

Case Report

Cytomegalovirus-associated colitis with colonic stricture mimicking necrotizing enterocolitis in a newborn: a case report

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ABSTRACT

Necrotizing enterocolitis is the most common gastrointestinal emergency in neonatal period. The etiology is considered multifactorial. Risk factors include prematurity, enteral feeding, hypoxia, and bacterial colonization. The etiologic role of viruses is unclear. Although cytomegalovirus (CMV) is a common congenital infection in neonates, most patients are asymptomatic. Gastrointestinal manifestation is unusual. In this report, we described a newborn with perinatal CMV infection presenting with symptoms mimicking necrotizing enterocolitis.

Keywords: Cytomegalovirus, Necrotizing enterocolitis, Newborn

INTRODUCTION

Necrotizing enterocolitis (NEC) is the most common gastrointestinal emergency in the neonatal period and a mortality rate as high as 50%. The most consistent risk factors are prematurity, enteral feeding, hypoxia, and bacterial colonization.¹ It has become evident that cytomegalovirus (CMV) is the most important cause of congenital infection in the developed world. Most exposed term infants are asymptomatic or not infected, however about 10% of congenitally infected newborns present with rash, jaundice, and microcephaly at birth. In contrast, preterm infants who lack antibody to CMV can develop serious infection. In neonates, viral culture of urine, saliva or tissue sample is the primary diagnostic tool.^{3,4}

In this report, we described a newborn with perinatal CMV infection presenting with symptoms mimicking NEC.

CASE REPORT

A 19-year-old gravida 1 para 1 woman delivered vaginally a female baby at 27 weeks of pregnancy. Birth weight was 964 g. The mother's prenatal serologies were negative for hepatitis B, VDRL, and HIV. At birth, he had a 5-minute APGAR score of <7 with respiratory distress. She was admitted to our intensive care unit with prematurity and respiratory distress syndrome. A single dose of surfactant (100 mg/kg body weight Curosurf®, Chiesi Farmaceutici, Parma, Italy) was tested via intubation cannula. She was extubated at her first day of life. She passed the meconium within the first 24 h.

Minimal enteral feeds with breast milk were started via nasogastric tube.

On day of life (DOL) 18, the baby developed abdominal distention and small amount of gastric residues. A plain abdominal film showed dilated bowel loops, but no evidence of pneumatosis. She was managed clinically as NEC, enteral feedings were discontinued, and parenteral antibiotic therapy cefotaxime and metronidazole was started. During the next 2 weeks, the baby was hemodynamically stable and then enteral feedings were restarted with breast milk. However, she suffered from intermittent abdominal distension. Rectal irrigations twice daily were started, and the abdominal distention gradually resolved. On DOL 38, enteral feeding was stopped again. Three days later, the baby developed acute abdominal distention with respiratory failure that requiring intubation and mechanical ventilation. Abdominal X-ray showed mistiness over left side of bowels. Laboratory studies showed a white blood cell count of $12.1 \times 10^9/L$, with 42% lymphocytes, 36% neutrophils and 15% band forms, hemoglobin of 13.6 g/dL, and platelet count of $47 \times 10^9/L$. C reactive protein (CRP) level was elevated to 51.3mg/L. She was managed as recurrent NEC and treated again with bowel resting and antibiotics (vancomycin with meropenem). However, there was worsening of condition. An emergent

exploratory laparotomy showed that the small bowel was dilated to the cecum with a stricture at the distal ileum. Matted bowel loops in the left upper quadrant adherent to the anterior abdominal wall were also seen. The adhesions were resected, and an ileostomy was performed. The histopathology of the stricture revealed ulceration, mixed inflammation and presence of cytomegalic intranuclear inclusion bodies within the stromal and endothelial cells, suggestive of CMV colitis (Figure 1). The baby's anti-CMV immunoglobulin (Ig) M and Ig G antibodies were negative. CMV was isolated in the urines and in the blood plasma by PCR in which the amount of viral DNA was 3.5×10^3 copies in plasma and 1.4×10^2 in urine. The baby has received multiple blood products throughout her admission. However, our practice is to administer CMV-negative, irradiated, leukocyte-depleted blood products. Transcranial ultrasonography (USG) was normal, and ophthalmologic examination showed no evidence of CMV retinitis. Antiviral therapy with intravenous (IV) ganciclovir 10 mg/kg/d divided twice a day was started. The baby showed clinical improvement in the following days. She began trophic enteral feeding on day of life 56. A 21-day course of IV ganciclovir was completed, and the baby was discharged from the hospital on day of life 115 with an ileostomy.

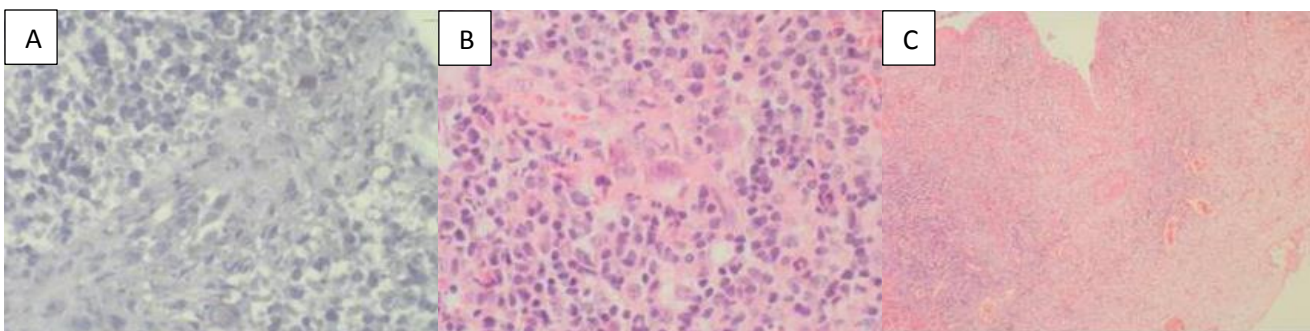


Figure 1: A) Immunohistochemical stain for cytomegalovirus, confirms the viral inclusions are CMV infected cells, HEx20. B) Intranuclear inclusion body in the vascular endothelium in inflammatory cells, HEx20. C) Mucosal ulcers, in the middle, vein thrombosis is seen, HEx4.

DISCUSSION

Cytomegalovirus enterocolitis is rarely seen in newborns and is frequently overlooked. CMV causes a wide spectrum of gastrointestinal (GI) involvement in immunocompromised hosts. Both prenatal and postnatal transmission of CMV can be implicated as a cause of GI involvement.⁵ CMV enterocolitis differs from NEC in that it presents with ulceration progressing to stricture, rather than gangrene and perforation.^{6,7} However, CMV infection is generally not included in the differential diagnosis of an infant with a clinical diagnosis of NEC. Our case presented initially with clinical picture

mimicking recurrent NEC and intestinal obstruction. The diagnosis of CMV enterocolitis was not made until the specimen was examined histologically.

The true incidence of CMV enterocolitis might be higher because screening of CMV is not routinely performed for neonates with refractory NEC and clinicians often overlook the diagnosis. There are few reports on the link between CMV or other enteric viruses and NEC. Marseglia et al reported a case of a full term infant presenting NEC, acquired CMV and post NEC colonic stricture.⁸ Tran et al also reported a case of NEC associated with CMV and Proteobacteria in a 48-day-old,

ex-premature infant.⁹ In a recent study, Skeath et al assessed the presence of candidate viruses in blood or stool of a case series of infants with NEC managed in one surgical centre.⁹ They did not detect any of these viruses in their 17 NEC infants and the authors concluded that CMV was unlikely to be causative for NEC.

By this case, we think that CMV enterocolitis must be considered in all patients with refractory NEC to establish an earlier diagnosis and appropriate treatment. Future research is needed in order to accurately determine the incidence of CMV infection in patients with NEC.

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