中文題目：在一位紅斑性狼瘡的 26 歲女性患者發生形似狼瘡性腸炎的巨細胞病毒腸炎－病例報告

英文題目：Cytomegalovirus colitis mimicking lupus enteritis in a 26-year-old woman with systemic lupus erythematosus: A case report

作　　者：石佳玉[1]、黃柏豪[1,2]

服務單位：中國醫藥大學附設醫院內科部[1]、中國醫藥大學附設醫院內科部風濕免疫科[2]

Case presentation:

A 26-year-old unmarried woman with history of systemic lupus erythematosus (SLE) presented with abdominal cramping pain after meal accompanied with watery diarrhea, nausea, vomiting and cough with sputum for 1 days.

She had a 4-year history of SLE with initial presentation of polyarthritis with right 3rd finger vasculitis, elevated anti-nuclear antibody(ANA) level (1:1280 homogenous), anti-ds-DNA level (>648 U/ml), anti-SS-A level(>240U/ml), anti-SS-B level(74U/ml), anti-Sm level(146 U/ml), and anti-RNP level(77U/ml). She was on low dose prednisolone and hydroxychloroquine 200mg twice a day initially with good response, but proteinuria around 3gm/24hours was mentioned 1.5 years later. Mycophenolate sodium 360mg twice a day was given from that time on, but proteinuria persisted off and on. One year prior to this time admission, she ever got sudden onset of diffuse abdominal pain with progressive abdominal distension for 3 days, and the final diagnosis was lupus enteritis with typical computed tomography (CT) finding(target sign, Figure 1), which good response to methylprednisolone(MTP) 1gm 3 days pulse therapy and maintenance medication of Mycophenolate sodium 360mg twice a day.

One day prior to this time admission, she got sudden onset of abdominal cramping pain after meal, accompanied with watery diarrhea, nausea, vomiting, and productive cough. At our emergent department, apart from diffuse abdominal pain on palpation the physical examination was normal. Laboratory test revealed elevated level of ESR (95 mm/hr) and hsCRP (2.23 mg/dl), and hemolytic anemia (Hb:6.9g/dl with positive direct Coomb's test). Other lupus serology profiles showed lower complement levels (C3:39.7 mg/dl, C4 4.27 mg/dl), and very high titer of anti-dsDNA (679 U/mL). Chest X-ray (CXR) disclosed left pleural effusion. Contrast-enhanced CT of chest and abdomen revealed left pleural effusion with adjacent lung atelectasis, diffuse bowel wall thickening involving colon and sigmoid colon, and minimal ascites. After admission, she was treated with intravenous (iv) flomoxef for pneumonia and iv hydrocortisone 300mg/day for suspicious lupus enteritis initially. Infection episode was then excluded according to negative finding on sputum culture, urine culture,

and blood culture. She was planned to administer 3 days iv MTP 500mg pulse therapy to control lupus enteritis since 7th days admission. However, deteriorated abdominal pain and fullness were mentioned after the 2nd day of MTP pulse therapy. Repeated contrast-enhanced CT of abdomen showed extended diffuse bowel wall swelling and target sign from ascending colon to rectum area, pleural effusion, pericardial effusion and massive ascites (Figure 2). Abdominal paracentesis around 1000c.c., and thoracic paracentesis from left lung around 200c.c. were performed, which fluid culture all showed negative finding and no malignant cells. Because of poor response to glucocorticoid pulse treatment, we suspected there may be atypical infection. On the 14th day of admission, serum Cytomegalovirus (CMV) pp65 antigen test was positive (2/5*10^{5}PMN), she was therefore started on iv ganciclovir 5mg/kg twice daily and dramatic good response after 2 days ganciclovir. Her symptoms of abdominal fullness and tenderness gradually improved after 14th days treatment of ganciclovir.

She has no further complications on follow up and switched to oral valganciclovir maintenance therapy for another 7 days. Currently she has no more abdominal pain or diarrhea, and continued stasis lupus serology.

Discussion:

Gastrointestinal symptoms are very common in SLE, and lupus enteritis is a major cause of acute abdominal pain in these patients. Characteristics finding of lupus enteritis on CT include bowel wall edema, abnormal bowel-wall enhancement (target sign), dilatation of bowel wall enhancement and ascites. But such CT findings could also be seen in CMV colitis. The distinction between lupus enteritis and CMV colitis is critical because both are life-threatening conditions requiring radically different therapeutic modalities. In lupus enteritis patient with poor response to standard glucocorticoid therapy, we need to think about the condition other than lupus enteritis, such as atypical infection; just as this case, first time illness was typical lupus enteritis, and the second time was CMV colitis. Timely detection of the symptomatic CMV infection with useful antigenemia assay help us to start the proper choice of medication for the patient and to avoid life-threatening condition.

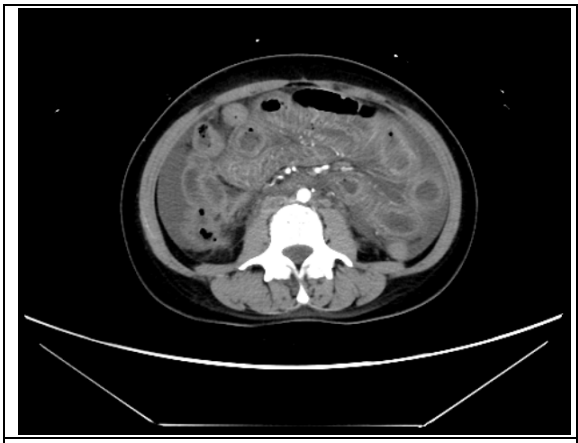	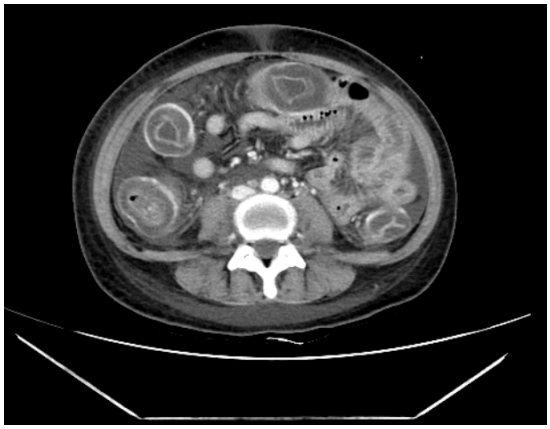
Figure 1.	Figure 2.

Reference:

1. Keigo Ikeda, Shihoko Nakajima, Kana Tanji, Takuya Hirai1, Kaori Uomori, Shinji Morimoto, Shigeki Tomita, Masaki Fukunaga, Naoto Tamura, and Iwao Sekigawa, et al. Intestinal perforation due to hemorrhagic Cytomegalovirus enteritis in a patient with severe uncontrolled lupus nephritis: a case and review of the literature. Rheumatol Int. 2017 Aug;37(8):1395-1399
2. Elisa Gravito-Soares, Marta Gravito-Soares, Maria Augusta Cipriano, Pedro Amaro, et al. Fatal colitis associated with active systemic lupus erythematosus complicated by cytomegalovirus superinfection. BMJ Case Rep. 2017 Nov 28;2017