternal group B streptococcus status was unknown, but the mother received ampicillin adequately prior to delivery. All other maternal infectious screening tests were unremarkable. After an uneventful caesarean delivery, the neonate required management in the neonatal unit for 10 days due to respiratory distress after birth. He received empiric antibiotic coverage with intravenous (IV) ampicillin and gentamicin, which were discontinued after 48 hours as blood cultures remained without growth. He was discharged home in stable condition.

On presentation to the ED, the infant had a fever of 38.4°C but was otherwise well appearing and well perfused. Cerebrospinal fluid (CSF) studies showed a white blood cell (WBC) count of >2,000 cells/µL (normal for age 0-6 cells/µL) with 74% neutrophils and 15% lymphocytes, glucose of 1 mg/dL (normal for age 34-119 mg/dL), protein of 415 mg/dL (normal for age 58 ±17 mg/dL), and a red blood cell (RBC) count of 107 cells/µL. The CSF Gram stain showed no organisms. Urinalysis was normal. Blood, CSF, and urine cultures were obtained. He received meningitis dosing of IV ampicillin and ceftriaxone as well as gentamicin and was then transferred to our pediatric hospital for further management.

At our facility, vancomycin and acyclovir were initiated upon admission. Shortly thereafter the patient developed brief eye twitching which self-resolved, but was then noted to have arching of the back and clonus in the lower extremities. An electroencephalogram (EEG) demonstrated excessive discontinuity and multifocal interictal epileptiform abnormalities arising from within both hemispheres independently. Lorazepam was administered and antiepileptic therapy with levetiracetam and phenobarbital was initiated. Subsequent brain magnetic resonance imaging (MRI) with contrast revealed findings consistent with meningitis, encephalitis and developing abscesses involving the supratentorial region (Figure 1). Neurosurgical intervention was deemed unnecessary at the time. The patient received IV acyclovir until CSF *Herpes simplex* virus PCR test was negative and the CSF culture returned positive for *Paenibacillus dendritiformis*. A repeat lumbar puncture was performed to ensure the organism was not a contaminant. It showed CSF WBC count of 940 cells/µL, RBC 35 cells/µL, glucose <10 mg/dL, protein 824 mg/dL, and repeated growth of *P.* dendritiformis on culture.

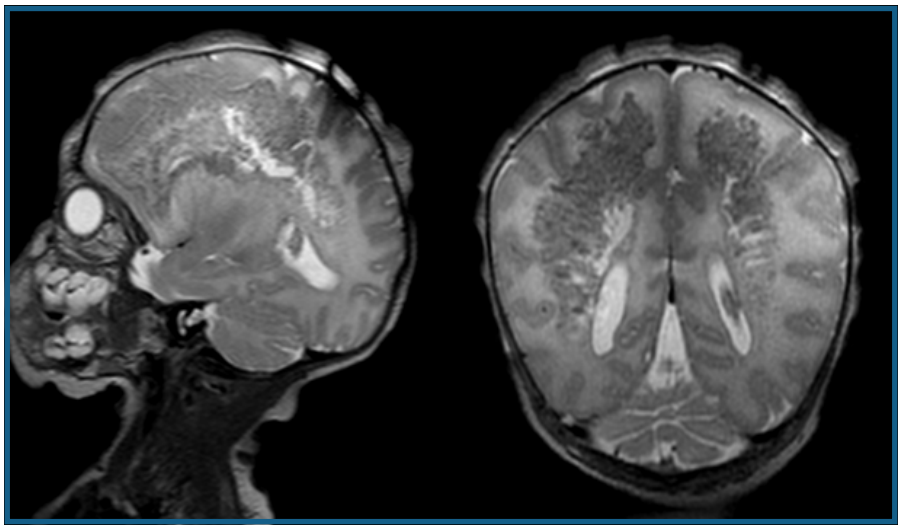

Figure 1
Brain MRI with contrast on admission: axial and sagittal views showing extensive bilateral abnormal signal in the supratentorial brain with restricted diffusion and hemorrhagic products. There is also extensive meningeal and parenchymal enhancement with large areas of peripheral enhancement involving the white matter bilaterally.

Ten days after admission, the infant had excessive crying, persisting fevers and a bulging anterior fontanel. A repeat brain MRI shown demonstrated an interval increase in ventricle size due to encephalomalacia, splaying of the cranial sutures, development of cystic spaces concerning for cerebral abscesses and findings consistent with hemorrhagic meningoencephalitis (Figure 2). Neurosurgery performed an urgent craniectomy for endoscopic fenestration of the ventricle and multiple infected cerebral cysts as well as placement of an external ventricular drain (EVD). The patient was continued on ceftazidime and vancomycin until the organism was shown to be susceptible to ampicillin (minimum inhibitory concentration [MIC]= 0.19 ug/mL). He was then switched to and maintained on high-dose IV ampicillin monotherapy for 6 weeks.

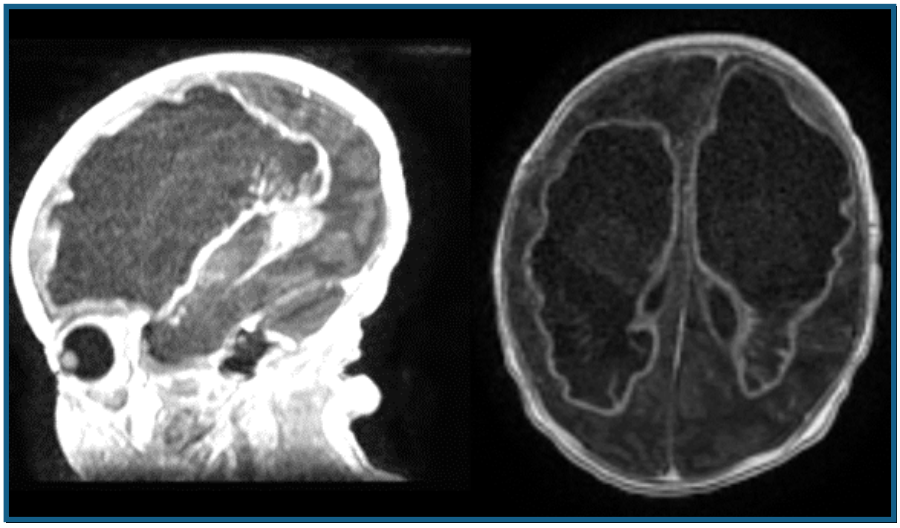

Figure 2
Brain MRI with contrast 10 days after admission: axial and sagittal views showing interval development of necrosis of the white matter bilaterally with peripheral enhancement and a central area showing restricted diffusion. There is enhancement of the meninges and ventricular lining bilaterally.

DISCUSSION

Paenibacillus dendritiformis and related species are rarely reported in the literature as human pathogens. *Paenibacillus polymyxa* has been shown to colonize the intestinal tract of termites, which are a relatively common household pests.[5] While a history of termite infestation in our patient's household was reported after further questioning of the family, we can only propose that the *Paenibacillus* species causing his infection may have originated from the intestinal flora of these pests.

Among the reported cases in the literature, *Paenibacillus* species have been isolated from human wound infections, gingiva, blood, urine, and CSF.[3,6-9] However, in several of these cases the organism was thought to be a contaminant.[3,9] Importantly, an apparent outbreak of *Paenibacillus macerans* was reported in a neonatal intensive care unit in 1999, which was eventually determined to be contaminated blood culture bottles.[10] One case report identified recurrent soft tissue *Paenibacillus* infection in a healthy adult, eventually requiring indefinite therapy with trimethroprim-sulfamethoxazole.[11] There is little consensus on susceptibility interpretation and preferred antibiotic therapy because of limited and anecdotal data on this organism.. Saez-Nieto et al. examined 138 *Paenibacillus* species isolated from human and environmental sources and found that 25% of the isolates represented true infections.[12] Antimicrobial susceptibility testing showed 95.6% of isolates were resistant to ampicillin, 44% were resistant to cotrimoxazole, 20-30% were resistant to cefotaxime and vancomycin and 13% were resistant to rifampicin and erythromycin.[12] Our patient had *Paenibacillus dendritiformis* that was susceptible to ampicillin (MIC= 0.19 ug/mL), gentamicin (MIC= 2 ug/mL), penicillin (MIC < 0.03 ug/mL), rifampin (MIC <= 1 ug/mL) and vancomycin (MIC= 4 ug/mL).

Three reports exist of neonatal sepsis attributed to a *Paenibacillus*. Two of the neonates described had poor neurologic outcome despite adequate antibiotic therapy, and one had a favorable outcome with similar therapy.[2,4,13] All three cases involved neonatal fever and seizures, commonly expected in neonates with meningoencephalitis. Our patient's clinical course was complicated by the need for neurosurgical drainage and EVD placement in addition to antiepileptic therapy. He was discharged home in stable condition following completion of his antibiotic therapy course. Currently, at the age of 21 months, he has not had any recurrent infection or the need for additional neurosurgical intervention. He has significantly delayed language and gross motor development for which he receives physical therapy.

To our knowledge, *Paenibacillus dendritiformis,* as was isolated from our patient's spinal fluid, has not been previously reported in the literature. Pediatric providers should be aware of the potential of serious neonatal infection with this organism, its possible relationship to termites in the patient's environment, and spectrum of outcomes.

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