

Diffuse Panbronchiolitis in a Patient With Ulcerative Colitis Treated With Ustekinumab

Meghan Marmor, MD¹, Gerald Berry, MD², and Rishi Raj, MD¹

¹Division of Pulmonary, Allergy and Critical Care Medicine, Stanford University School of Medicine, Stanford, CA

²Department of Pathology, Stanford University School of Medicine, Stanford, CA

CASE REPORT

A 51-year-old White man with no known medical history was diagnosed with ulcerative colitis (UC) based on moderate active colitis found on colon and rectal biopsies. His hematochezia was controlled with 1 week of systemic corticosteroids. Sulfasalazine was an effective maintenance therapy for his colitis, and he otherwise had no evidence of extraintestinal disease. Three months later, he presented with progressive shortness of breath. A chest computed tomography scan revealed diffuse panbronchiolitis (Figure 1), a rare extraintestinal manifestation of UC. The diagnosis was confirmed by surgical wedge lung biopsy demonstrating mononuclear bronchiolitis and the presence of foamy macrophages within the walls and lumens of the respiratory bronchioles (Figure 2). His disease was responsive to steroids, but recurred despite the use of azathioprine and mycophenolate as steroid-sparing agents. He received ustekinumab, a novel anti-interleukin-12 and interleukin-23 biologic agent, which was chosen to respect the patient's preference for subcutaneous administration. Within a month, he weaned from corticosteroids, his dyspnea resolved, and repeat chest computed tomography showed resolution of his bronchiolitis (Figure 3). His clinical and radiographic resolution persisted at 1-year follow-up where he continued on ustekinumab therapy. Clinically recognized UC-associated airway disease is rare, although bronchial involvement accounts for approximately half of UC-related pulmonary disease.¹⁻³ The pathogenesis is unknown. Diffuse panbronchiolitis is an especially rare form of UC-related pulmonary disease with few reported cases.⁴ This is the first case reported in the literature suggesting the efficacy of ustekinumab for the treatment of pulmonary manifestations of inflammatory bowel disease.

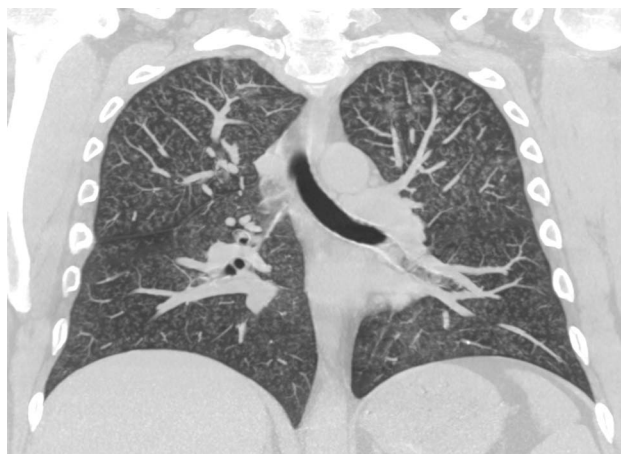


Figure 1. Coronal chest computed tomography images showing diffuse, innumerable centrilobular nodules throughout the lung fields.

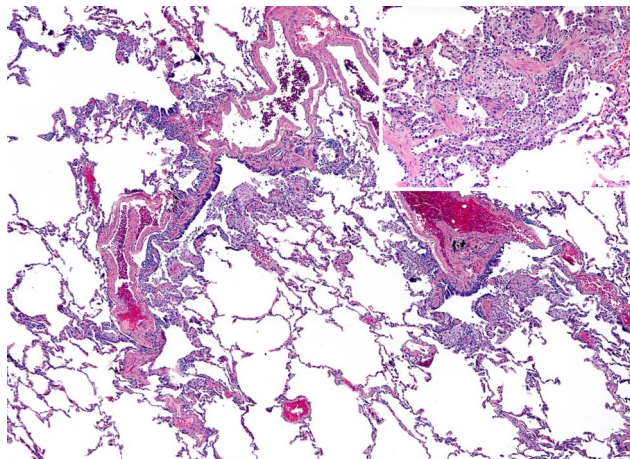


Figure 2. The left lower lobe surgical lung biopsy shows preservation of the pulmonary architecture with bronchiocentric inflammation of respiratory bronchioles and preservation of adjacent alveolar structures (hematoxylin and eosin 40×). Insert: Respiratory bronchiole with interstitial and luminal accumulations of foamy macrophages and bronchiolitis (hematoxylin and eosin 100×).

DISCLOSURES

Author contributions: M. Marmor cared directly for the patient and chose the treatments for his condition. G. Berry provided histologic interpretation and histology images. R. Raj provided diagnostic guidance. M. Marmor is the article guarantor.

Financial disclosure: None to report.

Previous presentation: This case report will be presented at the American Thoracic Society International Meeting; May 2023; Washington, DC.

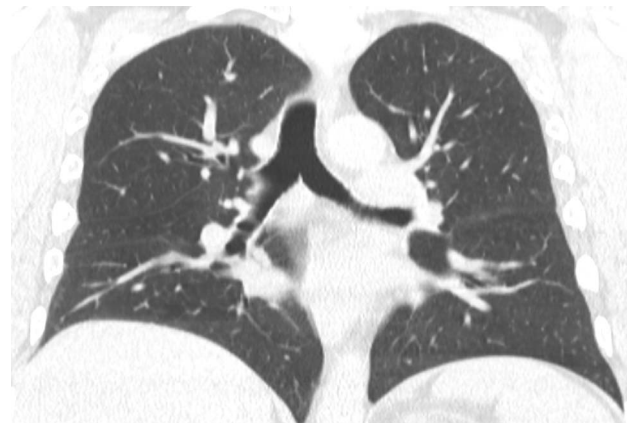


Figure 3. Coronal chest computed tomography images showing resolution of centrilobular nodularity.

Informed consent was obtained for this case report.

Received January 27, 2023; Accepted April 28, 2023

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