


## CASE REPORT

# Systemic lupus erythematosus as an unusual cause of intussusception in Sudanese woman: A case report

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## Abstract

Abdominal pain does have a wide differential diagnosis, however, gastrointestinal involvement is not unusual in SLE, and this report is to describe an unusual case of intussusception due to SLE in a Sudanese woman.

## KEYWORDS

adults, intussusception, lupus nephritis, systemic lupus erythematosus

## 1 | INTRODUCTION

Systemic lupus erythematosus (SLE) is a chronic autoimmune disease characterized by multisystem inflammation, antibodies to cytoplasmic and nuclear antigens, protean clinical manifestations, remitting, and a relapsing course. Intussusception is an uncommon diagnosis in adults; however, a reported case of intussusception as an early presentation for SLE has been described. The aim of this report is to describe an unusual case of intussusception due to SLE in a Sudanese female. A middle-aged Sudanese woman has been presented complained about multiple joint pain, skin rash, chest pain, hair loss, severe

abdominal pain associated with abdominal distension, bloody diarrhea, and vomiting. Lab investigations and computed tomography abdomen revealed the patient have an intussusception on top of SLE. The patient well responded after received prednisolone, hydroxychloroquine, azathioprine, lisinopril, tonics bone, and gastric protection. Now, she is on regular treatment along with hemodialysis as is SLE complicated to lupus nephritis.

Systemic lupus erythematosus is a complex autoimmune chronic inflammatory disorder with an extensive cascade of symptoms, the disease can involve the kidney, lung, nervous system, and heart.<sup>1</sup> Abdominal pain does have a wide differential diagnosis, however,

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gastrointestinal involvement is not unusual in SLE.<sup>2,3</sup> A lot of causes of abdominal pain have been described in lupus patients such as lupus enteritis drug reactions and infections due to steroids.<sup>4,5</sup> In addition, the likelihood of SLE as a contributing factor in jejunal diverticulosis has been reported.<sup>6</sup>

An intussusception itself as a complication or presenting symptom of SLE is rare, until now, there are no more than seven cases have been reported in SLE.<sup>4,6</sup> Two of those cases were secondary to mesenteric vacuities and five cases had an intussusception as an initial presentation of SLE.<sup>7</sup> In this case, we are reporting a case of SLE with intussusception as the initial presentation.

## 2 | CASE REPORT

A 40-year-old Sudanese female presented with complaints of facial skin rash, bilateral small and large joints pain, swelling, palpitations, shortness of breath, pleuritic chest pain, hair loss, and muscle pain. Soon after, she has been admitted to our rheumatology unite at Omdurman military hospital, Khartoum Sudan, the patient developed acute severe cramping periumbilical abdominal pain associated with abdominal distension,

vomiting, and infrequent bloody diarrhea. Her examination showed high blood pressure (150/100), pulse rate of 100, and temperature of 37.5, and she has malar rash sparing the nasolabial folds with hyperpigmented patches, no rashes in other sites, broken hair in the frontal and temporal areas, and no nail change was seen. Cardiovascular examination revealed short localized systolic murmur in the mitral area, and chest examination indicated decreased air entry bilaterally with dull percussion notes, her abdomen was distended, tender left periumbilical mass. Joints examination showed active synovitis involve meta-carpophalangeal joints bilaterally, knees, and shoulder pain with passive and active movements. General investigations were done (Table 1) suggestive of SLE, and the patient abdominal computer tomography (CT) showed rounded lumbar mass with a double layer concluded intestinal intussusception. The patient was seen by the surgical team and underwent laparotomy with bowel resection, and the biopsy examination report multiple fragments measuring 16 cm with attached binding of 12.5 cm showed congested vascular channels and hypertrophied muscular fibers with no granulomas or malignancy saw. All these features were consistent with a diagnosis of intussusception, no features of granuloma or neoplastic cells. The patient was

Investigations	Results	References
WBC	6.8 cells/ $\mu$ l	4–11 $\times 10^9$ /L
HB	7.3 g/dl	
Platelet	127 cells/ $\mu$ l	150–450 cells/ $\mu$ l
ESR	60 mm/h	Normal reference up to 20 mm/h
Serum urea	32 mg/dl	5–20 mg/dl
S. Creatinine	1.1 mg/dl	0.5–1.1 mg/dl
CRP	20	Less than 10 mg/L
S. Albumin	2.9 g/dl	2.4–4 G/dl
Total protein	5.9 G/dl	6–8.3 G/dl
Total bilirubin	0.4 mg/dl	0.2–1.3 mg/dl
ALT	33 U/L	10–130 U/L
AST	27 U/L	10–34 U/L
ALP	105 U/L (24–147 UL)	24–147 UL
Urine general	Albumin, no casts 7–9 pus	
Direct Coombs Test	Negative	
Antinuclear antibodies	positive Antidouble stranded DNA Antihistone positive AntiRibosomal p protein positive Anti nucleosome borderline (Picture attached)	

**TABLE 1** Lab results performed for the patient

diagnosed with a case of systemic lupus erythematosus complicated with intussusception and the possibility of lupus nephritis postoperatively. The patient received prednisolone 30 mg with tapering HCQ 200 mg/day, azathioprine 50 mg twice per day, lisinopril 20 mg/day, tonics bone and, gastric protection, although the patient therapy showed good response, she is still on regular hemodialysis because renal biopsy done to the patient showed grade 4 lupus nephritis.

### 3 | DISCUSSION

Systemic lupus erythematosus is a multisystem autoimmune disease in which the deposition of immune complexes and pathogenic autoantibodies lead to a wide variety of symptoms.<sup>8</sup> SLE is a kind of highly heterogeneous disorder, and the performance of each patient is varying. Steroids are still the first-line treatment options for SLE. Because of his wide anti-inflammatory and immunosuppressive effects.<sup>9</sup> Abdominal pain in SLE patients can occur for multiple reasons including severe vasculitis presenting as gastrointestinal perforation or mesenteric thrombosis, acute pancreatitis, cholecystitis, lupus mesenteric vasculitis (LMV), and hepatitis.<sup>10</sup> Intussusception is a condition in which the proximal segment of the intestine telescopes into the distal part of it.<sup>11</sup> Although intussusception is uncommon in adults and more likely to occur during childhood, adult have an underlying pathology in approximately 90% of cases. Intussusception in the small intestine is mainly due to secondary causes either due to extraluminal or intraluminal lesions (Meckel's diverticulum, lymphoma, lipoma, postoperative, adhesions, and metastases), however, intussusception in the large bowel is mostly due to malignant etiology.<sup>12</sup> The surgical specimen can show macroscopical aspects of intussusception in the intestine (Figure 1) and the microscopical histopathology features (Figure 2).

At usually CT scan for the abdomen can identify the potential cause. The presence of bowel with bowel configuration with or without the existence of adipose tissue and mesenteric vessels is pathognomonic for intussusception.<sup>5</sup>

The pathogenesis of the intussusception in our patient related to SLE could be mostly the initial manifestation of the disease. Only seven cases of intussusception in patients with SLE have been described in the literature. Four cases were secondary to LMV,<sup>7</sup> one of them, lymphadenopathy was the main cause, one was related to Burkitt's lymphoma, one was secondary to changes in the peristalsis of the edematous small intestine.<sup>4,7,13,14</sup>

The first report has been published by Hermann<sup>12</sup> on a 5-year-old child with known SLE who had an

intussusception as a complication to LMV. The exact pathology and mechanism of LMV causing intussusception is not fully understood.<sup>4</sup> A potential explanation is that vascular necrosis occurred by diffuse vasculitis with partial devitalization of the intestinal segment, this can cause the interruption of the normal neuromuscular function, with concomitant intussusception and the possibility of venous infarction necessitating bowel resection.<sup>4</sup>

Intussusception is a very serious complication carrying a high mortality rate,<sup>15</sup> CT abdomen has a central role in the early detection of LMV and delivers precise imaging of the lesions. However, the detection of the underlying causative factor of the intussusception can be challenging due to differentiation from bowel wall edema may not be possible (Figure 3). In our case, the CT abdomen of the patient showed bilaterally pleural effusion, moderate-free peritoneal fluid collection (Ascites), and rounded lumbar mass with double layers suggestive of intussusception.

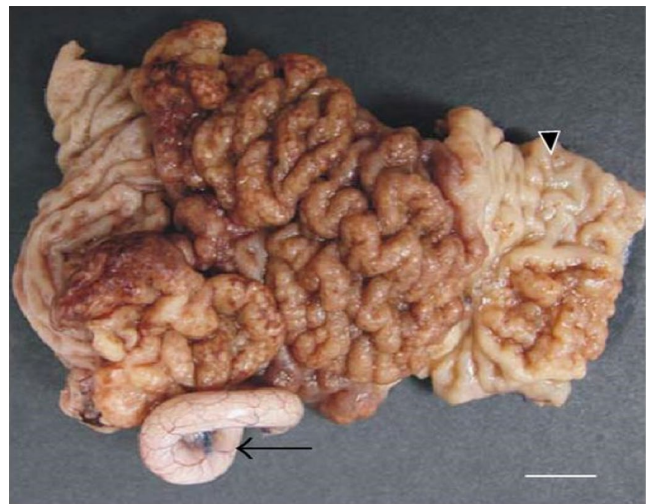


FIGURE 1 Morphological features of ileocecal surgical specimens with intussusception<sup>17</sup>

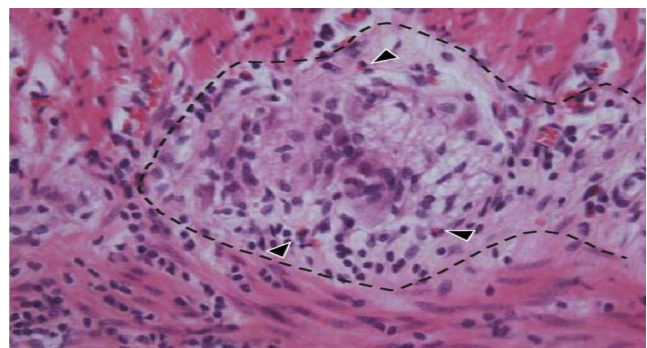
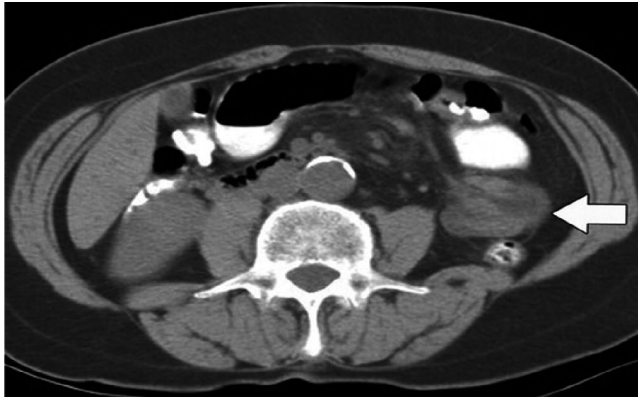


FIGURE 2 Tissue section of the terminal ileum demonstrates the plexus myentericus (dotted line) and several infiltrating lymphocytes and eosinophils (arrowheads)<sup>17</sup>



**FIGURE 3** CT scans shows an intussusception in the abdomen, the mass is isoattenuating relative to bowel wall edema, making differentiation difficult<sup>18</sup>

Another report described a known case of lupus with fungal infection with intussusception. It is not fully clear whether there is an association between these conditions.<sup>16</sup> However, in our case intussusception occur few days after the patient present complaining of the rheumatological features.

In conclusion, a 40-year-old female presented with multiple joints pain, skin rash, chest pain, hair loss, and intussusception, and bowel resection was done and they found no evidence of malignancy, infection causes, or mesenteric vasculitis. Therefore, it is more likely that the intussusception was a secondary cause of SLE in this patient.

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#### CONFLICT OF INTEREST

None.

#### AUTHOR CONTRIBUTIONS

All authors contributed equally.

#### ETHICS APPROVAL

Obtained.


#### CONSENT

Obtained.

#### DATA AVAILABILITY STATEMENT

All the data used in the study are available from the first and corresponding author on reasonable request.

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