Radiology Case Reports 12 (2017) 686–689

Available online at www.sciencedirect.com

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journal homepage: http://Elsevier.com/locate/radcr

Chest

# Delayed recurrence of ulcerative colitis manifested by tracheobronchitis, bronchiolitis, and bronchiolectasis

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ARTICLE INFO

*Article history:*
Received 17 April 2017
Received in revised form 18 July 2017
Accepted 8 August 2017
Available online

*Keywords:*
Ulcerative colitis
Trachea
Lung

ABSTRACT

Ulcerative colitis can cause inflammation of small and large airways, characterized by mucosal inflammation, tracheobronchial stenosis, bronchiestasis, and bronchiolitis. We present a case of tracheobronchitis and bronchiolitis associated with ulcerative colitis in a 58-year-old non-smoking man, 17 years after the total colectomy and complete resolution of intestinal findings. Computed tomography demonstrated wall thickening of trachea and left main stem bronchus, and multiple bronchi around the both hilum with mild to moderate stenosis. Fiberoptic bronchial biopsy showed inflammation of the airways, similar to histologic findings of ulcerative colitis within colon.



## Introduction

Ulcerative colitis (UC) is characterized by recurring episodes of inflammation limited to the mucosal layer of the colon. It commonly involves the rectum and may extend in a proximal and continuous fashion to involve other parts of the colon. UC is associated with a variety of extraintestinal manifestations. Although pulmonary manifestation is relatively rare, UC has been associated with upper airway stenosis, tracheobronchitis, bronchiectasis, constrictive bronchiolitis, panbronchiolitis, necrobiotic nodules, lung bullae, interstitial lung disease, organizing pneumonia, sarcoidosis, pulmonary vasculitis, pulmonary eosinophilia, Wegener granulomatosis without renal involvement, and apical fibrosis [1]. These abnormalities are generally related to the underlying bowel disease. Pulmonary parenchymal disease is seen more commonly with UC than with Crohn disease, and large airway disease is strongly associated with UC [2].

In this case report, we are presenting a patient with UC with diffuse tracheal and bronchial involvement, bronchiectasis, and bronchiolectasis. We will review airway complications of UC.

## Case report

The patient is a 58-year-old male never smoker, diagnosed with UC, in 1977. He had colectomy in 1999. He was surgically cured of his UC, and currently has no signs or symptoms of gastrointestinal disease. He had been off Azulfidine and prednisone

Competing Interests: The authors have no conflicts of interest.

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https://doi.org/10.1016/j.radcr.2017.08.003

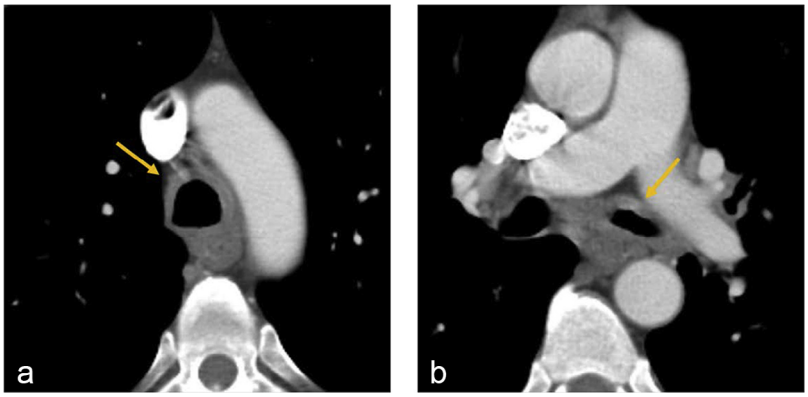


**Fig. 1 – Contrast-enhanced axial CT images of the chest at the level of the (A) mid-trachea and (B) left main bronchus (mediastinal window setting). There is circumferential thickening of the trachea that extends to involve the left mainstem bronchus (arrows).**

since 1999 after colectomy, previously treated with them for at least 15 years. He is referred to pulmonologist for evaluation of cough. Onset of cough occurred approximately 5 years ago and progressively worsened. Chest radiography showed left lower lobe infiltrate. Afterward, contrast-enhanced chest computed tomography (CT) examination was ordered for better characterization of clinical symptoms and chest X-ray findings. Contrast-enhanced CT showed circumferential, diffuse thickening of the tracheal wall that extends to involve the left main bronchus (Fig. 1). There was diffuse wall thickening of right upper, middle, left lingular, and lower lobe bronchi around both hila (Fig. 2). Extensive bronchiectasis, bronchiolectasis, and mucus plugging were seen in the left lower lobe (Fig. 3).

Pulmonologist performed bronchoscopy for further evaluation of extensive large airway disease.

Bronchoscopy demonstrated diffuse, severe inflammation with erythema, bleeding, sloughing mucosa, and edema from trachea to left lower lobe bronchus. It was difficult to identify tracheal and bronchial cartilaginous rings caused by edema or sloughing mucosa.

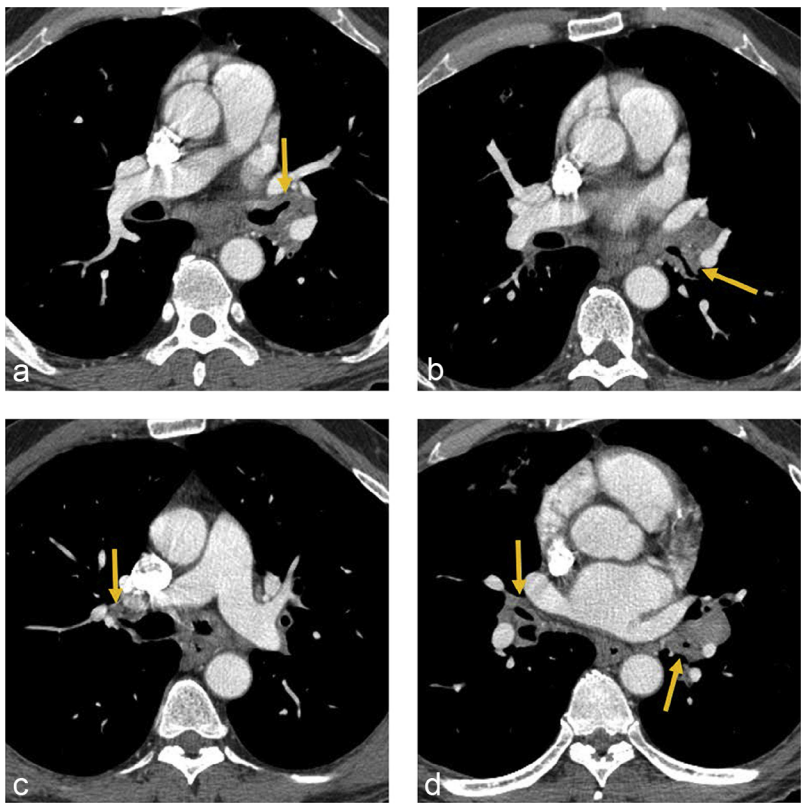


**Fig. 2 – Contrast-enhanced axial CT images of the chest at the level of the left lingular and left lower lobe superior segmental bronchus (A, B) show diffuse wall thickening within these airways. (C, D) Diffuse wall thickening within the right upper lobe, right middle lobe, and left lower lobe bronchus.**

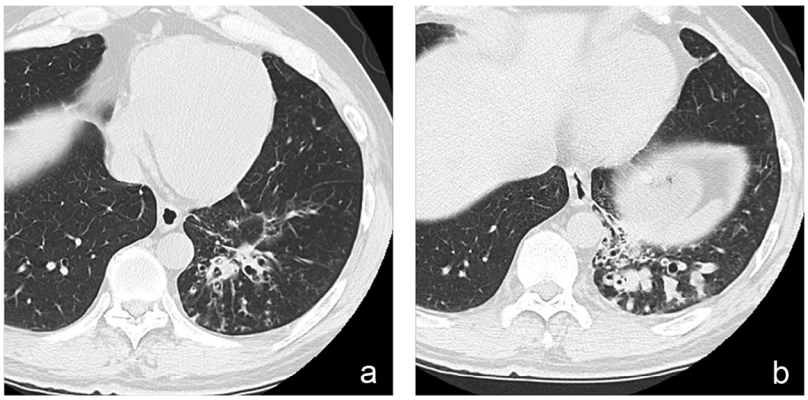


**Fig. 3 – Axial reformatted CT images (lung window setting) show bronchiectasis, bronchiolectasis, bronchial wall thickening, and mucus plugging within the left lower lobe.**

Bronchial wall biopsy histologic sections demonstrated acute inflammation associated with prominent lymphoplasmacytic inflammation, ulceration, and hemorrhage. Extent and severity of airway process suggested UC-related large airway disease refractory to colectomy.

Prednisone 20 mg/d was started. He returned 6 months later on prednisone 20 mg/d, his dry cough and mucus production have nearly resolved. Chest examination was normal. His resting SaO2 was improved from 96% to 98% on room air at the end of the 6-month treatment. Ronchi has resolved on chest auscultation. Repeat chest CT showed near complete resolution of wall thickening within trachea, left main stem bronchus, left lingular, left lower lobe, and right middle lobe bronchi (Fig. 4). Mucus plugging and airway wall thickening have also resolved. There was persistent bronchiectasis and bronchiolectasis within the left lower lobe.

## Discussion

Respiratory involvement in patients with IBD has been reported since 1976. The airways and alimentary tract have a common origin from the primitive gut, and both have a columnar epithelium that undergoes nonspecific inflammatory changes during the disease process. Various hypotheses have also been proposed in support of common pathogenic mechanisms. One interesting hypothesis suggests that common antigens, both inhaled and ingested, sensitize the lymphoid tissue of the lung and intestine, and trigger an allergic inflammatory response [3].

Patients with UC may present with cough, dyspnea, chest pain, or an abnormal chest radiograph obtained for another reason. Chest radiographs are frequently normal or show nonspecific changes resulting from bronchial wall thickening or bronchiectasis. High-resolution chest CT is more sensitive than conventional chest radiographs, often demonstrating findings of bronchial wall thickening, bronchiectasis, or tree-in-bud opacities, wall thickening within trachea, and main bronchi [4,5].

Pulmonary complications of UC is associated with inflammation of the trachea, bronchi, and bronchioles [6–8]. Large airways are the most common location of UC involvement in literature [9]. Tracheitis can develop at any time during the disease course and usually follow the onset of bowel disease, occurring in the fifth decade of life. It is more common in female patients with UC [10]. Tracheitis can also be the initial presentation of UC [11,12]. Edema, ulceration, and hemorrhage within the trachea and large airways have histologic appearance similar

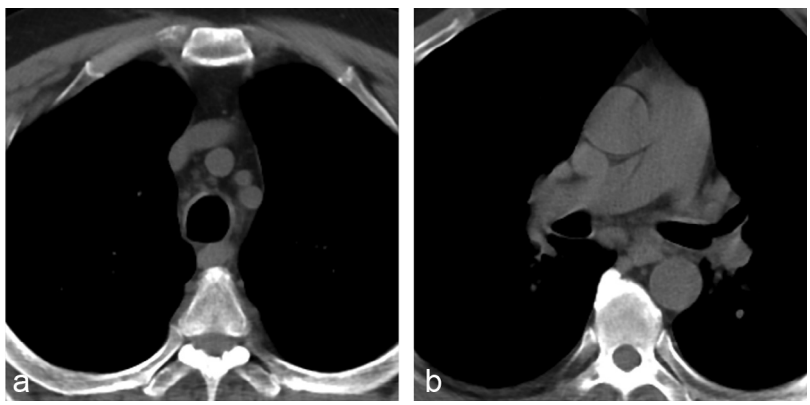


**Fig. 4 – Axial noncontrast CT images 6 months after steroid therapy (A, B) show complete resolution of wall thickening within the trachea and left mainstem bronchus.**

to the colonic epithelium of patients with UC. Tracheal inflammation, if not treated, may cause an irreversible destruction in trachea, leading to tracheal stenosis [7]. Severe stenosis because of edema and ulceration can compromise the airway and require urgent intervention [13]. In our case, there was diffuse wall thickening of trachea and left main stem bronchus, including posterior membranous wall of the trachea and bronchi.

Bronchiectasis is the most common disease of the upper airways, noted in 66% of patients with IBD involving the large airways [10]. Acute and chronic bronchitis, and suppurative large airway disease without bronchiectasis are other manifestations of IBD within lung bronchitis [10]. Chest CT findings are bronchiectasis and bronchial wall thickening with or without mucoid impaction [2]. In our case, bronchiectasis and bronchial wall thickening were moderate within the left lower lobe.

Bronchiolitis, or small airway involvement, is a less common pulmonary manifestation of UC. CT findings of bronchiolitis are tree-in-bud–type opacities, mosaic attenuation, and centrilobular ground glass nodules. We observed scattered tree-in-bud–type opacities within the left lower lobe in our case.

Other pulmonary complications of IBD include cryptogenic organizing pneumonia, usual interstitial pneumonia, eosinophilic pneumonia, Langerhans granulomatosis, serositis, and pulmonary embolism [1,3,14]. However, the most common pulmonary manifestations of IBD are drug-induced lung disease and infections because of immunosuppressive medication, used for treatment of bowel disease. Sulfasalazine and mesalamine are associated with eosinophilic pneumonia, pulmonary fibrosis, and pleural effusion [14].

## Conclusion

In summary, tracheobronchitis and bronchiolitis are rare pulmonary complications of UC that should be considered in patients with UC with chronic productive cough, even after total colectomy. For these patients, workup with chest CT would be warranted to detect tracheobronchitis and bronchiolitis. Bronchoscopy and biopsy are required for final diagnosis. Treatment is usually initiated with corticosteroids and may result in complete control of symptoms as well as significant reversal in pulmonary findings. It is very important to maintain a high index of suspicion for pulmonary complications of UC to initiate early appropriate treatment and avoid complications.

## REFERENCES


[1] Mahadeva R, Walsh G, Flower CD, Shneerson JM. Clinical and radiological characteristics of lung disease in inflammatory bowel disease. Eur Respir J 2000;15(1):41–8.

[2] Betancourt SL, Palacio D, Jimenez CA, Martinez S, Marom EM. Thoracic manifestations of inflammatory bowel disease. AJR Am J Roentgenol 2011;197(3):W452–6.

[3] Sostegni R, Daperno M, Pera A. Pulmonary manifestations of inflammatory bowel disease. Dig Liver Dis 2007;39(3):239–41.

[4] Garg K, Lynch DA, Newell JD. Inflammatory airways disease in ulcerative colitis: CT and high-resolution CT features. J Thorac Imaging 1993;8(2):159–63.

[5] Spira A, Grossman R, Balter M. Large airway disease associated with inflammatory bowel disease. Chest 1998;113(6):1723–6.

[6] Kuzniar T, Sleiman C, Brugière O, Fournier M. Severe tracheobronchial stenosis in a patient with Crohn's disease. Eur Respir J 2000;15(1):209–12.

[7] Bayraktaroglu S, Basoglu O, Ceylan N, Aydın A, Tuncel S, Savas R. A rare extraintestinal manifestation of ulcerative colitis: tracheobronchitis associated with ulcerative colitis. J Crohns Colitis 2010;4(6):679–82.

[8] Camus P, Piard F, Ashcroft T, Gal AA, Colby TV. The lung in inflammatory bowel disease. Medicine (Baltimore) 1993;72(3):151–83.

[9] Vutcovici M, Brassard P, Bitton A. Inflammatory bowel disease and airway diseases. World J Gastroenterol 2016;22(34):7735–41.

[10] Black H, Mendoza M, Murin S. Thoracic manifestations of inflammatory bowel disease. Chest 2007;131(2):524–32.

[11] Schleiermacher D, Hoffmann JC. Pulmonary abnormalities in inflammatory bowel disease. J Crohns Colitis 2007;1(2):61–9.

[12] Kar S, Thomas SG. A case of tracheobronchitis in ulcerative colitis: a review of literature. Clin Respir J 2009;3(1):51–4.

[13] Yamamoto AK, Babar JL. Case 184: ulcerative tracheobronchitis. Radiology 2012;264(2):609–13.

[14] Basseri B, Enayati P, Marchevsky A, Papadakis KA. Pulmonary manifestations of inflammatory bowel disease: case presentations and review. J Crohns Colitis 2010;4(4):390–7.