Case Report

Plesiomonas shigelloides Sepsis and Meningoencephalitis in a Surviving Neonate

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In this study, we report the case of a 2.5-day-old neonate with septicemia and meningitis due to Plesiomonas shigelloides. Culture of the cerebrospinal fluid showed Gram-negative rods, although the glucose, protein and leukocyte counts were normal. The patient was treated with meropenem and survived without any sequelae, although we were not able to identify the source of the infection. In addition, ten previously reported cases of this infection are reviewed.

KEYWORDS: meningoencephalitis, newborn, Plesiomonas shigelloides, sepsis

Introduction

Plesiomonas shigelloides was first isolated in 1947 by Ferguson and Henderson and is the only species in the genus. Humans are infected with P. shigelloides through ingestion of contaminated food or water, or by contact with colonized animals. P. shigelloides is an uncommon cause of acute bacterial gastroenteritis and an extremely rare cause of extra-intestinal infections, e.g. sepsis, meningitis, cellulitis, septic arthritis, and osteomyelitis. Underlying illnesses associated with extra-intestinal infections are immunodeficiency, sickle cell disease, and cirrhosis. To date, 10 cases have been reported in which neonates developed P. shigelloides sepsis and meningitis and 70% of these cases resulted in the death of the patient. We report a case of a neonate with P. shigelloides sepsis and meningoencephalitis; the second case in Europe, but the first to survive.

Case Report

A healthy 34-year-old primipara woman delivered a 2,800-g female infant at term after an uncomplicated pregnancy. The labor was spontaneously vaginal and uneventful. The Apgar scores were 8 in the 1st minute and 10 in the 5th minute. The baby was doing well for the first 24 hours. However, on the second day of life (DOL 2), jaundice, fever, irritability and poor feeding were noted. Lethargy, jaundice, and abdominal tension were present and the Moro reflex was absent. Upon initial evaluation, laboratory findings revealed leucopenia, with a leukocyte count of $1 \times 10^9/L$ (40% lymphocytes, 30% segmented granulocytes, 20% band forms, and 10% metamyelocytes) and 10 red cell
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precursors per 100 leukocytes were found in the differential blood count. The patient was also thrombocytopenic (platelet count=74×10^9/L) and C-reactive protein was 73 mg/L. Other laboratory tests yielded the following: hemoglobin=12 g/dL; blood glucose=80 mg/dL; serum total bilirubin=19.7 mg/dL; serum direct bilirubin=1.1 mg/dL; serum calcium=6.5 mg/dL; prothrombin time=29 seconds; partial thromboplastin time=81 seconds; fibrinogen=330 mg/dL; and D-dimer=2,113 μg/dL. Serum electrolyte values were normal. The cerebrospinal fluid (CSF) contained eight leucocytes, six polymorphonuclear leukocytes, and two lymphocytes per mm³. The glucose concentration in the CSF was 67 mg/dL and protein was 96 mg/dL. Blood and the CSF were obtained for culture. On admission, the cranial ultrasonogram was normal. A right suprarenal hematoma (27×22 mm) was noted on the abdominal ultrasonogram, which resolved spontaneously. According to the laboratory findings, the reasons for the hematoma may have been thrombocytopenia and/or a coagulation disorder. A provisional diagnosis of sepsis was made and treatment with intravenous cefotaxime, amikacin, calcium gluconate and positive inotropic drugs was instituted.

Shortly after the first examination, the body temperature of the baby rose to 41°C (axillary). Recurrent convulsions occurred and were treated with intravenous phenobarbital. Antibiotic therapy was changed to meropenem when Gram-negative rods were seen on the stained CSF smear. In spite of phototherapy, an exchange transfusion was performed as the level of indirect bilirubin was 25 mg/dL after 6 hours. On DOL 3, a cardiorespiratory arrest ensued and the infant responded to resuscitation efforts. As the patient was not breathing spontaneously, she was put on positive pressure ventilation therapy for 2 days. On DOL 4, cultures of both blood and the CSF produced a motile, Gram-negative rod, which was subsequently identified as P. shigelloides by the API 20E test. The antibiotic susceptibility testing showed the bacteria to be resistant to ampicillin, but susceptible to meropenem. On DOL 5, the patient’s spontaneous activity was improved. After DOL 6, C-reactive protein levels gradually started to decline, while leukocyte and platelet counts gradually increased.

We had some thoughts on the origin of the infection. However, neither the mother nor the father had consumed any kind of seafood prior to their child’s birth. There was no family history of foreign travel, diarrhea or environmental water exposure. Moreover, there were no fish, frogs, snakes or lizards at their home. However, we suspected the relatives of the family (who had arrived from Germany 23 days before the birth) may have been the source of infection. P. shigelloides was not found in cultures from the labor room or the maternal urine and stool.

On DOL 16, the control cranial and abdominal ultrasonograms were normal. All laboratory findings, including leukocyte counts, glucose and protein levels and CSF culture were normal. The patient was treated with meropenem for 21 days. The patient’s neurological examinations at 3, 12, and 18 months of age were normal. She remains on a follow-up program.

Discussion

P. shigelloides is a very rare causative organism of neonatal meningitis and sepsis. Its phenotypic characteristics are based on polar flagella, oxidase production, and fermentative properties. The Gram-negative rod is classified into the Vibrionaceae family. Its primary natural reservoirs are water and soil surfaces, as well as fish and other marine animals, especially oysters. The organism is recovered from freshwater and estuaries in temperate and tropical regions and occasionally from seawater during the summer months. To date, the infection has been described in 10 babies, seven of whom died. Our description of a neonate suffering from P. shigelloides sepsis and meningoencephalitis is the second case, but the first survivor, in Europe. The relevant features of the 10 reported cases and our own case of neonatal sepsis with meningitis caused by P. shigelloides are summarized in the Table.

P. shigelloides sepsis and meningoencephalitis in neonates is a progressive and often fatal disease. P. shigelloides is mostly resistant to penicillin and ampicillin. It is usually sensitive to third-generation cephalosporin. We diagnosed a more serious case of neonatal P. shigelloides sepsis and meningoencephalitis due to leucopenia, thrombocytopenia and suprarenal hematoma. According to the experiments in our hospital, fatal Gram-negative rods, especially Klebsiella pneumoniae and Pseudomonas aeruginosa, are resistant to cefotaxime therapy. Because of the severe neonatal septic course and the pending cultures, cefotaxime therapy was changed to meropenem. As the antibiotic treatment of meningitis and sepsis caused by Gram-negative microorganisms usually lasts for 21 days, meropenem was stopped at the end of the 3rd week.
It was interesting that the CSF culture produced Gram-negative *P. shigelloides*, because CSF glucose levels, protein levels and leukocyte counts were all normal in our patient. Conversely, biochemical and cellular responses were detected in the CSF in the other reported cases. This extraordinary situation might be explained by the time of meningitis diagnosis, which was made early, and this early diagnosis may have played a part in the success of the therapy.

However, there is still a discussion point regarding neonatal *P. shigelloides* infection. It is well known that early-onset neonatal sepsis is rarely seen with meningitis or meningoencephalitis. All the reported cases of neonatal *P. shigelloides* sepsis have been seen in the first four DOL. However, in these cases, sepsis was diagnosed along with meningitis or meningoencephalitis. We consider this point remarkable and open to further discussion.

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### References


