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Olgu Sunumu / Case Report

Intractable Diarrhea from Cytomegalovirus Colitis in a Case with Hereditary Spherocytosis

Herediter Sferositozlu Bir Olguda Sitomegalovirus Kolitine Bağlı İntraktable Diyare

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ABSTRACT

Cytomegalovirus (CMV) infection is a common viral infection worldwide, with a frequency as high as 90% in developing countries. Only 10% of primary CMV infection is symptomatic in immunocompetent patients, and it rarely causes specific complications. We report CMV colitis in an immunocompetent adolescent with hereditary spherocytosis and spontaneous rupture of the spleen, who was critically ill with septicaemia and significant non-bloody diarrhoea that responded to specific CMV treatment.

Key words: CMV, colitis, hereditary spherocytosis, immunocompetent

ÖZET

Sitomegalovirüs (CMV) infeksiyonu tüm dünyada sıkça görülen bir viral infeksiyon olup, gelişmekte olan ülkelerde infeksiyonu geçirme oranı %90'lara çıkmaktadır. İmmünkompetan kişilerde primer CMV infeksiyonu %10 semptomatik olup, nadiren organ spesifik komplikasyonlara neden olur. Biz herditer sferositozlu ve spontan dalak rüptürü olan immünkompetan kritik sepsisli ve kansız ishali olan hastada CMV tedavisine yanıt veren CMV kolitli hastayı bildirdik.

Anahtar kelimeler: CMV, kolit, herediter sferositoz, immünkompetan

INTRODUCTION

Cytomegalovirus (CMV) infection causes severe diseases and significant morbidity and mortality in immunocompromised patients. In contrast, it typically runs a more benign course in immunocompetent individuals, who may usually remain asymptomatic[1]. The less common but significant manifestations include colitis, encephalitis, haemolytic anaemia, thrombocytopenia, myocarditis, and pneumonitis[2].

We present CMV colitis in an immunocompetent adolescent with hereditary spherocytosis, who manifested significant intractable diarrhoea requiring gancyclovir treatment.

CASE

A previously known hereditary spherocytosis 15-year-old man was admitted with a 14 day history of acute jaundice and abdominal pain.

Laboratory values on admisson were haemoglobin 8 g/dL, alanine aminotransferase 188 U/L, total bilirubin 49 mg/dL, and direct bilirubin 28 mg/dL. Batın ultrasonography was showed cholelithiasis and choledoch stones and dilated proximally bile ducts. The choledoch stones were dropped by duodenum and external biliary drainage tube was placed into the choledoch. The drainage tube cultures were growed gram positive and negative bacils. The patient developed sign of sepsis such as fever and anorexia. Due to sudden pallor and abdominal distension in the patient, batın ultrasonography was re-performed, and determined spontaneous spleen rupture. Therefore, the patient was underwented splenectomy, cholecystectomy, and liver biopsy. The liver biopsy revealed chronic cholestatic hepatitis.

Three week after admission, the patients's external biliary drain tube was closed. After the tube was closed, severe dehydration secondary watery stool (15-20 times a day) and vomiting that was showed in the patient. A stool test for parasites and culture for the detection of pathogenic organisms were negative. In spite of the specific and supportive treatments were not corrected to the hypoalbuminemia, and severe dehydration findings. Because the patient developed severe malnutrition, a central venous catheter was placed, and parenteral nutrition support was begun.

Gastroduodenoscopy and colonoscopy was performed 25 days after admission. While the upper gastrointestinal mucosa was seen normal, a macroscopic colitis was seen with an erythematous mucosa with edema and diffuse microulceration in the transvers and rektosigmoid colon. The colonic biopsies showed acute inflammation with destruction of crypts with inflammation in the lamina propria consisting of extensive polymorphonuclear leucocytes and eosinophils in clusters (Figure-1). Some biopies showed the typical "owl eye" intranuclear inclusion bodies in epithelial and endothelial cells (Figure-2). Morever, the polymerase chain reaction (PCR) for CMV DNA in blood and colonic biopsy speciments was positive (6142 copy /mL).

Whereas total serum IgE was elevated (>2500 U/mL), and the specific IgE inhale and food was positive. Total serum IgE level was elevated follow-up six months. The other immunoglobuline levels were normal The peripheral blood lymphocyte subsets by flow cytometry revealed normal number of both CD4 and CD8 T cell.

Parenteral gancyclovir (12 mg/kg a day, for 21 days) was administered. Two weeks after parenteral gancyclovir treatment, the patient's complaints were improvemented as the clinically and laboratory. The oral gancyclovir therapy was continued two weeks after discontinued parenteral therapy until serum CMV PCR was negative. Two months after admission, serum CMV PCR was detected negative and the patient resolved completely.

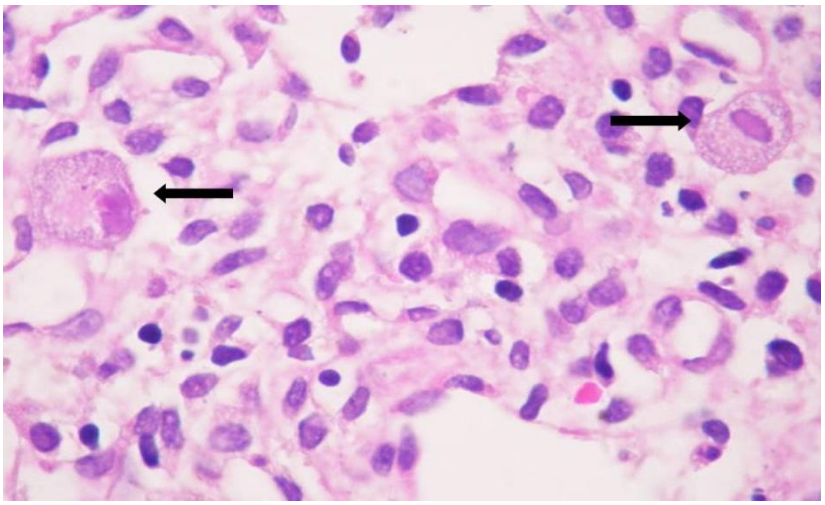

Figure-1. Extensive polymorphonuclear leucocytes and eozinophiles in lamina propria that caused to destruction of crypts (HE x 200).

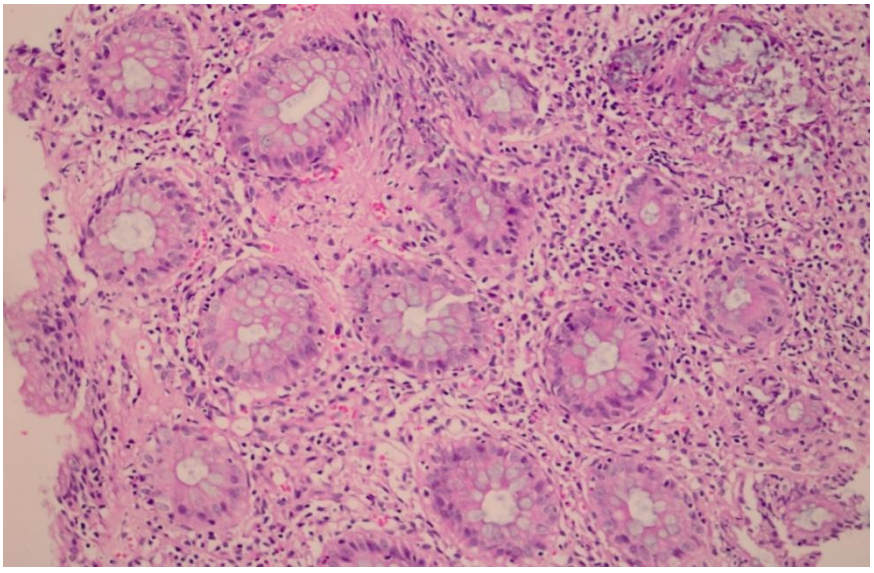

Figure-2. Intranuclear inclusion bodies in epithelial and endothelial cells (Arrows) (HE x 1000)

DISCUSSION

Clinically significant CMV-related disease among immunocompetent hosts is uncommon. Most of these infections occur during childhood. However, seroprevalence studies for CMV among adult populations have shown that the rate of infection ranges from 40% to 100% (1). This suggests that subclinical or mild primary infections are common. Both primary and secondary infection with CMV can occur, although it is often clinically difficult to differentiate them. Reactivation of latent CMV infection is less common, and apart from

those associated with immunosuppressant therapies and immunodeficiency.

The gastrointestinal tract in immunocompetent patients is one of the most commonly affected systems, especially the colon[2]. The reason for the predilection of the colon is unknown. Gastrointestinal infections caused by CMV also have been reported in immunocompetent patients with underlying gastrointestinal disease[3]. In our case had not any underlying colonic disorders according to the history, laboratory and histopathologic findings. The immunological screen tests of the patient were only elevated serum IgE. We believed that this finding can be to secondary sepsis, CMV infection, and atopic susceptibility.

The pateint's diarrhoea was started after the splenectomy. Therefore, we believed that this operation contributed sepsis and severe CMV colitis. Furthermore, we considered that spontaneous rupture of the spleen may be caused by CMV infection. In literature, the majority of the adult cases that were reported both the spontaneous rupture of the spleen due to CMV infection and CMV infection after splenectomy[4,5].

CMV infections in immunocompetent intensive care unit patients have been associated with poor outcomes[6]. Therefore, CMV-related diseases, including colitis, need to be treated, especially among severely ill patients. Early diagnosis and appropriate treatment are essential. Ganciclovir, a guanosine analog that selectively inhibits CMV DNA polymerase, is the antiviral agent[7]. Whitley et al, report a phase II study of ganciclovir in 42 neonates with symptomatic congenital CMV infection, and treatment with ganciclovir was associated with a clinical and histologic response with decreased stool output, weight gain, and histopathologic improvement[8]. In our case, ganciclovir therapy was administered for five weeks period and showed rapid improvement in the second week.

In our case, the manifestations and colonoscopic findings of CMV infections can resemble other common conditions. Colonoscopic findings in the patient were consistent with acute extensive colitis. According to the medical condition and age of the patient, these findings are not unexpected. Therefore, a high index of doubt is important, and adequate biopsies and serological studies are vital for early diagnosis.

In conclusion, CMV colitis, although rare in immunocompetent adolescent, should be considered in the differential diagnosis of severe colitis when other causes fail to explain the course of disease, especially the patients with underwent splenectomy secondary spontaneous rupture of spleen .

Declaration of Conflicting Interests

The authors declare no potential conflicts of interest with respect to the authorship and/or publication of this article.

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