

Ureteritis associated with systemic lupus erythematosus: a case report

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Chunyan Li*, Fei Huang* , Yu Wang and Mei Tian 

Abstract

We report herein an unusual case of systemic lupus erythematosus in a 35-year-old woman who developed acute abdominal pain while hospitalized. Abdominal computed tomography (CT) scan with enhancement indicated long-segment inflammatory lesions in the right ureter. The patient received spasmolytic and analgesic drugs with poor effect and continued to have persistent severe abdominal pain and signs of peritonitis. We suspected that the patient had acute abdominal disease, but no abnormality was detected during laparoscopic surgery. Therefore, we considered the possibility of right upper urinary tract hydronephrosis; the patient's abdominal pain was relieved after double-J tube implantation. The patient's clinical symptoms improved after hormone and mycophenolate mofetil therapy for 1 year, and all laboratory indicators returned to normal. Reexamination by abdominal CT showed that the long-segment inflammatory lesions of the right ureter had resolved. Early identification and diagnosis are important for ureteritis associated with systemic lupus erythematosus.

Keywords

Systemic lupus erythematosus, ureteritis, double-J tube, immunosuppressant, case report, vasculitis

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Introduction

Systemic lupus erythematosus (SLE) is an autoimmune disease. Injury of the urinary system often manifests as lupus nephritis, and relevant bladder and ureter injury have been reported in previous studies

Department of Nephrology and Rheumatology, Affiliated Hospital of Zunyi Medical University, Zunyi, Guizhou, P. R. China

*These authors contributed equally to this work.

Corresponding author:

Mei Tian, Department of Nephrology and Rheumatology, Affiliated Hospital of Zunyi Medical University, 149 Dalian Road, Huichuan District, Zunyi, Guizhou 563003, P. R. China.

Email: 348820517@qq.com



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with a low incidence.^{1,2} Bladder or ureter injury may be the first symptom of SLE but it often appears during disease complications or recurrence. It is characterized by frequent or urgent urination, inadequate urination, or dysuria, ultimately causing bilateral dilatation of ureters and hydronephrosis. The underlying pathological basis could be that diffuse small vasculitis in SLE leads to neuritis and dysfunction of smooth muscle of the bladder and ureter.³⁻⁵ This case highlights another clinical possibility for SLE with acute abdominal pain. For SLE-related ureteritis with acute abdominal pain, imaging and ureteroscopy should be performed promptly. If necessary, double-J tube implantation may be an effective treatment to relieve obstruction and spasm.

Case presentation

A 35-year-old woman was admitted to our hospital on 13 August 2018 with a systemic rash that had lasted for 2 months and hair loss and systemic pain that had lasted for 1 month. The patient had alopecia, a red maculopapular rash over her entire body, and pain (like that associated with acupuncture) mainly at the end of both feet and fingers. Physical examination showed a scattered red maculopapular rash that did not fade when pressed throughout the body and no edema in the lower limbs. A nervous system examination showed that both feet were sensitive with tingling pain. The results of her blood tests are shown in Table 1. Ultrasonography identified bilateral pleural and abdominal effusion. Abdominal B-type ultrasonography showed polypoid changes in the gallbladder, enlarged spleen, and normal kidneys (Figure 1). Electromyography and nerve conduction tests showed partial nerve damage of the posterior tibial nerve and common peroneal nerve in both lower limbs. On the basis of the history of related medication,

symptoms, signs, and examination, the patient was diagnosed as having SLE, including (1) neuropsychiatric lupus with peripheral neuropathy, (2) lupus nephritis, and (3) pancytopenia.

The patient was treated with methylprednisolone (MP; 80 mg intravenously [IV] once daily) and hydroxychloroquine (200 mg orally twice a day) for 4 days. However, the patient's foot pain worsened and she experienced upper and right middle abdominal pain, which presented as persistent and paroxysmal aggravation. Physical examination showed a soft abdomen, with mild tenderness below the xiphoid and right middle abdomen. We considered acute gastric mucosal lesions, although mesenteric vasculitis could not be ruled out. Magnetic resonance imaging of the head revealed no abnormality. Abdominal computed tomography (CT) scan with enhancement and 3-dimensional reconstruction showed long-segment lesions of the right ureter, indicating a high possibility of inflammatory lesions, and mild hydronephrosis of the right kidney (Figure 2). B-type ultrasound of the urinary system showed hydronephrosis of the right kidney and dilation of the right upper ureter (Figure 3). Therefore, a diagnosis of mesenteric vasculitis could be excluded. The patient's treatment was then adjusted as follows: MP 500 mg IV once daily for 3 days, followed by 80 mg IV once daily. Abdominal pain was treated with anisodamine, atropine, meperidine, and promethazine. On the basis of various clinical manifestations and laboratory data, the patient scored >15 points on the systemic lupus erythematosus disease activity index (SLEDI): 8 points for cranial neuropathy, 4 points for proteinuria, 2 points for rash, 2 points for alopecia, 2 points for low complement, 2 points for high anti-double-stranded DNA antibody, 1 point for thrombocytopenia), indicating a period of severe activity, and we cannot exclude ureteral spastic pain caused by ureteral vasculitis.

Table 1. Results of laboratory tests performed during the patient's hospitalization.

Item	Before treatment	After treatment	Reference range
WBC ($\times 10^9/L$)	2.61	5.58	3.5–9.5
Neu ($\times 10^9/L$)	1.70	3.05	1.8–6.3
Lym ($\times 10^9/L$)	0.34	1.12	1.1–3.2
HB (g/L)	70	115	130–175
PLT ($\times 10^9/L$)	97	240	100–300
ESR (mm/h)	40	16	<21
CRP (mg/L)	141.61	1.8	0.068–8.2
IgG (g/L)	20.70	15	7.51–15.6
C3 (g/L)	0.15	0.42	0.79–1.52
C4 (g/L)	<0.017	0.039	0.16–0.38
ALT (U/L)	45	16	7–40
AST (U/L)	84	23	13–35
Alb (g/L)	29	42.4	40–55
Urinary occult blood test	+++	+ –	–
24-hour urinary protein quantity (g/24 h)	0.738	0.135	0–0.15
BUN (mmol/L)	12.36	5.73	2.8–7.2
Ccr ($\mu\text{mol/L}$)	207	74	30–90
Anti-dsDNA	+++	–	–
Anti-SSA	+++	++	–
Anti-SSB	++	–	–
Anti-Ro-52	+++	+++	–
AnuA	+++	++	–
AHA	++	++	–
ARPA/Rib-P	+++	+++	–
ANA (nuclear particle type)	1:1000	1:100	–

WBC, white blood cell; Neu, neutrophil; Lym, lymphocytes; HB, hemoglobin; PLT, platelets; CRP, C-reactive protein; ESR, erythrocyte sedimentation rate; IgG, immunoglobulin G; C3/4, complement 3/4; ALT, alanine transaminase; AST, aspartate aminotransferase; Alb, albumin; BUN, blood urea nitrogen; Ccr, creatinine clearance rate; anti-dsDNA, anti-double-stranded DNA antibody; anti-SSA and anti-SSB, anti-Sjögren's-syndrome-related antigen A/B; AnuA, anti-nucleosome antibody; AHA, anti-histone antibody; ARPA/Rib-P, anti-ribosomal P protein antibody; ANA, anti-nuclear antibody.

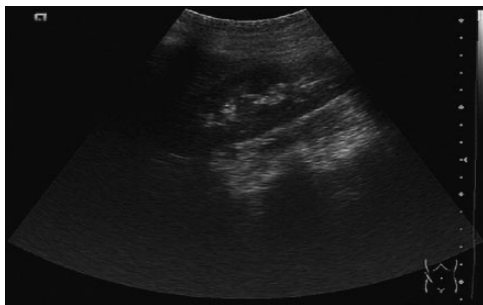


Figure 1. Abdominal B-type ultrasonography showing both kidneys (normal size) with no separation observed in the collecting system and no dilation in the upper ureter.

To alleviate the patient's abdominal pain, we proposed a double-J tube implantation to remove the obstruction.

On 24 August 2018, after providing informed consent, the patient underwent abdominal exploratory surgery and lysis of intestinal adhesions under general anesthesia. Intraoperative exploration found only edema of the right renal capsule. The patient's abdominal pain was thought to be caused by hydrops in the right upper urinary tract. Postoperative right ureteroscopy and double-J internal drainage showed that the bladder mucosa was intact, the bilateral ureteral opening was clear, the ureterscope

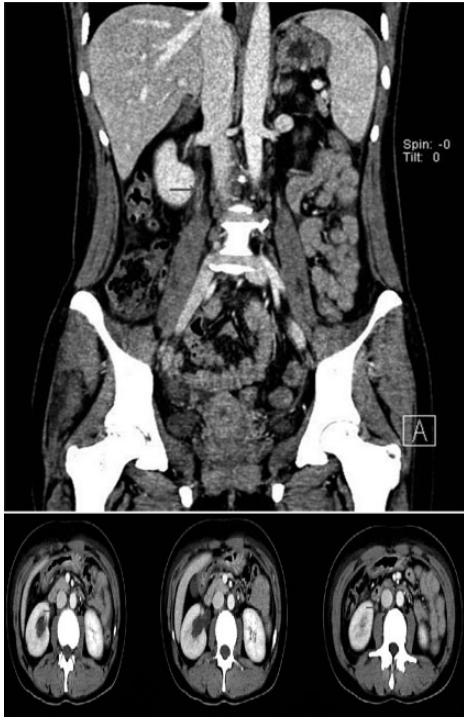


Figure 2. Abdominal computed tomography scan with enhancement and 3-dimensional reconstruction, showing decreased density in the right kidney, long-segment lesions in the right ureter (considered inflammatory lesions), and mild hydronephrosis in the right kidney.



Figure 3. B-type ultrasonography of the urinary system showing right hydronephrosis and dilatation of the right upper ureter.

entered the right ureter to the right renal pelvis, the lumen was patent, and the double-J tube was successfully implanted. After the operation, the patient's

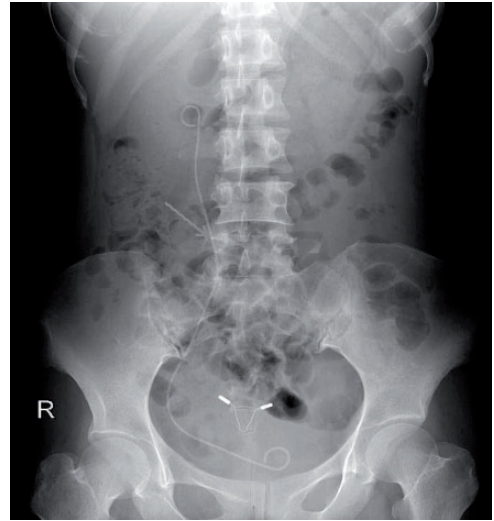


Figure 4. Postoperative abdominal X-ray showing performance of double-J tube drainage in the right urinary system.

abdominal pain was relieved. Abdominal X-ray confirmed the postoperative performance of the double-J tube drainage in the right urinary system (Figure 4). At the same time, MP was replaced with prednisone 40 mg orally once daily plus the immunosuppressant mycophenolate mofetil 0.75 g orally twice a day.

After treatment, the patient's abdominal pain and rash disappeared and the pain in both feet resolved significantly. The double-J tube was removed 3 months after implantation. On 14 December 2018, the patient underwent another abdominal CT scan; no separation was observed in the bilateral renal collecting system and there was no dilation in the upper ureter bilaterally (Figure 5). Routine blood tests and liver and kidney function tests returned to normal (Table 1). A routine urinalysis showed occult blood (+ -) in the urine.

Discussion

Acute attacks of lupus are often accompanied by a variety of gastrointestinal

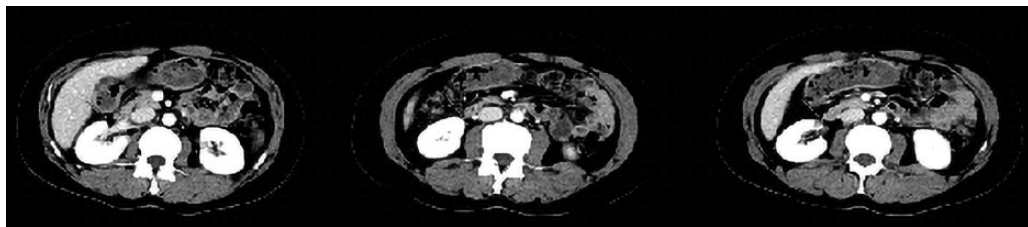


Figure 5. Postoperative abdominal computed tomography scan with enhancement showing no separation in the bilateral renal collecting system and no dilation in the bilateral upper ureter.

symptoms and involvement of the urinary and reproductive systems. Lupus cystitis and damage to the ureters is a relatively rare complication of SLE, and its clinical features remain obscure.^{1,3,6-9} Generally, ureteritis refers to inflammatory changes in the ureter caused by bacterial invasion, and it mainly manifests as lower back pain, hematuria, fever, and urinary irritation. However, the underlying pathological basis of SLE-associated ureteritis is vasculitis. In addition to the clinical manifestations caused by ureteritis, patients often experience clinical features of SLE and multiple positive autoantibodies. A review of clinical data of 18 patients with lupus cystitis admitted to Peking Union Medical College Hospital indicated that the interval from SLE onset to ureteropelvic involvement ranged from 0 to 5 years.¹⁰ The most common clinical manifestations are gastrointestinal symptoms, and urinary symptoms are less pronounced. Prednisone combined with cyclophosphamide has a therapeutic benefit for lupus cystitis.¹⁰ A 13-year-old girl diagnosed with SLE had left hydronephrosis and ureteral stricture and dilatation detected by intravenous pyelography. Cystoscopy showed erythema in the left ureteral orifice, and pathological examination showed vasculitis. Her condition improved after she received gammaglobulin combined with prednisone and azathioprine.¹¹

Ureteritis in SLE with acute abdominal pain as the main symptom has not been

reported to date and it can be easily misdiagnosed. In our case, the patient was in an acute stage of lupus activity and more prone to develop systemic symptoms. Abdominal color ultrasound did not indicate hydronephrosis or ureteral dilatation initially. In our patient, acute abdominal pain occurred suddenly during hospitalization. An abdominal CT scan with enhancement suggested long-segment inflammatory lesions of the ureter and bilateral hydronephrosis. No stone signs or urinary tract stimulation symptoms were found, and the abdominal CT showed no thickening of the bladder wall. However, the basic pathological change of SLE is systemic vasculitis, which can affect all systems of the body. Therefore, the patient's long ureteral inflammatory lesion could have been caused by ureteritis and dysfunction of ureteral smooth muscle related to diffuse small vasculitis.

Although the incidence of ureteral vasculitis in SLE is low, early identification is important, and a combination of hormones and immunosuppressants is recommended.^{10,12-14} If ureteral smooth muscle dysfunction as a result of ureteral vasculitis is suspected to be the cause of acute abdominal pain, prompt double-J tube implantation to remove the obstruction combined with treatment for muscle spasms and pain can avoid the surgical trauma caused by misdiagnosis of acute abdominal disease. Because of the difficulty of a ureteral biopsy, we unfortunately did not obtain

pathological evidence. After active treatment with hormones and immunosuppressants, the patient recovered gradually. Laboratory indicators and tests of multiple systems (blood, kidney, and ureter) returned to normal levels during regular follow-up.

The clinical manifestations of SLE are very complicated. This case highlights the need for vigilance in patients with SLE and acute abdominal pain so that the problem can be identified and diagnosed promptly.

Ethics statement

The study protocol was approved by the Medical Ethics Committee of the Affiliated Hospital of Zunyi Medical University. The patient provided written informed consent for the publication of this case report.


Declaration of conflicting interest


The authors declare that there is no conflict of interest.

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ORCID iDs

Fei Huang  <https://orcid.org/0000-0001-6954-3036>

Mei Tian  <https://orcid.org/0000-0002-1633-2988>

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