Contents lists available at ScienceDirect

Journal of Microbiology, Immunology and Infection

Journal homepage: http://www.e-jmii.com

Case Report

Plesiomonas shigelloides Sepsis and Meningoencephalitis in a Surviving Neonate

Osman Ozdemir[a]*, Sinan Sari[b], Serdar Terzioglu[c], Aysegul Zencirogl[d]

[a]Department of Pediatrics, Kecioren Training and Research Hospital, Ankara, Turkey.
[b]Department of Pediatrics, School of Medicine, Gazi University, Ankara, Turkey.
[c]Department of Microbiology, Doctor Sami Ulus Children's Hospital, Ankara, Turkey.
[d]Department of Neonatology, Doctor Sami Ulus Children's Hospital, Ankara, Turkey.

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, cellulites, septic arthritis, and osteomyelitis. Underlying illnesses associated with extra-intestinal infections are immunodeficiency, sickle cell disease, and cirrhosis.[1] 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.[2–11] We report a case of a neonate with *P. shigelloides* sepsis and meningoencephalitis; the second case in Europe, but the first to survive.[9]

*Corresponding author. Kecioren Egitim ve Arastirma Hastanesi, Sanatoryum Caddesi, Pinarbasi Mahallesi, Ardahan Sokak, 1 Kecioren, Ankara, Turkey.

E-mail: pedkard@gmail.com

Article History:
Received: Apr 20, 2009
Revised: Jun 5, 2009
Accepted: Jul 28, 2009

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 leucopoenia, with a leukocyte count of 1×10^9/L (40% lymphocytes, 30% segmented granulocytes, 20% band forms, and 10% metamyelocytes) and 10 red cell

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 mm^3. 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.[1] To date, the infection has been described in 10 babies, seven of whom died.[2–11] Our description of a neonate suffering from *P. shigelloides* sepsis and meningoencephalitis is the second case, but the first survivor, in Europe.[9] 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.[1–12] *P. shigelloides* is mostly resistant to penicillin and ampicillin. It is usually sensitive to third-generation cephalosporin.[1] We diagnosed a more serious case of neonatal *P. shigelloides* sepsis and meningoencephalitis due to leucopoenia, 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 micro-organisms usually lasts for 21 days, meropenem was stopped at the end of the 3rd week.

Table. Cases of neonatal *Plesiomonas shigelloides* sepsis and meningitis

Case	Sex	Weight (kg)	Age (d)	Treatment	Course	Year of publication [Reference]
1	F	2.2	4	Rifampin + Ampicillin	Died	1978 [2]
2	F	3.7	2	Penicillin + Gentamicin	Died	1980 [3]
3	M	3.5	3.5	Penicillin + Gentamicin	Survived	1981 [4]
4	M	2.8	4	Ampicillin + Gentamicin	Died	1982 [5]
5	M	3.3	2.5	Ampicillin + Kanamycin	Died	1983 [6]
6	M	3.8	2	Cefotaxime	Survived	1988 [7]
7	M	2.4	2	Netilmicin + Ampicillin + Cefotaxime	Died	1989 [8]
8	M	–	4	Netilmicin/Gentamicin + Mezlocillin + Cefotaxime	Died	1992 [9]
9	–	–	3	Cefotaxime	Survived	1994 [10]
10	–	–	–	–	Died	1999 [11]
11	F	2.6	2.5	Meropenem	Survived	Present study

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.[2–11] 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.[12] 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.[2–11] We consider this point remarkable and open to further discussion.

Acknowledgments

The authors would like to thank Dr Cigdem Uner (MD, Radiologist, Doctor Sami Ulus Children's Hospital, Ankara, Turkey) for their exhaustive revision of this manuscript.

References

1. Brenden RA, Miller MA, Janda JM. Clinical disease spectrum and pathogenic factors associated with *Plesiomonas shigelloides* infections in humans. *Rev Infect Dis* 1988;10:303–16.
2. Appelbaum PC, Bowen AJ, Adhikari M, Robins-Browne RM, Koornhof HJ. Neonatal septicemia and meningitis due to *Aeromonas shigelloides*. *J Pediatr* 1978;92:676–7.
3. Dahm LJ, Weinberg AG. *Plesiomonas (Aeromonas) shigelloides* septicemia and meningitis in a neonate. *South Med J* 1980;73: 393–4.
4. Su S, Ee CK. *Plesiomonas shigelloides* meningitis in newborn. *J Singapore Paediatr Soc* 1981;23:156–8.
5. Dudley Ag, Mays W, Sale L. *Plesiomonas (Aeromonas) shigelloides* meningitis in a neonate: a case report. *J Med Assoc Ga* 1982;71: 775–6.
6. Pathak A, Custer JR, Levy J. Neonatal septicemia and meningitis due to *Plesiomonas shigelloides*. *Pediatrics* 1983;71:389–91.
7. Waecker NJ, Davis CE, Bernstein G, Spector SA. *Plesiomonas shigelloides* septicemia and meningitis in a newborn. *Pediatr Infect Dis J* 1988;7:877–9.
8. Billiet J, Kuypers S, Van Lierde S, Verhaegen J. *Plesiomonas shigelloides* meningitis and septicemia in a neonate: report of a case and review of the literature. *J Infect* 1989;19:267–71.
9. Terpeluk C, Goldmann A, Bartmann P, Pohlandt F. *Plesiomonas shigelloides* sepsis and meningoencephalitis in a neonate. *Eur J Pediatr* 1992;151:499–501.
10. Fujita K, Shirai M, Ishioka T, Kakuya F. Neonatal *Plesiomonas shigelloides* septicemia and meningitis: a case and review. *Acta Paediatr Jpn* 1994;36:450–2.
11. Bravo L, Cabrera R, Ramírez M, Llop A, Fernández A, Ariosa C, Ferrer R. Fatal *Plesiomonas shigelloides* in a newborn. *Mem Inst Oswaldo Cruz* 1999;94:661–2.
12. Stoll BJ. Infections of the neonatal infant. In: Kliegman RM, Behrman RE, Jenson HB, Stanton BF, eds. *Nelson Textbook of Pediatrics.* Philadelphia: Saunders Elsevier, 2007:794–811.