

An Adolescent with Undiagnosed Ulcerative Colitis Presenting with Toxic Megacolon and Cavitory Pulmonary Nodules

RAHIYA REHMAN, MD; STEPHANIE CAMHI, MD; JASON SHAPIRO, MD; SHOVA SUBEDI, MD; LINDA SHALON, MD

ABSTRACT

Toxic megacolon and pulmonary nodules are not seen frequently on diagnosis in pediatric ulcerative colitis patients. This report emphasizes the importance of carefully evaluating and managing complications in pediatric ulcerative colitis cases, especially in the presence of pulmonary nodules.

KEYWORDS: pediatrics, ulcerative colitis, inflammatory bowel disease, lung nodules

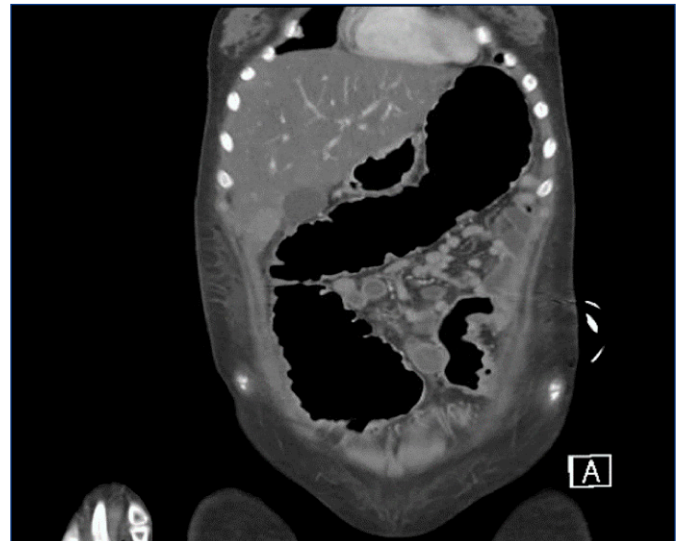
INTRODUCTION

Toxic megacolon (TM) is an infrequent complication of ulcerative colitis (UC), especially in children, with a reported incidence of only 1–2%.¹ TM is characterized by colonic dilation (≥ 5 –6 cm), along with signs of system toxicity. TM is most often associated with infectious colitides, such as *Clostridioides difficile* (*C. difficile*) and cytomegalovirus.^{1,2} It can lead to mortality if left untreated. Inflammatory bowel disease (IBD) can also lead to several extra intestinal manifestations including rare occurrence of cavitory pulmonary nodules (CPN) which can be a diagnostic dilemma due to their resemblance to other infectious conditions.^{3,4} We present a unique case of a pediatric patient who initially presented with two rare complications of ulcerative colitis (UC): TM and CPN.

CASE PRESENTATION

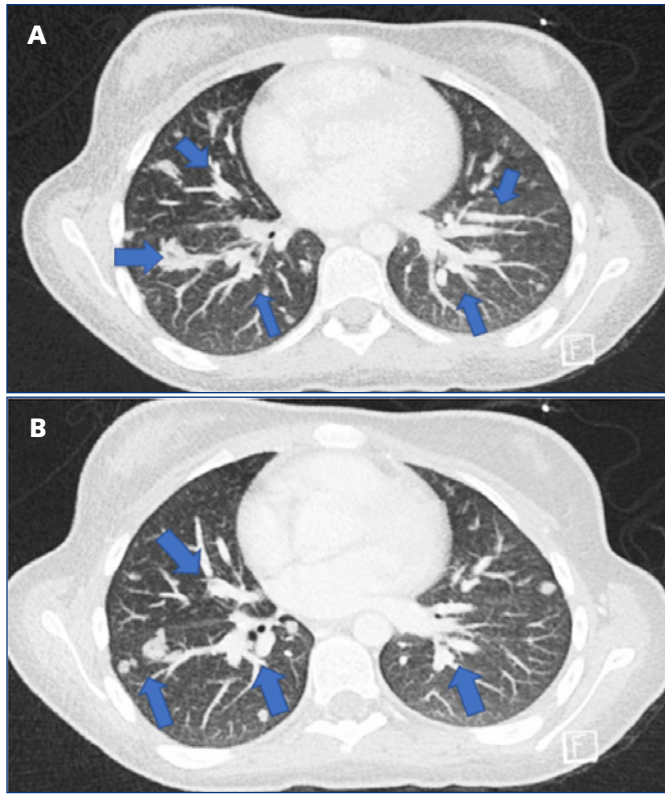
A 12-year-old girl came to the emergency department with a four-week history of abdominal pain, poor appetite, diarrhea, and weight loss. She had an unremarkable exam except for persistent tachycardia. Labs showed microcytic anemia, leukocytosis with bandemia, elevated erythrocyte sedimentation rate, and hypoalbuminemia. Tachycardia initially responded to fluid resuscitation; however, next day she developed abdominal tenderness and bloody diarrhea. Abdominal X-ray showed dilated transverse colon. Chest X-ray revealed right-sided atelectasis vs pneumonia. Magnetic resonance enterography (MRE) showed diffuse colonic distention (~6–8 cm in diameter) with circumferential thickening and hyperenhancement of the colon without terminal ileum involvement (**Figure 1**). She received antibiotics and red blood cell

Figure 1. MRE of the abdomen showed diffuse distention of the colon along with air-fluid levels, marrow edema, circumferential wall thickening and hyperenhancement on postcontrast images.



transfusions. Stool infectious studies including *Clostridium difficile* were negative and high dose steroids were started for presumed inflammatory bowel disease (IBD). Further review of the MRE demonstrated basal lung nodules. Follow-up chest computed tomography (CT) revealed bilateral pulmonary nodules with a basilar and peripheral predominance, and cavitation (**Figure 2**) suggestive of either septic emboli or inflammatory nodules seen in IBD. Tuberculosis infection was ruled out with QuantiFERON gold testing. Her colitis was unresponsive to high-dose steroids, and therapy with anti-tumor necrosis factor (anti-TNF) agent was initiated given patient's hesitancy for any surgical intervention. However, she developed a bowel perforation resulting in subtotal colectomy and ileostomy. Distal sigmoid colon and rectum appeared healthy and were left in place at that time given unclear diagnosis while the remaining inflamed colon was removed. Histopathology was consistent with UC, further confirming diagnosis of UC. After four weeks she was discharged home with plans for future completion proctocolectomy, creation of J pouch and eventual ileostomy closure following nutritional rehabilitation in future. A repeat chest X-ray and CT scan at follow-up visit demonstrated almost complete resolution of the pulmonary nodules.

Figure 2A,2B. CT scan of chest with IV contrast showed innumerable pulmonary nodules noted throughout the lungs with a basilar and peripheral predominance along with some cavitation.



DISCUSSION

Our case highlights two rare complications of ulcerative colitis in an adolescent: acute severe ulcerative colitis presenting as toxic megacolon and pulmonary nodules. Acute severe UC is managed initially with high-dose steroids, but this decision must be carefully weighed in a patient with lung nodules of uncertain etiology. Parenchymal lung disease and lung nodules seen in UC can mimic autoimmune processes such as granulomatosis with polyangiitis, sarcoidosis and vasculitis, and are commonly confused with septic pulmonary emboli.^{5,6} Both septic emboli and necrobiotic nodules may appear as cavitating lesions on imaging. Septic emboli are suggested by bilateral peripheral nodules with identifiable feeding vessels and may have accompanying lung abscesses with a predominant neutrophilic infiltrate on histopathology.⁷ Inflammatory pulmonary nodules are also bilateral but have a necrotic granulomatous infiltrate on histopathology.^{5,6} While septic emboli always respond to antimicrobial therapy, they usually get worse with immunosuppressive therapy. Inflammatory nodules on the other hand do not respond to antibiotics but show a favorable response to steroids and anti-TNF therapy.⁶⁻⁸ Lung biopsy was not recommended in our patient given the acuity of her illness. She likely had inflammatory nodules as suggested by the lack of documented infection and near complete resolution of lung nodules on follow-up imaging after treatment of her ulcerative colitis.

To the best of our knowledge, this is the first report of a pediatric patient presenting with both TM and CPN as initial presentation of UC. This case highlights the dilemma of treating acute severe colitis in the presence of CPN.

References

1. Russell RK, Protheroe A, Roughton M, Croft NM, Murphy MS, Spray C, et al. Contemporary outcomes for ulcerative colitis inpatients admitted to pediatric hospitals in the United Kingdom. *Inflamm Bowel Dis.* 2013;19: 1434–1440. PMID: 23624885
2. Benchimol EI, Turner D, Mann EH, Thomas KE, Gomes T, McLennan RA, et al. Toxic megacolon in children with inflammatory bowel disease: clinical and radiographic characteristics. *Am J Gastroenterol.* 2008;103: 1524–1531. PMID: 18510624
3. Barfield E, Deshmukh F, Slighton E, Lentine J, Lu Y, Ma X, et al. Pulmonary Manifestations in Adolescents with Inflammatory Bowel Disease. *Clin Pediatr.* 2020;59: 573–579. PMID: 32146830
4. Pemmasani G, Loftus EV, Tremaine WJ. Prevalence of Pulmonary Diseases in Association with Inflammatory Bowel Disease. *Dig Dis Sci.* 2022. PMID: 35142913
5. Turner D, Ruemmele FM, Orlanski-Meyer E, Griffiths AM, de Carpi JM, Bronsky J, et al. Management of Paediatric Ulcerative Colitis, Part 2: Acute Severe Colitis-An Evidence-based Consensus Guideline from the European Crohn's and Colitis Organization and the European Society of Paediatric Gastroenterology, Hepatology and Nutrition. *J Pediatr Gastroenterol Nutr.* 2018;67: 292–310. PMID: 30044358
6. Mukherjee S, Harne P, Mirchia K, Sharma A, Manocha D. Necrobiotic Pulmonary Nodules in Ulcerative Colitis: Not Just a “Crohnic” Phenomenon. *ACG Case Rep J.* 2020;7: e00438. PMID: 32903944
7. Weerakkody Y, Murphy A, Lukies M, et al. Septic pulmonary emboli. Reference article, Radiopaedia.org (Accessed on 23 Jul 2023) <https://doi.org/10.53347/rID-22848>
8. Kellish A, Soal V, Caskey E, Hassinger G, Terrigno N. Pulmonary Necrobiotic Nodules at Time of Diagnosis in a Patient with Ulcerative Colitis. *Cureus.* 2020;12: e7474. PMID: 32351852

Authors

Rahiya Rehman, MD, Brown Pediatric Gastroenterology Fellow, Division of Pediatric Gastroenterology, Hepatology and Nutrition, Hasbro Children's Hospital, and Department of Pediatrics, The Warren Alpert Medical School of Brown University, Providence, RI.

Stephanie Camhi, MD, Brown Pediatric Resident, Department of Pediatrics, The Warren Alpert Medical School of Brown University, Providence, RI.

Jason Shapiro, MD, Attending Physician, Division of Pediatric Gastroenterology, Hepatology and Nutrition, Hasbro Children's Hospital, and Department of Pediatrics, The Warren Alpert Medical School of Brown University, Providence, RI.

Shova Subedi, MD, Attending Physician, Division of Pediatric Gastroenterology, Hepatology and Nutrition, Hasbro Children's Hospital, and Department of Pediatrics, The Warren Alpert Medical School of Brown University, Providence, RI.

Linda Shalon, MD, Attending Physician, Division of Pediatric Gastroenterology, Hepatology and Nutrition, Hasbro Children's Hospital, and Department of Pediatrics, The Warren Alpert Medical School of Brown University, Providence, RI.

Disclosures

There are no conflict of interest or financial disclosures to report.

Correspondence

Rahiya Rehman, MD
Division of Pediatric Gastroenterology, Hepatology and Nutrition
Hasbro Children's Hospital, 593 Eddy St, Providence RI 02903
401-444-2851
Rahiya_rehman@brown.edu