Case Report

Citrobacter sedlakii – A rare cause of meningitis and brain abscess in a neonate

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ABSTRACT

Citrobacter sedlakii is a Gram-negative bacillus, usually a non-pathogenic organism, found in human stool. However, it may rarely cause devastating illness in human beings. We are describing a case of *C. sedlakii* meningitis and brain abscess in a neonate. To the best of our knowledge, this is the second case, of *C. sedlakii* causing complicated meningitis in a neonate in the world.

Key words: Brain abscess, *Citrobacter sedlakii*, Neonate

Citrobacter sedlakii is a Gram-negative bacillus, which is frequently found in the stool and is non-pathogenic in most of the cases [1]. It was isolated first from the human stool and wound as strains of *Citrobacter freundii* [2]. In 1993, Czech microbiologist, Jiri Sedlak isolated six strains of *C. freundii* as *C. sedlakii* based on DNA hybridization. It can be differentiated from other *Citrobacter* species by its ability to produce indole, arginine dihydrolase activity, and ornithine decarboxylase activity [2]. It expresses the O157 antigen which is commonly found on pathogenic *E. coli*; however, this does not cause disease. In the literature in pediatric age group, only a single case is described earlier in a newborn causing brain abscess.

CASE REPORT

A male neonate presented to pediatrics emergency with complaints of refusal to feed and convulsions at day 12 of life. He was delivered by lower segment cesarean section (LSCS) at full term with a birth weight of 3500 g and was discharged on day 4 of life and was thriving well till day 12 of life. The indication for emergency LSCS was cord around neck and the baby cried soon after birth. On examination, anterior fontanel was at level, tone was increased, sucking was poor, and cry was inconsolable. A provisional diagnosis of late-onset sepsis with meningitis was made.

At presentation, blood sugar was 90 mg/dl and convulsions were present, so injection phenobarbitone (20 mg/kg) was given. As per the unit protocol, intravenous piperacillin-tazobactam (100 mg/kg/dose 8 hourly) and amikacin (15 mg/kg/dose 24 hourly) were started. The cerebrospinal fluid (CSF) examination was suggestive of pyogenic meningitis (Table 1). Antibiotics were upgraded to meropenem (40 mg/kg/dose 8 hourly) and amikacin. Meanwhile, blood culture sensitivity showed the growth of *C. sedlakii* sensitive to amikacin, meropenem, imipenem, ciprofloxacin, amoxicillin/clavulanic acid, piperacillin/tazobactam, levofloxacin, tetracycline, and cefoxitin. The isolate was resistant to ampicillin, cefazolin, cephalexin, ceftriaxone, septran, aztreonam, tobramycin, cefepime, and gentamicin. CSF culture also showed growth of *C. sedlakii*. The first cranial ultrasound on day 2 of admission was suggestive of chinked right ventricle.

The baby improved, so shifted to postnatal ward with mother. He started having high-grade fever after being normal for 4 days. Sepsis screen, blood culture, CSF culture sensitivity, urine routine microscopy and culture sensitivity, and USG cranium were repeated. Repeat USG cranium was suggestive of a well-defined hypoechoic lesion measuring 20 ml noted on right cerebral cortex with internal cystic component with septations. On repeat USG after 72 h, multiple hypoechoic rounded lesions with internal echoes were noted in both the cerebral hemispheres, largest measuring 4.3×4.3 cm on the right side with a few of them showing anechoic component measuring 1.7×1.7 cm in bilateral parietal lobes with midline shift to the left.

Magnetic resonance imaging (MRI) brain was suggestive of few well defined intra-axial space occupying lesions involving right frontal lobe with fluid levels and mass effect, suggestive of cerebral abscesses (Fig. 1). Neurosurgeons did a burr hole with USG-guided aspiration of the right frontal abscess. The aspirated pus was sent for Gram stain, culture sensitivity including mycobacterial cartridge-based nucleic acid

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Table 1: Comparison between the present case and previous case (Dyer *et al.*, 1997)

	Our case	Dyer *et al.* (1997)
Gestational age	38 weeks	30 weeks
Birth weight	3500 g	1740 g
Maternal age	28 years	15 years
Mode of delivery	LSCS for cord around neck	NVD
History of consanguinity	Present	Not known
Apgar score at 1 and 5 min	7,8	8,9
Presented as	Late-onset sepsis, PM	RDS on day 1 with EONS
Blood culture		
1st	*Citrobacter sedlakii*	*Enterobacter cloacae* and *Citrobacter sedlakii*
2nd	Sterile	*Citrobacter sedlakii*
CSF analysis		
TLC	50/cc	113/cc
Erythrocytes	None	8750/ml
Protein	221.4 g%	105 g%
Sugar	56.4 mg%	63 mg%
Culture	*Citrobacter sedlakii*	*Citrobacter sedlakii*
Antibiotics	IV meropenem amikacin IV levofloxacin amikacin metrogyl IV meropenem vancomycin 6 weeks	IV cefotaxime; IV gentamicin – 2 weeks 1V cefotaxime; oral cotrimoxazole 2 weeks IM ceftriaxone; oral CTX 3 weeks Discharged on oral CTX 2 weeks
Surgical procedure	Burr hole with ultrasound-guided aspiration – twice	None
MRI	Few well-defined intra-axial space-occupying lesion involving right frontal lobe showing fluid levels and mass effect – cerebral abscess	Abscesses in the right deep frontal, left frontal, and left occipitoparietal areas
EEG	Normal	Bilateral temporal positive and negative sharp waves
ABER	Normal	Normal
Outcome	Normal till 5 ½ months	Normal at 5 months

PM: Pyogenic meningitis, EONS: Early-onset neonatal sepsis, CSF: Cerebrospinal fluid, TLC: Total leukocyte count, CTX: Cotrimoxazole, EEG: Electroencephalogram, ABER: Auditory brainstem evoked response, MRI: Magnetic resonance imaging

amplification test, fungal culture, all were normal. Post-aspiration and contrast-enhanced computed tomography (CECT) head were done to follow the size of abscess which was reduced.

After 15 days of procedure, the baby again developed high-grade fever. Hence, CECT head was repeated and revealed that abscess cavity was reexpanding. Gadolinium-enhanced MRI brain with MR spectroscopy was done and antibiotics were planned to continue for a minimum of 6 weeks. After 2 weeks of the 1st aspiration, again USG-guided aspiration was done. In view of recurrent fever and an unusual organism, neonate was investigated for primary immunodeficiency which was be normal.

Gradually, the patient became afebrile and cavity size remained constant, so the baby was discharged at the age of 12 weeks. At discharge, the neurological examination of the neonate was abnormal with hypotonia of neck muscles on pull to sit maneuver but appendicular tone was normal. Social smile was present at discharge, and he was able to fix the gaze and track. At the time of follow-up at 6 months of age, he gained good neck control, was able to sit with support, and could transfer objects from one hand to another.

DISCUSSION

In 1993, six strains of *C. freundii* were identified as a separate organism and were named as *C. sedlakii* [2]. Brenner *et al.* classified citrobacteria by DNA hybridization. According to this, *C. freundii* complex consists of 11 germospecies, of which 8th was named as *C. sedlakii*.

On searching the literature on neonatal *C. sedlakii* infection, we found only one case report by Dyer *et al*. in a preterm baby (30 weeks gestation) with a birth weight of 1400 g. This neonate presented as early-onset neonatal sepsis with respiratory distress. Our case presented as late-onset neonatal sepsis with meningitis. On MRI, abscesses were there in the right deep frontal, left frontal, and left occipitoparietal areas. The salient features of our case versus the previous one are described in Table 1.

Although *Citrobacter* is an uncommon organism causing sepsis in neonates, it is associated with high risk of CNS involvement in the form of meningitis and abscesses. Among all the species, *Citrobacter koseri* is the most common. The reported cases in neonates are both acquired vertically and postnatally. Doran in his review article states the role of prolonged antibiotic therapy and surgical treatment in most of the neonates [3]. They related abscesses formation to the presence of a specific 32 kDa outer membrane protein.

Agarwal *et al*. had described a case of vertically acquired neonatal meningitis with multiple brain abscess by *C. koseri* who responded to 4-week IV imipenem with a burr hole aspiration [4]. Our case too complicated with brain abscesses treated by aggressive surgical approach with 6 weeks of

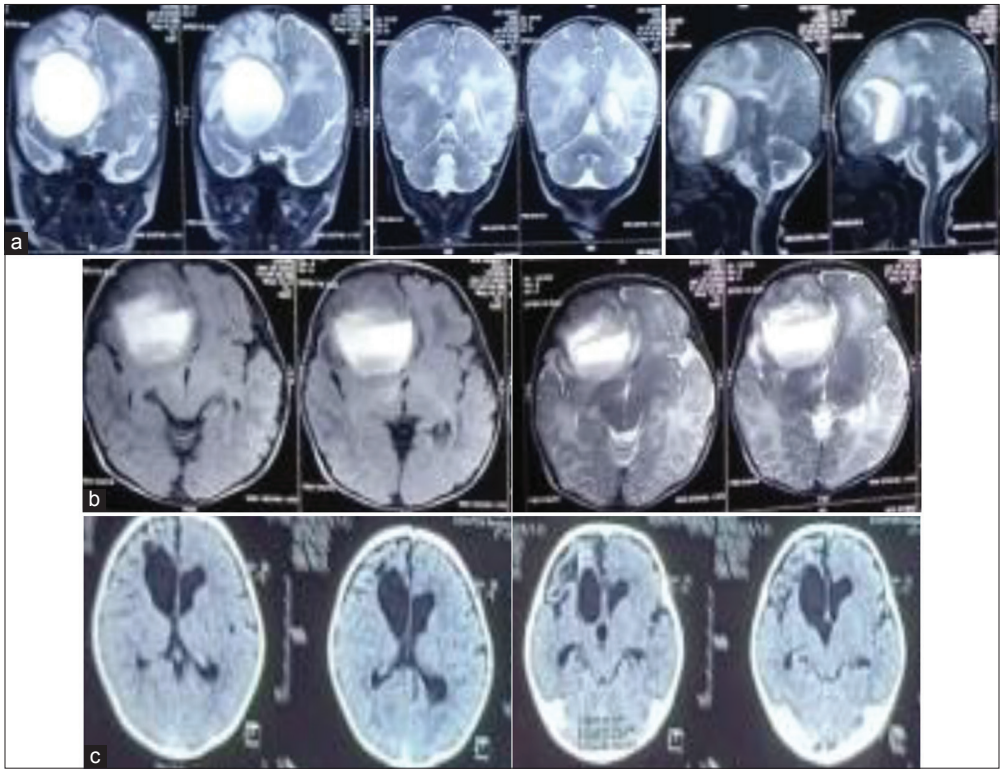

Figure 1: Neuroimaging before (a and b) and after (c) treatment

intravenous antibiotics. *Citrobacter diversus* meningitis has also been reported previously as fatal sporadic meningitis in a newborn with multiple brain abscess and spinal abscess. The portal of entry was omphalitis [5]. Another case of *C. diversus* meningitis was reported in a term male neonate with hydrocephalus which did not respond to intraventricular meropenem, improved on IV trimethoprim and sulfamethoxazole [6].

Chowdhry *et al*. described two cases of *C. koseri* brain abscesses in a 6-week, 2-month-old infant. Both had favorable outcomes with aggressive surgical and medical management [7]. Plakkal *et al*. [8] described a case of *C. freundii* meningitis with brain abscess in a 27-week preterm newborn, which developed brain abscess following meningitis in the parieto-occipital lobe. In our case, the neonate was full term, had involvement of frontal lobe. They treated with IV meropenem, ciprofloxacin for 5 weeks and surgical drainage with serial neuroimaging. The infant had normal neurological outcome at follow-up. Baumeister FAM monitored neonatal *C. koseri* meningitis and venticulitis with IL-6 in CSF and used high-dose cefotaxime (300 mg/kg/day) for the treatment without surgery [9].

CONCLUSION

Our case is the second case reported in world of neonatal brain abscess by *C. sedlakii.* He behaved like the previously reported case. The newborn presented with extensive brain involvement, required prolonged antibiotic therapy, sequential imaging, and burr hole aspirations. As with the former case, the final outcome was reassuring.

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