

— Case Report —

A Case of Systemic Lupus Erythematosus Initially Presenting as Lupus Enteritis Triggered by PregnancyTomoko Miyoshi¹⁾²⁾ and Fumio Otsuka¹⁾*1) Department of General Medicine, Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, Okayama University, Okayama, Japan**2) Center for Medical Education and Internationalization, Kyoto University, Kyoto, Japan***Abstract**

Systemic lupus erythematosus (SLE) predominantly affects young women, with pregnancy commonly triggering flares. Lupus nephritis is the most common type of pregnancy-induced SLE flare. Herein, we report the case of a young woman who developed lupus enteritis and cystitis triggered by pregnancy. The patient was a 28-year-old woman who had frequently presented to the hospital with abdominal pain, nausea, diarrhea, and frequent urination at approximately 30 weeks of gestation when pregnant with her first child. At 37 weeks of gestation, she underwent a cesarean section because of abnormal fetal rotation; however, her symptoms did not improve after delivery. Abdominal computed tomography revealed circumferential bowel edema from the stomach to the rectum, bilateral hydronephrosis, ureteral dilatation, and bladder wall thickening. She was diagnosed with SLE based on fever, polyarthralgia, extremity erythema, lymphocytopenia, anti-double-stranded DNA antibody, low complement level, and proteinuria. The gastrointestinal symptoms were considered to be caused by lupus enteritis. The abdominal symptoms improved with one month of steroid treatment, and the peripheral intestinal edema and bilateral hydronephrosis were improved after one year of treatment. Our findings indicate that pregnancy and childbirth can trigger SLE onset. SLE is difficult to diagnose during pregnancy because of the low anti-DNA antibodies and elevated complement levels.

Keywords

systemic lupus erythematosus, lupus enteritis, pregnancy, case report

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<https://doi.org/10.60227/jhgmeibun.154>**Introduction**

Systemic lupus erythematosus (SLE) predominantly affects young women, with pregnancy being a common trigger. Notably, 20% of SLE flares are caused by pregnancy, with lupus nephritis being the most common type of pregnancy-induced SLE flare. Lupus nephritis flare-ups tend to occur at any stage of pregnancy or during the puerperium period [1]. In this study, we report the case of a young woman with lupus enteritis triggered by pregnancy.

Case

A 28-year-old Japanese woman reported nausea, diarrhea, abdominal pain, and mild frequent urination around the 30th week of pregnancy and was admitted to a hospital. In the

37th week of pregnancy, she underwent a cesarean section because of abnormal fetal rotation. However, she experienced abdominal pain, nausea, diarrhea, and severe frequent urination (approximately 20 times daily) after delivery. Her food intake was severely affected because she experienced vomiting and diarrhea, with the excreted substances resembling digestive fluids. Five weeks after delivery, the patient was admitted to our hospital for further examination. The patient and her baby had been determined to be healthy at every medical examination between the first and third trimesters of pregnancy. During pregnancy, proteinuria was slightly positive. The patient had no significant medical history.

Physical examination revealed that she was 150.5 cm tall, weighed 41.06 kg, and had a body mass index of 18.13 kg/m². Her blood pressure, heart rate, and body temperature

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Table 1. Laboratory deta on admission

Complete blood count		Chemistry		Coagulation	
WBC	4310 / μ l	TP	5.8 mg/dl	PT	13.6 sec
neutrophyl	84 %	Alb	2.4 mg/dl	APTT	27.8 sec
lymphocyte	9 %	T.Bil	1.52 mg/dl	D-dimer	2.4 mg/ml
monocyte	4 %	AST	98 IU/l	Immune system	
eosinocyte	0 %	ALT	78 IU/l	CH50	28 U/ml
basophyl	1 %	ALP	1469 IU/l	C3	75 mg/dl
RBC	408 $\times$ 10 ⁵ / μ l	γ GTP	599 IU/l	C4	13.6 mg/dl
Hb	11.7 g/dl	ChE	150 IU/l	RF	(-)
platelet	43.9 $\times$ 10 ⁵ / μ l	LDH	259 IU/l	ANA	43.5 index
Ulynarisis		CK	5 IU/l	Anti-Sm Ab	44.3 index
protein	(2+)	BUN	3.3 mg/dl	Anti-dsDNA Ab	43.8 IU/ml
occult blood	(-)	Cr	0.34 mg/dl	Anti-cardiolipin Ab	2.7 U/ml
Sediment		UA	3.2 mg/dl	LAC	(-)
RBC	0~1 /hf	Na	139 mEq/l	Anti-RNP Ab	28.2 index
granular casts	19 /wf	K	3.4 mEq/l	Anti-SS-A Ab	73.0 index
		Cl	101 mEq/l	Anti-SS-B Ab	9.4 index
urine protein	0.868 g/day	CRP	0.35 mg/dl	P-ANCA	<10 EU
Ccr	93.3 ml/min			C-ANCA	<10 EU

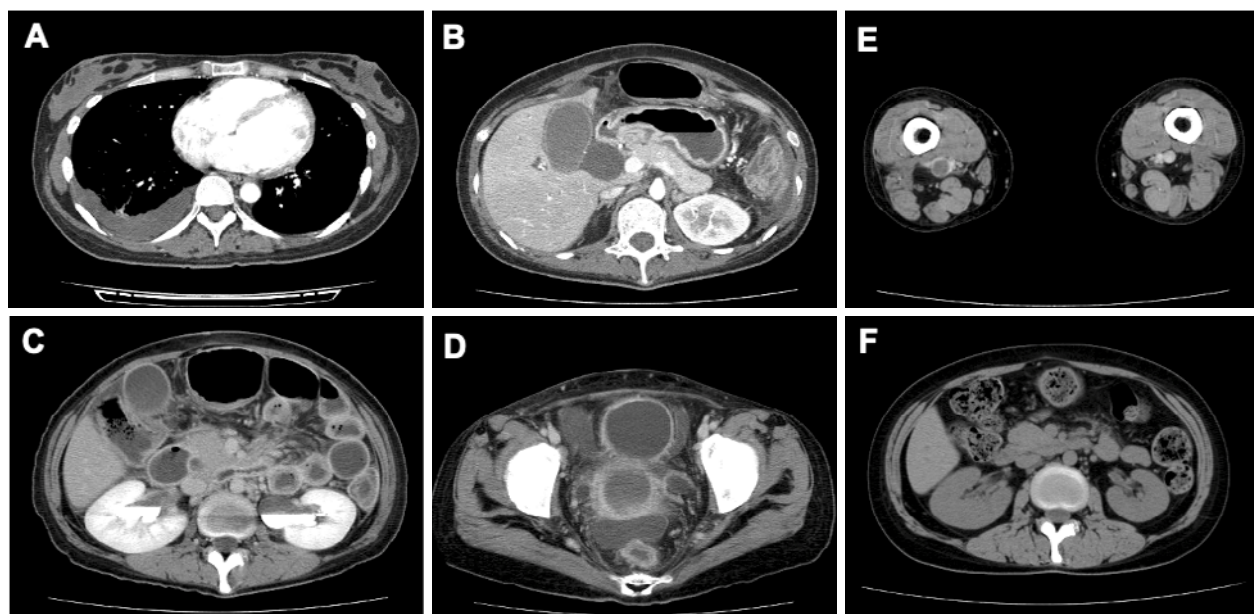


Figure 1. Contrast-enhanced computed tomography.

Examination showed A. pleural effusion on the right side, B. thickening of the stomach wall, C. thickening of the peripheral intestinal wall and bilateral hydronephrosis, D. thickening of the urinary bladder wall, E. thrombosis in the right femoral vein, and F. the bladder and bowel walls after 1 year of treatment.

were 122/78 mmHg, 96 beats/min, and 36.7°C, respectively. No facial erythema, jaundice, pallor, oral aphthous ulcers, or cervical lymphadenopathy were observed. Her pulmonary sounds were clear, her heart sounds were regular, and there were no murmurs. A scar from a recent cesarean section was observed in the middle portion of the lower abdomen. Further abdominal examination revealed the absence of bowel sounds, and generalized abdominal guarding and tenderness were observed. However, ascites were not confirmed. No pretibial edema, purpura, or erythema was observed in the lower extremities. No joint swelling or tender-

ness was observed.

Laboratory findings revealed normal complete blood and platelet counts. However, hypoproteinemia and liver dysfunction were observed (Table 1). Contrast-enhanced computed tomography revealed peripheral wall edema from the stomach to the rectum, bilateral hydronephrosis, ureteral dilation without any obstructive lesions, bladder wall thickening, right-sided pleural effusion, minimal ascites in the pelvic cavity, and thrombosis in the right femoral vein (Figure 1).

After admission, the patient gradually developed fever,

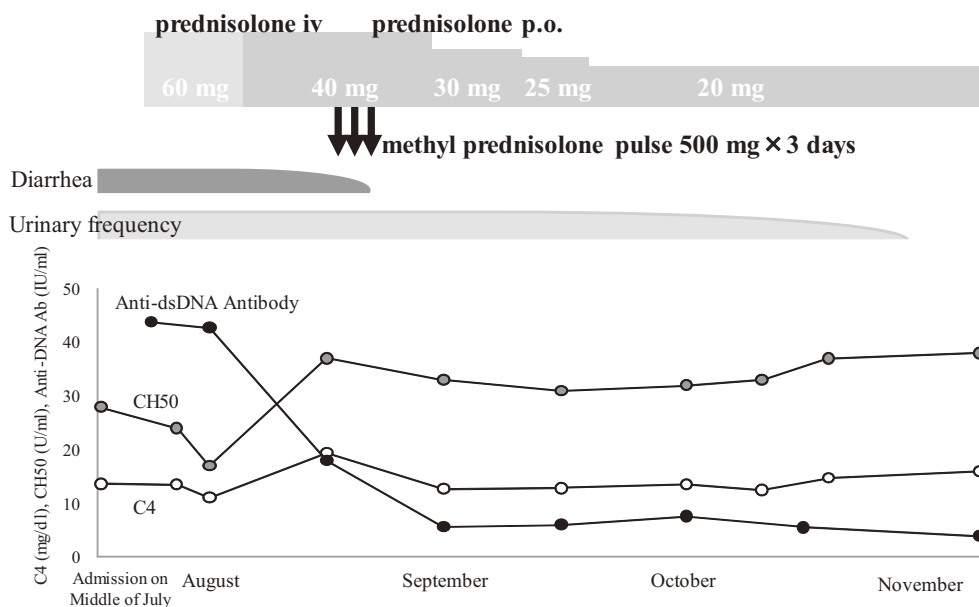


Figure 2. Time course.

The patient's symptoms and levels of anti-ds-DNA antibodies and complement improved with steroid treatment.

Abbreviations: C, complement; CH50, homolytic complement activity; iv, intravenous injection; p.o., per os

polyarthralgia of the wrists and fingers, and erythema of the wrists and lower extremities. She was diagnosed with SLE based on subsequent findings of lymphocytopenia, anti-double-stranded DNA antibody, mildly low complement level, proteinuria, and granular casts in the urine sediment. No anticardiolipins or lupus anticoagulants were detected.

On day 9 of admission, the patient was started on intravenous prednisolone (60 mg/day), which improved the diarrhea but not the frequent urination. Pulsed methylprednisolone (500 mg/day) was added to the treatment routine and administered for 3 days. After approximately 2 weeks, the patient urinated approximately 10 times daily. After 1 month, the peripheral intestinal edema and bilateral hydronephrosis observed on computed tomography improved. Upon discharge from the hospital 3 months after treatment initiation, the patient's appetite had returned to normal, and her abdominal pain had alleviated; however, she experienced nocturnal urination once or twice per night (Figure 2). Regarding proteinuria, a tendency to improve with treatment was observed; however, it persisted at 100 mg/day upon discharge.

Discussion

Notably, 15-60% of individuals with SLE exhibit gastrointestinal symptoms, which are often nonspecific and may be caused by treatment side effects or infection [2]. However, gastrointestinal symptoms can be a rare early manifestation of SLE, with lupus enteritis and intestinal pseudo-obstruction occurring in 0.59-10.7% and 0.5-4% of cases, respectively. It is important to observe these potential complications in patients with SLE [2]. The primary histopa-

thologic feature of lupus enteritis is vasculitis from the submucosa to the subserosal tissue [3]. Lupus cystitis is a relatively rare condition, with an estimated incidence of 0.01% [4]. However, severe cases can result in persistent bilateral hydronephrosis and end-stage renal failure; therefore, the diagnosis and treatment of this condition are crucial. Notably, 81.7% of patients with lupus cystitis also have lupus enterocolitis [5]. Computed tomography revealed symptomatic lupus cystitis with hydronephrosis and urinary bladder wall thickness. In most reported cases of concomitant lupus enteritis and cystitis, gastrointestinal symptoms were the primary presentations, whereas urinary symptoms were rare. Furthermore, steroid therapy was reported to be successful in such cases [6-8]. In the case in the present report, gastrointestinal symptoms, frequent urination, and abnormal findings on computed tomography were improved by steroid therapy.

Pregnancy and delivery can trigger SLE onset, which can be aggravated during pregnancy [9,10]. During pregnancy, fetal-mother interactions and physiological increases in cortisol, progesterone, and estrogen levels can lead to systemic T helper 2 cytokine polarization [11-13]. Suppression of the immune response by T helper 1 cytokines may be essential for fetal survival [7]. However, during pregnancy, the production of C3 and C4 in the liver is enhanced by estrogen; therefore, hypocomplementaemia is absent and may be difficult to diagnose [14,15]. Furthermore, high erythrocyte sedimentation rates and low hemoglobin levels due to hemodilution are common during pregnancy. The patient in the present report also presented with postpartum lupus enteritis, which was difficult to diagnose using anti-DNA antibodies, and the serum complement titer was normal immediately af-

ter pregnancy. Therefore, a diagnosis of SLE should be carefully considered during pregnancy.

In the case in the present report, the peripheral intestinal edema and bilateral hydronephrosis observed on computed tomography were likely caused by lupus enteritis and cystitis, respectively.

A limitation of this report is that it is a case of SLE that developed due to peritonitis after delivery, and further reports on the pathogenesis of postpartum-onset SLE are desirable.

Conclusions

Herein, we report the case of a patient with SLE who presented with lupus enteritis and cystitis in the third trimester of pregnancy. Because gastrointestinal symptoms during and immediately after pregnancy are rare, a potential early manifestation of SLE should be considered. A careful diagnosis of SLE is necessary during and immediately after pregnancy.

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