

Intussusception as a Rare Gastrointestinal Manifestation of Systemic Lupus Erythematosus: A Report of Two Cases and Literature Review

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How to cite this paper: Mirza, H.I. and Banks, S. (2026) Intussusception as a Rare Gastrointestinal Manifestation of Systemic Lupus Erythematosus: A Report of Two Cases and Literature Review. *Case Reports in Clinical Medicine*, 15, 139-146.
<https://doi.org/10.4236/crcm.2026.154019>

Received: March 9, 2026

Accepted: March 30, 2026

Published: April 2, 2026

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Abstract

Systemic lupus erythematosus (SLE) is a multisystem autoimmune disorder with lesser known diverse gastrointestinal manifestations, occurring in up to 40% - 50% of patients. Intussusception, however, remains an exceedingly rare complication, with fewer than 15 cases reported worldwide, often linked to mesenteric vasculitis or serving as an initial presentation. We present two patient cases highlighting the spectrum of this rarity: one as the inaugural manifestation leading to SLE diagnosis in a young male, and another as a complication in a post-kidney transplant female on immunosuppression, precipitated by an underlying malignancy. These cases underscore diagnostic challenges, the role of imaging, and the need for prompt multidisciplinary intervention. Literature review reveals predominance in young females, with management ranging from conservative immunosuppression to surgical resection. Clinicians should maintain high suspicion for SLE in unexplained adult intussusception to mitigate life-threatening outcomes.

Keywords

Systemic Lupus Erythematosus, Intussusception, Lupus Enteritis, Immunosuppression, Mesenteric Vasculitis

1. Introduction

Systemic lupus erythematosus (SLE) is a chronic autoimmune disease characterized by diverse clinical manifestations, including gastrointestinal involvement in 40% - 50% of cases, typically presenting as abdominal pain, diarrhea, or vasculitis-

related complications like ischemia or perforation [1] [2]. Intussusception, a telescoping of bowel segments, is exceptionally uncommon in adults, accounting for only 5% of all intussusception cases [3], and even rarer in SLE, with literature documenting fewer than 15 instances globally [4]-[10]. These often arise from lupus mesenteric vasculitis (LMV), bowel edema, or altered peristalsis, and may present as the initial SLE symptom or a complication in immunocompromised states, such as post-transplant [11] [12]. Pathological lead points, defined as lesions that initiate the intussusception (e.g., tumors, polyps, or inflammatory masses), including malignancies, are more common in adults and may be exacerbated by chronic immunosuppression [3] [13].

This report describes two distinct presentations, emphasizing diagnostic pitfalls, therapeutic strategies, and the importance of multidisciplinary care in optimizing outcomes for this underrecognized entity. These cases contribute to the limited literature by illustrating diverse patient backgrounds; one in a treatment-naïve immigrant male and another in a post-transplant female with malignancy, highlighting the need for heightened awareness in varied populations to prevent delays in diagnosis and progression to complications like end-stage renal disease (ESRD).

2. Case Presentations

Case 1. A 28-year-old Spanish-speaking male from the Dominican Republic presented to the emergency department with acute intermittent epigastric and right upper quadrant abdominal pain for 2 days, rated 9/10 at worst, exacerbated by food intake, and associated with nausea and vomiting. He denied other acute symptoms and reported no alcohol, smoking, or drug use. Past medical history included unspecified kidney disease.

Vital signs showed blood pressure 150/100 mmHg, heart rate 100 beats/min, respiratory rate 20 breaths/min, and temperature 37.5°C. Abdominal exam revealed epigastric and right upper quadrant tenderness without rebound or guarding; bowel sounds were hypoactive.

Laboratories included hemoglobin 10.2 g/dL, neutrophils 82.3%, absolute lymphocytes $0.40 \times 10^9/L$, BUN 37 mg/dL, creatinine 2.58 mg/dL, and eGFR 37 mL/min/1.73m². Urinalysis showed 2+ blood, 3+ protein, and 6 - 10 RBCs/HPF. Abdominal CT demonstrated thickened, fluid-filled small bowel loops in the right lower quadrant with pneumatosis (**Figure 1**). Emergent exploratory laparotomy with extended right colectomy was performed due to CT findings of bowel wall thickening, fluid-filled loops, and pneumatosis intestinalis raising concern for ischemia or impending perforation. Pathology showed congested vascular channels and hypertrophied muscular fibers without granulomas or malignancy. He was discharged after 7 days, with new hypertension diagnosis and initiation of lisinopril and nifedipine.

The next day, he re-presented with hematochezia, loose stools, abdominal pain, tachycardia, and tachypnea. Hemoglobin was 5.9 g/dL, creatinine 2.27 mg/dL,



Figure 1. Axial CT abdomen demonstrating thickened loop of bowel within the right lower quadrant with swirling of the adjacent mesentery (see arrow) findings concerning for partial intussusception.

and eGFR 44 mL/min/1.73m². CT showed post-surgical changes without extravasation. He was transfused and discharged.

One week later, he returned with fatigue and bilateral leg weakness. Additional history revealed chronic malar rash, decreased appetite, hand/knee joint pains, fatigue, weight loss, and low-grade fevers. Laboratories showed hemoglobin 10.2 g/dL, BUN/creatinine 33/2.53 mg/dL, eGFR 38 mL/min/1.73m², urinalysis with 2+ protein and 1+ blood. Lower extremity Dopplers were negative. CT confirmed post-surgical changes.

Rheumatology evaluation established the diagnosis of SLE on the basis of clinical features (malar rash, polyarthralgias, constitutional symptoms including fatigue, weight loss, and fevers) and serologic abnormalities (ANA titer 1:1280, dsDNA titer 1:80, SSA > 240 AU/mL, SSB >320 AU/mL, CRP 7.93 mg/L; negative anti-Sm, anti-centromere, Scl-70, anti-CCP, anti-RNP, U1-RNP, and ANCA). These findings, together with biopsy-confirmed class IV/V lupus nephritis (proteinuria quantified at approximately 2 g/24 hours), fulfilled the 2019 EULAR/ACR classification criteria for SLE. Intussusception was suspected as a rare SLE presentation with lupus nephritis. Referrals were made to hematology/oncology for anemia and nephrology for biopsy.

Mycophenolate mofetil 1500 mg BID and hydroxychloroquine 200 mg daily were started, but mycophenolate was reduced to 1.5 g daily due to nausea and hypertension. Nephrology biopsy confirmed proliferative glomerulonephritis

with membranous features consistent with class IV/V lupus nephritis. He remains on hemodialysis for ESRD under multidisciplinary care.

Case 2. A 48-year-old female with a medical history significant for chronic anemia, antiphospholipid (APL) syndrome managed with warfarin, systemic lupus with lupus nephritis status post kidney transplant in 2010 on chronic mycophenolate mofetil and tacrolimus, history of small bowel obstruction (SBO) status post partial colectomy in 2014, cerebrovascular accident (CVA), and liver hemangioma presented to the emergency department on November 24 with a one-week history of nausea, vomiting, bloody diarrhea, and generalized weakness. She had experienced multiple prior admissions for recurrent abdominal pain and diarrhea, which had led to a recent adjustment in her immunosuppressive regimen from mycophenolate mofetil (CellCept) to azathioprine (AZA) suspecting medication-related cause; however, her symptoms persisted despite this change.

On physical examination, the abdomen was distended with tenderness, particularly in the right lower quadrant, but without rebound or guarding. Vital signs were stable, and laboratory studies revealed chronic anemia (hemoglobin 8.5 g/dL) but no acute leukocytosis or electrolyte derangements. Initial imaging with computed tomography (CT) of the abdomen and pelvis demonstrated a long segment small bowel intussusception measuring at least 13 cm in length, with significant dilation of the small bowel proximal to this, with fluid and air suggestive of mechanical obstruction (**Figure 2**). Surgical management with diagnostic laparoscopy and extracorporeal small bowel resection at two sites was pursued the following day due to these obstructive features and the need to evaluate for a pathologic lead point in this immunosuppressed adult patient. Intraoperative findings included no evidence of ischemia or perforation. Histopathological examination



Figure 2. Coronal CT abdomen and pelvis demonstrating a long segment small bowel intussusception measuring at least 13 cm in length, with significant dilation of the small bowel proximal to this, with fluid and air (see arrow).

of the resected specimens revealed a 5 mm adenocarcinoma arising within a tubular adenoma, with negative surgical margins and one incidental lymph node negative for malignancy. Postoperative workup for metastatic disease, including positron emission tomography (PET) scan and tumor markers, was negative.

Following surgery, the patient's gastrointestinal symptoms resolved completely. However, her renal allograft function subsequently declined, with rising creatinine levels and proteinuria, prompting evaluation for a second kidney transplant. She remains under close multidisciplinary follow-up with rheumatology for SLE management, oncology for surveillance of the adenocarcinoma, and the transplant service for ongoing assessment and optimization for transplantation.

3. Discussion

This report illustrates the heterogeneous presentation of intussusception in SLE, spanning from an initial diagnostic clue in a treatment-naïve patient (Case 1) to a malignancy-driven complication in a post-transplant, immunosuppressed individual (Case 2). Both cases represent rare gastrointestinal manifestations of SLE, highlighting the disease's protean nature. In Case 1, the patient presented with intussusception prior to any immunosuppressive therapy, arguing against a causal link to agents like mycophenolate mofetil (MMF) or other immunosuppressants (IS). Instead, this underscores that intussusception can arise directly from SLE-related mechanisms, such as lupus mesenteric vasculitis (LMV), even in the absence of treatment-related complications [4] [8] [9]. Gastrointestinal manifestations in SLE are multifaceted, ranging from mild mucosal inflammation to severe vasculitis, with lupus enteritis—a form of mesenteric vasculitis—being a key contributor to acute abdominal syndromes [1] [2]. Intussusception's rarity is underscored by literature reviews, with retrospective analyses identifying only 7 - 15 cases worldwide, predominantly in young females and involving ileocolic or small bowel segments [4]-[10]. Many cases, like Case 1, feature intussusception as SLE's inaugural manifestation, presenting with acute pain, vomiting, and diarrhea, often in diverse populations including Sudanese, Chinese, and now a Dominican immigrant [4] [5] [8]. A 2021 report detailed a 40-year-old Sudanese woman presenting with intussusception as the first sign of SLE, managed conservatively with high-dose corticosteroids after CT confirmation, emphasizing early serological testing in unexplained abdominal emergencies [4].

Pathophysiologically, SLE-associated intussusception is primarily attributed to LMV, causing vascular necrosis, bowel wall edema, and peristaltic disruption, potentially leading to telescoping segments [2] [12] [14]. Other mechanisms include lymphadenopathy, altered motility from edematous intestines, or secondary malignancies like Burkitt's lymphoma or, as in Case 2, adenocarcinoma [8] [9] [15]. In immunocompromised patients, such as post-transplant on agents like mycophenolate or azathioprine, the risk of neoplasm development escalates 1.8- to 2-fold due to impaired immune surveillance [3]. In Case 2, histopathological examination revealed a 5 mm adenocarcinoma arising within a tubular adenoma. The

occurrence took place in the setting of long-term post-transplant immunosuppression, which is a known risk factor for de novo malignancies; this may have contributed alongside other factors. A similar 2022 case involved a 38-year-old female with SLE and prior Roux-en-Y gastric bypass who developed alimentary loop intussusception, presenting with severe epigastric pain, hematemesis, and requiring multiple laparotomies for ischemia, perforation, and anastomotic leaks; pathology revealed atrophic villi and chronic inflammation, attributed to lupus enteritis exacerbating post-bariatric motility issues [6]. This highlights how comorbidities like bariatric surgery can intersect with SLE to precipitate intussusception, with reported incidence post-bypass at 0.64%, often retrograde and in the alimentary limb [6].

Recent literature further expands on this rarity. A 2023 case report described acute intestinal obstruction in systemic lupus erythematosus [10]. Abstracts from gastroenterology meetings [13] [16] have noted the scarcity of SLE-associated intussusception cases, with one reviewing ileocecal intussusception alongside prior literature. These updates align with earlier findings but emphasize demographic shifts, including more males and immigrants affected like our Case 1, potentially due to delayed diagnosis in underserved populations [4]-[10]. Prior cases have noted the demographic diversity affected by intussusception and stressed the need for early CT and serologic evaluation for potential autoimmune disease [4] [5] [8] [9].

Diagnostic challenges are evident, as symptoms overlap with medication side effects, infections, or pseudo-obstruction—a rarer SLE complication with about 30 reported cases [11] [17]. CT imaging is pivotal, revealing the "target sign," bowel-within-bowel configuration, or LMV features like the "comb sign" and ascites [2] [12]. In Case 1, concurrent musculoskeletal symptoms prompted serological workup, unveiling ANA positivity and lupus nephritis, while in Case 2, the CT findings prompted urgent surgery, uncovering malignancy. Recent guidelines recommend integrating abdominal CT imaging with lupus serology in high-risk patients to differentiate from mimics like ischemic bowel or infections [2].

Given the rarity of intussusception as a presenting feature of SLE, particularly in treatment-naïve individuals as in Case 1, clinicians should maintain a low threshold for ruling out SLE in young adults with unexplained intussusception. This is especially pertinent if accompanied by systemic symptoms such as arthralgias, fatigue, or mucocutaneous findings, warranting prompt ordering of serological tests including ANA, anti-dsDNA, and complement levels [2] [4] [5] [8]. Early recognition can prevent diagnostic delays and mitigate progression to severe complications like ESRD.

Management varies by etiology and severity. Vasculitis-driven cases without obstruction may resolve with high-dose corticosteroids (e.g., prednisone equivalent of 1 mg/kg/day or intravenous methylprednisolone pulses of 500 - 1000 mg daily for 3 days, as reported in earlier cases [4] [8]), intravenous immunoglobulin (IVIG), or pulse methylprednisolone, as seen in a pediatric case and retrospective

series of five patients [7] [8]. Persistent symptoms or necrosis necessitate laparoscopy/laparotomy with resection, as in both our cases, to avert perforation or sepsis [4] [6]. Post-transplant considerations include balancing immunosuppression to prevent rejection while monitoring for malignancies [3]. A recent review advocates for tailored regimens, such as switching from mycophenolate to azathioprine in refractory GI symptoms but warns of persistent risks of infection or malignancy in comorbid patients [16]. Immigrant status and limited healthcare access underscore the need for heightened suspicion in young adults with unexplained intussusception to halt progression to ESRD. Emerging studies suggest exploring biologic therapies targeting vascular inflammation to reduce intussusception incidence in high-risk SLE subsets, potentially including anti-type I interferon agents or B-cell depletion to downregulate vasculitis pathways [10] [16].

4. Limitations

The causal relationship between SLE and intussusception cannot be definitively proven in these cases, particularly given the absence of histopathological vasculitis in Case 1. This report involves only two patients and relies on a narrative (not systematic) literature review, which is subject to selection and publication bias. Larger prospective studies are required to better characterize incidence, mechanisms, and optimal management.

5. Conclusion

Intussusception represents a rare, potentially catastrophic SLE complication, manifesting across disease stages and warranting early advanced imaging in patients with refractory abdominal symptoms. These cases and literature review highlight the need for multidisciplinary collaboration among rheumatology, surgery, oncology, and transplant services to achieve favorable outcomes. Enhanced awareness and further mechanistic studies are essential to improve preventive and therapeutic approaches in SLE-associated gastrointestinal pathologies. Future research into preventive strategies, such as biomarkers for early vasculitis detection, and the role of routine GI surveillance with advanced imaging, could reduce these rare but life-threatening events.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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