Stercoral Colitis in a Patient with Pediatric Onset Systemic Lupus Erythematous: Case Analysis and Review of Literature

Chunchun Gau  
Chang Gung Memorial Hospital  
https://orcid.org/0000-0002-2852-5154

Li-lun Lin  
Chang Gung Memorial Hospital Linkou Main Branch: Chang Gung Memorial Hospital

Chao-Yi Wu  
Chang Gung Memorial Hospital Linkou Main Branch: Chang Gung Memorial Hospital

Jing-Long Huang  
Chang Gung Memorial Hospital Linkou Main Branch: Chang Gung Memorial Hospital  
long@adm.cgmh.org.tw

Case Report

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Abstract

Background: Systemic lupus erythematosus (SLE) is an autoantibody-related disease that affects multiple organs. Stercoral colitis (SC) is a rare type of inflammatory colitis with a high mortality rate in the elderly. We aimed to report the first case of pediatric-onset lupus in a patient complicated by stercoral colitis and to conduct a literature review of patients with SC under 30 years old to persuade a helpful clues for a rapid diagnosis in young ages.

Case presentation: A 28-year-old female presented with a history of lupus, was admitted for severe abdominal pain with unremarkable laboratory data and imaging and found to have stercoral colitis during the surgery. Two years later, the patient underwent Hartman's operation due to ischemia colon. In addition, 10 patients younger than 30 years old (ranged 2 year old to 28 year old) including 7 females and 3 males with a diagnosis of SC were analyzed with clinical presentation, physical examination, laboratory exam, image and treatment. All 10 cases had a favorable outcome without mortality.

Conclusions: Stercoral colitis is a rare but lethal complication and should highlight the importance of a multidisciplinary approach. The differential diagnosis should include stercoral colitis in patients with SLE developing unexplained sharp abdominal pain.

Background

Systemic lupus erythematosus (SLE) is an autoantibody-related disease that affects multiple organs, including those in the cutaneous, musculoskeletal, cardiovascular, renal, pulmonary, hematological and nervous systems. In addition, gastrointestinal presentation has been involved in more than half of SLE patients in their lifetime and is life-threatening[1]. These involvements may be due to lupus enteritis, lupus mesenteric vasculitis, inflammatory bowel disease or the adverse reactions of drugs used to treat SLE[2]. In addition, pediatric-onset lupus patients, accounting for 15–20% of lupus cases, have a severe presentation and are reported to have 19% gastrointestinal manifestations [3, 4].

Stercoral colitis is a rare type of inflammatory colitis with a high mortality rate ranging from 32 to 57%; it occurs mainly in elderly individuals with a history of chronic constipation, and it presents in a moribund state[5]. Chronic constipation may lead to fecaloma formation in the large bowel and cause an increase in intraluminal pressure, which eventually induces bowel wall necrosis and perforation, known as stercoral perforation. Most patients are diagnosed during emergency laparotomy or postmortem[6].

To our knowledge, this is the only reported pediatric-onset lupus patient complicated by stercoral colitis. Hence, the importance of the possibility of this disease occurrence in patients with SLE should be highlighted, even in young adults, and especially in those with early childhood-onset disease. Nevertheless, due to the high mortality rate of stercoral colitis, awareness is promptly required for immediate diagnosis and treatment.

Case Presentation

A 28-year-old female had been diagnosed with pediatric-onset lupus at the age of 8 and presented with fever, malar rash, arthritis, positive antinuclear antibodies (ANAs), and elevated anti-dsDNA antibodies. In addition, she also had episodes of neuropsychiatric SLE (NPSLE) at the ages of 19, 22 and 24 with presentations of seizure, cranial neuropathies of the facial nerve and transverse myelitis with neurogenic bladder, respectively; she had been treated with one dose of 0.5 g/m² intravenous cyclophosphamide pulse therapy at the age of 22. Furthermore, she experienced a brain abscess at 25 years old, and culture yielded Listeria. After the above presentations, she improved without obvious sequelae and regained clear consciousness without cognitive dysfunction or neurologic deficits. Daily medication was maintained with 5 ~ 30 mg prednisolone (i.e., a cumulative dose of approximately 144 g) and 100 mg azathioprine daily for approximately 20 years with regular follow-up at our pediatric rheumatology clinic.

Previous to admission 12 days ago, she visited our outpatient department with complaints of urinary frequency and urgency for one week without fever, abdominal pain, vomiting or diarrhea. The urinalysis results showed pyuria (white blood count, WBC 69/µL), and urinary culture yielded Klebsiella pneumonia (3000) and yeast-like 200000. We treated the patient with oral antibiotics, cefuroxime and fluconazole.

At the time of admission, she presented to our emergency department with developing severe bouts of diffuse abdominal pain and fever for one day. She had a history of constipation for many years, and her last bowel movement was 3 days prior. Upon arrival, she had tachycardia (123 beats per minute) with temperature of 38 degrees Celsius but no hypertension or hypotension (121/70 mmHg). On physical examination, the abdomen was distended with severe tenderness over the lower abdomen, positive peritoneal signs, rebounding pain and hypoactive bowel sounds, but a rectal exam was not performed. The blood laboratory work-up revealed raised inflammatory markers with a C-reactive protein (CRP) level of 77.35 mg/L and a WBC count of 7900/µL with normal serum concentrations of creatinine and electrolytes and normal liver function test results. The SLE activity remained stable with C3 and C4 concentrations of 63.4 and 19.1 mg/dL, respectively, and the anti-dsDNA antibody concentration was 382.0 U/mL; these levels were lower than the concentrations over the past six months, and disease activity, quantified by the SLEDAI-2K score[7], was calculated to be 6, which indicated that a lupus flare was not likely.

The initial abdominal plain film shown in Fig. 1 showed non-specific bowel gas distribution without abnormal calcification or pneumoperitoneum. Ultrasoundography revealed colon dilatation with air-fluid levels and ascites but large amount of residual urine. A Foley catheter was inserted, but abdominal pain was not improved. Empirical intravenous antibiotics with ceftriaxone 1000 gram and metronidazole 500 mg for suspected peritonitis were prescribed, but six hours after admission, due to aggravating abdominal pain, repeated blood tests proved that inflammatory markers had progressed with a CRP level of 197 mg/L. Subsequent abdominal computed tomography (CT) indicated substantial fecal impaction and colon dilatation, along with colon wall thickening and pneumatosis coli, as shown in Fig. 2. Exploratory laparoscopy indicated poor perfusion of the upper rectum with pericolonic brin with turbid ascites in the pelvic cavity without identified perforation. During the operation, rigid sigmoidoscopy revealed ischemic rectal mucosa. The ischemic rectum was resected, and primary anastomosis of the descending colon and rectum was performed with a diverting loop transverse colostomy over the right quadrant of the abdomen (Fig. 3). The pathology shown in Fig. 3 demonstrated focal ischemic changes, intensive mucosal injury and focal transmural necrosis of the large...
In the end, all cultures were negative, and the patient was discharged smoothly 46 days after the operation. However, 2 years later, the patient encountered abdominal pain again with shock status, while abdominal CT revealed inferior mesenteric artery occlusion and descending sigmoid colon ischemia. During the reoperation, ischemic colitis with gangrene changes was observed, and the Hartmann procedure was conducted. Since then, the patient has recovered smoothly under our outpatient follow-up.

**Literature review-based case series**

The medical records, including clinical notes, physical examination results, laboratory results and imaging series, were reviewed by Lin LL and Gau CC from a tertiary center in Taiwan. This study was approved by the Ethics Committee on Human Studies at Chang Gung Memorial Hospital in Taiwan, R.O.C. (IRB 201601678A3C501). Informed consent was obtained from the patient. We reported this case according to the CARE (for Case Reports) guidelines[8].

In a review of the literature by Lin LL and Gau CC, the clinical presentations, image findings, management strategies and outcomes were analyzed. We used the Medline subheading keywords “systemic lupus erythematosus (SLE)”, “stercoral colitis” and “stercoral perforation” to search for studies in PubMed published between 1965 and August 2020 in English. Relevant articles and additional references were found by checking the citations in the articles retrieved.

To date, fewer than 200 cases of stercoral colitis or stercoral perforation have been reported, and only 10 patients (3 males[9–11] and 7 females[12–17]) less than 30 years old have been reviewed in the literature based on PubMed. Patients aged 2 to 28 years old are analyzed in Table 1. According to the table, all of the young adult or adolescent patients experienced psychiatric problems or were under substance use except our patient, while only half of the children had underlying or drug problems. According to the presentations, most patients (8 of 10) complained of abdominal pain, but only 40% of patients presented with fever. According to the physical examinations, signs of peritonitis were only observed in six of the cases (case 1, case 3, case 5, case 8, case 9 and case 10). According to the laboratory work-up, 6 patients had remarkable inflammatory processes, 2 patients had metabolic acidosis, and one patient had both anemia and acute kidney injury. According to Kumar et al.’s [18] indication, the diagnostic sign of stercoral perforation and the CT feature of fecal protrusion through the colonic wall were found in seven of the cases, and extraluminal air was noted in half of the cases. Seven of the patients had undergone surgery, and all of them were alive.
| Case    | Patient | Pass history                        | Presentation | Vital sign | Physical finding                  | Laboratory | Image                              | Operation                        | Course                          |
|---------|---------|-------------------------------------|--------------|------------|------------------------------------|------------|------------------------------------|----------------------------------|---------------------------------|
| Our case (Case 1) | 28 year female | Lupus, NPSLE, brain abscess | severe abdominal pain with fever for 3 day | BT 38 °C HR 123 bpm BP 121/70 mmHg | Abdomen tenderness, peritoneal sign | inflammatory process | X-ray, non remarkable | CT: fecal impaction and colon dilatation with pneumatisis | Diverting loop transverse colostomy | Discharge 46 days postoper  |
| Hussain et al[13] (Case 2) | 28 year female | Opioids for chronic pain | diffuse, chronic abdominal pain, nausea, anorexia, and the complete inability to defecate for 6 weeks | Normal | Abdomen soft, diffusely tender, hypotonic bowel sound without peritoneal sign | Unremarkable | CT: gastric distension and a moderate to large amount of colonic stool | No | Endoscopic decompression and discharge |
| Brown et al[12] (Case 3) | 27 year female | 3-year heroin use and depression medication | 1-week abdominal pain and 12-hour vomiting | HR 160 bpm RR 30/min SpO2 100% BP 90/60 mmHg | Abdomen distended and peritonitic | Metabolic acidosis, AKI, inflammatory process, anemia | X-ray: no subdiaphragmatic air | CT: portal venous gas and free intraperitoneal fluid and gas and massive fecal distension | Hartmann's procedure perforation in the sigmoid colon | Discharge weeks postoper  |
| Canders et al[9] (Case 4) | 26 year male | Anxiety around using the restroom | Cramping abdominal pain in the lower quadrants and shortness of breath | BT 38.3 °C HR 120 bpm RR 16/min SpO2 96% BP 109/81 mmHg | Abdomen distended and nontender, with stool palpable in the left lower quadrant and normal bowel sounds | Rectal exam: hard stool | X-ray: dilated colon with severe fecal impaction without pneumoperitoneum. | CT: fecal impaction with bowel ischemia | No | Intravenous fluids, oral laxatives, water enemas | Discharge hospital discharge |
| Lundy et al[10] (Case 5) | 25 year male | Chronic constipation Narcotics | severe, diffuse abdominal pain that began three hours | tachycardic, diaphoretic, and tachypneic | Abdomen rigidity with involuntary guarding. | N/A | N/A | A resection of distended sigmoid and rectum | an unexpected recovery and discharge |
| McHugh et al[14] (Case 6) | 17 year girl | Chronic constipation Eating disorder | 24-hour history of left-sided abdominal pain | HR 140 bpm BP 100/77 mmHg | Abdomen distended and tender across her lower abdomen without guarding or rebound | Anemia Inflammatory process | X-ray: severe fecal throughout the large intestine, without free air | CT: colonic perforation, with sigmoid colon dilated to 11 cm | Total colectomy with end ileostomy | Repeated laparotomy |

NPSLE neuropsychiatric systemic lupus erythematosus, Yr year-old, HR heart rate, RR respiratory rate, SpO2 saturation oxygen, BP blood pressure, AKI acute kidney injury, N/A not applicable
Stercoral perforation represents a rare but lethal complication of constipation and fecal impaction. Since the first report in 1894, fewer than 200 cases have been evaluated prior to our patient[19]. Maurer et al assert that stercoral perforation persists in 1.2% of all emergency colorectal procedures and 3.2% of all colonic perforations[6]. Most cases of colonic perforation are associated with diverticulitis, malignancy, trauma, inflammatory bowel disease, ischemia, infection, or iatrogenic conditions[20]. In a retrospective review, stercoral perforation occurred in 81% of the patients, particularly associated with a long history of chronic constipation and typically in elderly or bedridden patients with a median age of 62 without propensity for either sex.[19] Patients taking antacids, codeine-containing compounds, narcotics, nonsteroidal anti-inflammatory drugs (NSAIDs), major tranquilizers, or tricyclic anti-depressants have been reported to have a higher risk of stercoral perforation[21, 22]. This is almost certainly due to the propensity of these drugs to travel slowly and transiently through the bowel rather than due to a direct effect on the bowel mucosa. However, some have speculated that NSAIDs, by abolishing the cytoprotective effect of prostaglandins, may compromise intestinal integrity[23].

Upon exploring the published English literature (PubMed search from 1965 through August 2020), only two cases of stercoral colitis in patients with SLE have been reported[24, 25], while constipation is demonstrated in 6% of lupus patients[26]. In one case report, a 45-year-old woman with SLE presented with epigastric pain for 12 hours. She also had other significant comorbidities, including sarcoidosis, hypertension, and a previous history of congestive heart failure. The author thought that the cause of stercoral perforation in the sigmoid colon was possibly related to nonsteroidal anti-inflammatory drug (NSAID) use[24]. The other case report addressed a 44-year-old female presenting with stercoral perforation but a complication of SLE attributed to long-term steroid use who had a worse prognosis[25]. In addition, our patient is the first pediatric-onset lupus patient complicated by stercoral colitis without a disease flare, and

NPSLE neuropsychiatric systemic lupus erythematosus, Yr year-old, HR heart rate, RR respiratory rate, SpO2 saturation oxygen, BP blood pressure, AKI acute kidney injury, N/A not applicable

| Case   | Patient | Pass history | Presentation | Vital sign | Physical finding | Laboratory | Image | Operation | Course          |
|--------|---------|--------------|--------------|------------|-----------------|------------|-------|-----------|----------------|
| Proulx et al[15] | 9 year girl | Chronic constipation | Nausea, vomiting, and diarrhea for several hours | BT 97.2°F (36.2°C) HR 154 bpm RR 22 bpm SpO2 95% BP 70/50 mmHg SpO2 95% | Abdomen palpable suprapubic mass, mild generalized tenderness without peritoneal sign | Leukocytosis | CT large stool burden in the rectum, comprising a mass of approximately 7 cm | No | Fluid supplement manual decompa antibiotic anorectal irritation 6 hours | Discharge hospital c |
| Park et al[11] | 6 year boy | Ehlers-Danlos Syndrome | abdominal pain for 4 days without nausea, vomiting, or fever, Tachycardic Normotensive | Abdomen tenderness to percussion, pain with movement, and voluntary guarding | Normal | X-ray: unremarkable | Loop colostomy perforation on the lateral mesenteric border of descending colon | Discharge 11 postoper | But reope 14 month later |
| Huang et al[17] | 4 year girl | Chronic constipation | sudden and severe abdominal pain | Fever | Abdomen diffuse peritonitis | Leukocytosis | X-ray: Subdiaphragmatic air | Segmental colectomy Perforation at anti-mesocolic site over mid-sigmoid Colon | No complica |
| Omran et al[16] | 2 year girl | Overdose of Ibuprofen | a 3-day history of cough, fever and general aches | BT 39.1°C HR 155 bpm RR 50/min BP 90/34 mmHg | Abdomen distended, with guarding and rebound tenderness | Leukocytosis | Coronal anteroposterior CT : free air and abundant fecal in the pelvis Double-barrel colostomy Perforation on the antimesenteric side of the sigmoid colon | Discharge week afte surgery |
several causes were postulated. First, stenosis of the superior mesenteric arteries[26] and persistent inflammatory vessel status in the gut mucosa[27] have been found in lupus, and the imbalance of SLE patients’ bowel mesenteric blood flow or damage to the gut mucosa should be considered to cause ischemic changes in the bowel. Second, gut microbiome alterations in SLE patients may also contribute to gut inflammation[28, 29]. Third, but most importantly, the consequence of neurologic events (NPSLE and brain abscess) may lead to chronic constipation and this catastrophic result. In terms of the multiple manifestations of lupus described above, making a correct diagnosis is difficult among our population.

A variety of causes of abdominal pain may suggest misdiagnosis in lupus patients, such as lupus enteritis, primary peritonitis and chronic lupus peritonitis[30, 31]. Lupus enteritis, which includes mesenteric arteritis or vasculitis, gastrointestinal vasculitis and intestinal vasculitis, is a rare but life-threatening complication of SLE that can present insidiously with postprandial abdominal pain due to chronic mesenteric ischemia[30]. Primary peritonitis secondary to SLE can develop rapidly during a lupus flare and can present with abdominal pain suspicious for abdominal surgery, although symptoms may be masked by concurrent immunosuppressive use[30]. Chronic lupus peritonitis usually presents with gradually developing ascites that is painless.[32] Given the challenge of misleading diagnosis mentioned previously, stercoral colitis can be easily neglected but requires prompt treatment due to the rapidly progressive course of the disease.

According to Maurer et al.’s[6] definition, there are three diagnostic criteria: (1) round or ovoid colonic perforation more than 1 cm in diameter on the antimesenteric side; 2) fecalomas observed in the colon, protruding through the perforation site or lying within the abdominal cavity; and 3) pressure necrosis or ulceration with chronic inflammatory reactions around the perforation site. In addition, mortality has been reported to be 32%-60% regardless of age[6]. In our case analysis, all of our patients met the diagnostic criteria, were alive and had been discharged, and young age may be attributed to the favorable outcomes.

The early diagnosis of stercoral colitis is still a dilemma. According to a previous review, fever, peritoneal signs, elevation of inflammatory markers, metabolic acidosis, and electrolyte imbalance are not noted in all cases[16, 33]. In addition, urinary retention, incontinence or frequency may mimic the early signs of fecal obstruction[34]. Sometimes, a tubular mass can be palpable in the left lower quadrant, as a stool-filled rectosigmoid and rectal exam can disclose stool impacted proximally located in the sigmoid colon[35, 36]. In our review, one of the patients, at the age of two, could not verbalize the abdominal discomfort; thus, it may be difficult to confirm the diagnosis. In our presentation, our lupus patient had urinary tract infection and retention episodes, which should be one of the early signs of chronic constipation. Our review is also in line with a previous study showing that peritoneal signs and inflammatory laboratory data could only be observed in 60% of cases.

Although previous experts have mentioned in their reports that pain during upright abdominal X-ray can offer clues regarding stercoral colitis or perforation, including distention of the colon at the site of impaction, calcified fecaloma, or free air, fecal matter may obscure these findings[6, 37]. The pivotal diagnostic role of radiologic investigation, including CT, has been mentioned for decades, including large fecaloma with distention > 6 cm of the affected colon, wall thickening > 3 mm of the affected colon, pericolonic fat stranding, mucosal discontinuity, free fluid, pericolonic abscess and extraluminal gas bubbles, which suggest perforation[18, 37–39]. In addition, ascites can be found on CT and may be a result of severe systemic inflammation[40]. However, in our review of patients with a young age, remarkable findings on plain abdominal X-ray could not be found in most patients, and only half of the patients displayed perforation on CT images.

According to Wu et al.’s research, 52% of patients can be treated nonoperatively with a bowel regimen[40]. However, if peritonitis occurs, an emergency laparotomy should be arranged for the possible association of stercoral perforation, and stercoral colitis can be diagnosed straightforwardly by pathology and intraoperatively[6]. The usual operation is Hartmann’s procedure with resection of the possible lesion[41]. Intraoperative colonoscopy should be utilized to identify if there are additional stercoral ulcers and to remove a specimen of the altered or dilated colon to prevent another event of colon stercoral perforation[17]. The sigmoid colon and rectum, particularly the rectosigmoid junction, are the parts of the colon most susceptible to stercoral colitis. There are several reasons for enhancing stone-hard fecaloma formation, including the decreasing water content of stool where feces are dry and inspissated, the relatively narrow diameter and the poor blood supply[17]. The poor blood supply at the rectosigmoid junction is especially defined as Sudeck’s point with insufficient or absent Anastomosis between the superior rectal artery and inferior mesenteric artery branch at the watershed area[17, 42]. Additionally, pathology can demonstrate mucosal hemorrhage, submucosal congestion, sharp demarcation without undermining at the ulcer margins and transmural necrosis on the perforated side.[33, 40] In our case analysis, 30% of patients were treated conservatively rather than surgically, but some patients had to receive a second operation. One of these was a patient with Ehlers-Danlos syndrome, a connective tissue disease that may lead to poor wound healing. The other was our lupus patient. Although the lupus condition was stable, bowel inflammation may also have contributed to the poor tissue recovery and the need for a second operation. The presented pathologic specimens also confirmed the diagnosis of stercoral colitis.

**Conclusion**

It is important to identify SLE patients with severe abdominal pain who may be at risk of stercoral colitis or perforation. Stercoral colitis is a rare yet lethal complication that can occur in a 28-year-old female patient, and the differential diagnosis should highlight the importance of a multidisciplinary approach. In addition to appendicitis, peritonitis, or vasculitis, the differential diagnosis should include stercoral colitis in patients with SLE who develop unexplained sharp abdominal pain.

**Abbreviations**

ANAs: antinuclear antibodies, CRP: C-reactive protein, CT: computed tomography, NSAIDs: nonsteroidal anti-inflammatory drugs, NPSLE: neuropsychiatric SLE, SC: Stercoral colitis, SLE: Systemic lupus erythematosus, WBC: white blood cell

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Declarations

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not applicable

Authors' contributions:

Gau CC and Lin LL carried out the case analysis studies, participated in the sequence alignment and drafted the manuscript. Wu CY and Huang JL participated in the design of the study and performed the statistical analysis. Huang JL conceived the study, and participated in its design and coordination and helped to draft the manuscript. All authors read and approved the final manuscript.

Competing interest:

none

Ethics approval and consent to participate:

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Consent for publication:

Written informed consent was obtained from the patient

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**Figures**
Figure 1

(A) The initial abdominal plain film showed non-specific bowel gas distribution without abnormal calcification or free air  (B) The ultrasonography of abdomen revealed colon dilatation with air-fluid level and ascites.

Figure 2

Abdominal CT revealed much fecal impaction and colon dilatation along with colon wallthickening and pneumatosi coli (arrow)
Figure 3

(A) An exploratory laparoscopy revealed poor perfusion of upper rectum with pericolonic fibrin and turbid ascites in pelvic cavity (B) The section showed ischemic change, intensive mucosal injury, and focal transmural necrosis of the large intestine (arrow) (C) The specimen showed focal ischemic change and transmural necrosis of the large intestine.