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Recurrent Meningitis in an Infant with Unusual Bacteria and A Spinal Dermal Sinus Fistula

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1. Abstract

We report a 3-month-old male infant who had two meningitis events with Citrobacter koseri and was eventually diagnosed as having a spinal dermal sinus tract. Recurrent meningitis is rare but fatal in newborns, and congenital spinal dermal sinus tracts are considered a crucial factor. Thus, it is essential to carefully reexamine the paravertebral region for patients with recurrent meningitis or meningitis with unusual bacteria in cerebral spinal fluid. Also, an MRI is helpful when spinal dermal sinus tracts are suspected.

2. Introduction

A spinal dermal sinus tract is often subtle and asymptomatic at birth. However, if untreated, it may cause problems with leg weakness, back pain, and problems related to the urinary system. Recurrent meningitis in children is unusual and life-threatening [1]. We report a 3-month-old male infant who had two meningitis events and was eventually diagnosed as having a spinal dermal sinus tract. He was soon given surgical and medical treatment. He had no further meningitis episodes during follow-up.

3. Case Report

A three-month-old male infant presented in our emergency room with a six-day fever accompanied by intermittent vomiting. It was his third postnatal fever. His first was a neonatal fever of unknown etiology when he was six days old and in a different hospital; that fever remitted after three days of antibiotic treatment. His second admission was because of a three-day fever; a complete septic workup, including lumbar tapping and a blood culture, isolated Citrobacter koseri in his cerebrospinal fluid. After three weeks of antibiotic treatment, his condition improved and he was discharged. This time, he was brought to the emergency room and treated for gastroenteritis. After three days, the fever persisted. He appeared ill and lethargic. He had a soft neck but increased deep tendon reflexes over his upper and lower limbs. After it was discovered that he had a small sacral dimple without a discharge (Figure 1A, arrow), he was transferred to the Pediatric Neurology division. A full septic workup was done: a complete blood count, a blood culture, and a lumbar culture. The blood count showed the patient's white blood cell count at 8000/μL and C-reactive protein at 3.38 mg/dL. His spinal fluid showed white blood cells at 1590/μL and a low glucose level of 13 mg/dL. He was treated empirically on cefotaxime (200 mg/kg/day) and penicillin (40 mg/kg/day) for suspected bacterial meningitis. The Gram stain revealed a Gram-negative, non-spore-forming bacillus, and showed Citrobacter koseri growing on his anaerobic culture, which was consistent with his previous meningitis organism. The patient's original

antibiotic regimen was replaced with piperacillin-tazobactam (80 mg/kg/q6h). The blood culture was negative after three days of incubation. Spinal Magnetic Resonance Imaging (MRI) revealed a sinus tract extending from the subcutaneous tissue to the spinal canal (S2/S3 level). A small S1/S2-level lesion with fat content made us suspicious of a dermoid cyst with an associated spinal fistula (Figure 1B, C). On the patient's seventh post-admission day, he underwent spinal neurosurgery to totally dissect the dermal sinus. Postoperatively, we continued his three-week meningitis treatment, and he fully recovered. After 25 days of hospitalization, he was discharged without any discomfort. He had no recurrent fever after he turned six months old.

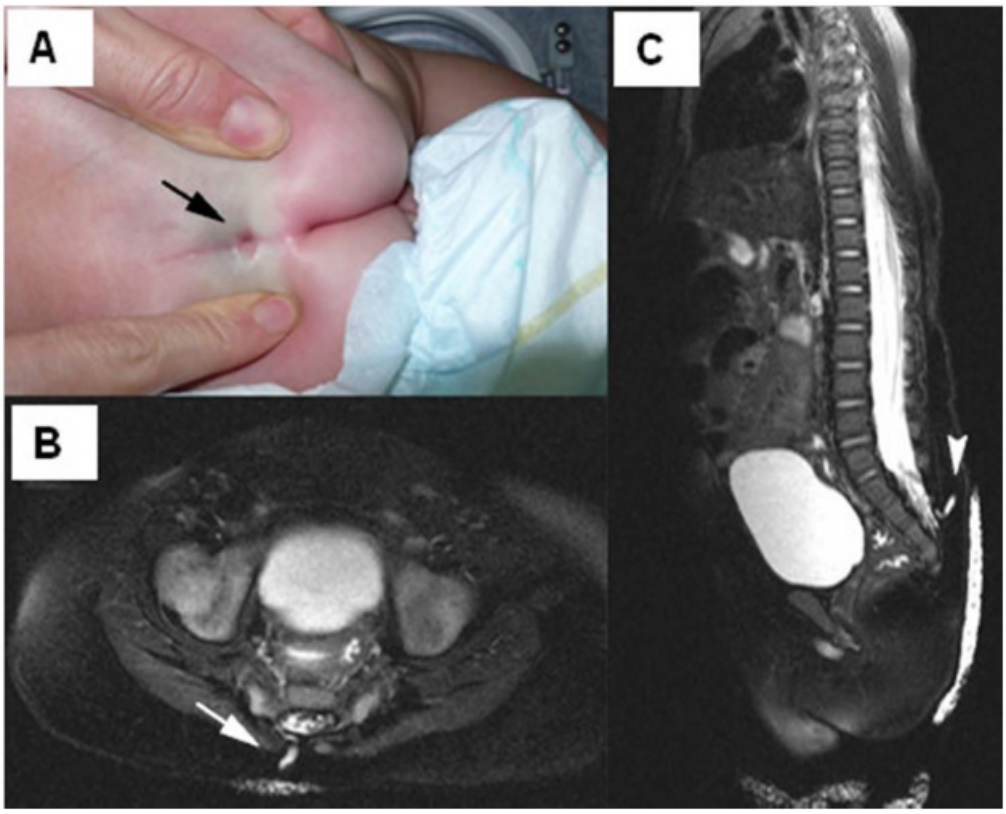

Figure 1A: A small dimple is on the lumbosacral area (*black arrow*);
Figure 1B, 1C: Spinal magnetic resonance imaging scan reveals a tubular structure extending from skin surface into spinal canal (**B:** axial view*, white arrow*; **C:** sagittal view, *white arrowhead*).

4. Discussion

A spinal dermal sinus tract, one type of spina bifida, is often undetected if it is asymptomatic. However, its existence is not only a potential risk for spinal cord dysfunction, but also a high risk for a central nervous system infection [2]. Hence, recurrent meningitis should raise the possibility of a parameningeal focus of infection [3]. In addition to immunodeficiency disorders, several anatomic defects have been reported [3] to be associated with recurrent meningitis, such as dorsal spinal dermal sinus tracts, defects in the cribriform plate, in the sphenoid or other sinuses, or in the temporal bone. In a pediatric population, recurrent bacterial meningitis implies that the patient might have a structural defect of the cribriform plate or a Mondini defect of the inner ear [4]. However, those particular infection routes often bring out particular organisms, which are quite different from those in community acquired or nosocomial meningitis [5].

Recurrent meningitis is rare the predisposing factors are significant. Congenital dermal sinuses occurring in the midline are among the etiological factors. The most common organisms cultured from dermal sinus tracts include *staphylococci, E. coli*, and *Proteus*. In the lesions situated in lumbosacral Gram-negative organisms are often the offending agent [6]. In our case, *Citrobacter koseri*, a Gram-negative, non-spore-forming bacillus, was cultured twice from cerebrospinal fluid-preemptive and prolonged antibiotic treatment made the fistula sterile for cultures. However, the negative findings of an abdominal computed tomography scan, a brain MRI, and a complete set of immune function tests suggested that the lumbosacral dermal sinus tract was crucially involved in this patient's recurrent meningitis.

5. Conclusion

Recurrent meningitis is rare but fatal in newborns, and congenital spinal dermal sinus tracts are considered a crucial factor. Thus,

it is essential to carefully reexamine the paravertebral region for patients with recurrent meningitis or meningitis with unusual bacteria in cerebral spinal fluid. Also, an MRI is helpful when spinal dermal sinus tracts are suspected.

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