CASE REPORT

Tracheobronchitis as an extraintestinal manifestation of ulcerative colitis

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SUMMARY

Respiratory involvement is a rare extraintestinal manifestation of ulcerative colitis (UC). It commonly presents as bronchiectasis and rarely as tracheobronchitis. It can occur before or after the presentation of gastrointestinal symptoms. Only rarely does it occur after the patient undergoes colectomy. Diagnosis should be considered in any patient with UC who presents with unexplained upper respiratory symptoms and a negative work up for infectious aetiologies. It responds well to immunosuppressive therapy. We present a case of a 21-year-old woman who underwent colectomy for ulcerative colitis and later presented with new onset severe reversible inflammation of the upper respiratory tract.

BACKGROUND

Respiratory manifestations of ulcerative colitis (UC), although rare, may extend from the bronchiectasis to bronchiolitis and may involve any part of the respiratory system such as the pleura, the parenchyma or the airway. These manifestations can occur before or after the presentation of gastrointestinal (GI) symptoms. It can also occur after the patient undergoes colectomy for UC. We present a case of a 21-year-old woman who underwent colectomy for ulcerative colitis and later presented with new onset severe reversible inflammation of the upper respiratory tract.

CASE PRESENTATION

A 21-year-old woman with a history of UC status postcolectomy presented to the emergency room with cough along with greenish sputum of 3 weeks duration. She was given multiple empiric broad-spectrum antibiotic treatments but her symptoms failed to improve.

INVESTIGATIONS

The patient's sputum and endobronchial lavage cultures did not grow any bacteria, fungi or viruses. Four months prior to the index hospital admission she was diagnosed with UC with the help of colonoscopy with biopsy along with inflammatory bowel disease (IBD) serology, both of which were consistent with UC. The relevant serology tests that supported the diagnosis included: elevated IBD p-ANCA auto antibody elevated, IBD p-ANCA perinuclear pattern, IBD p-ANCA DNase sensitivity, ATG16L1 homozygous G/G and ECM1 homozygous T/T. She subsequently underwent a bronchoscopy with a biopsy that showed inflamed and cobblestoned tracheobronchial mucosa (figure 1).

Figure 1 Bronchoscopy showing inflamed and cobblestoned mucosa.

The bronchial biopsy showed acute inflammation with neutrophils (figure 2). Pulmonary function tests (PFT) demonstrated an obstructive pattern with no significant response to bronchodilator therapy. A diagnosis of UC with airway involvement was then made.

TREATMENT

The patient was subsequently treated with high-dose intravenous methylprednisolone.

OUTCOME AND FOLLOW-UP

The patient's symptoms and lung auscultation findings showed significant response and resolved completely within a few days.

Figure 2 Bronchial biopsy showing acute inflammation with neutrophils.

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DISCUSSION

Extraintestinal findings of IBD are well documented.¹ Involvement of the respiratory system in the literature, although reported, is rare.² Standard textbooks for IBD do not commonly discuss direct pulmonary involvement as an extraintestinal manifestation of IBD.³ Respiratory and GI systems possess common embryonic origin from the primitive foregut and have goblet and submucosal cells as a part of the luminal structure. This could be the possible physiological basis for pulmonary involvement in IBD.⁴ Respiratory involvement in UC tends to occur in the fifth decade of life, although there is a wide range for ages of onset. In 85% of the cases, most symptoms develop following the onset of disease from intestinal manifestations. In about 10–15% cases, respiratory symptoms predate the IBD. The respiratory symptoms may also occur after proctocolectomy.^{2 5} Bronchiectasis is the most common pulmonary manifestation of IBD noted in 66% cases involving airway disease in IBD. Other less common pathologies of the large airways include chronic bronchitis, bronchiolitis, tracheitis, tracheobronchitis and bronchiolitis obliterans organising pneumonia.⁴

Tracheal involvement in UC is rare and has only been mentioned in the literature as case reports few and far between.^{6–10} Tracheobronchitis can either be the initial presentation of ulcerative colitis or can develop during the disease course.^{6 7} It usually follows the onset of bowel disease.⁸ It has also been reported uncommonly to occur right after colectomy.² Rarely, it has also been noted that people may present with tracheobronchitis years after colectomy.⁹ In our case, the patient had undergone colectomy 1 month prior to the onset of respiratory symptoms.

Lung infections should be considered in the differential diagnosis of pulmonary parenchymal disease in patients with IBD, particularly when patients present with fever, dyspnoea and cough. For patients of IBD taking medication that has been associated with pulmonary toxicity, the implicated drug is empirically discontinued while awaiting culture results and a response to drug discontinuation.^{11 12} In our case, infectious causes of the tracheobronchitis were ruled out with the negative sputum, endobronchial and bronchoalveolar lavage cultures and negative viral serology. Drug-induced (allergic) lung injury was an unlikely differential for the patient's tracheobronchitis as she was not on any biological agents, immunomodulators or 5-ASA agents at the time of presentation. She was taken off biological agents (antitumour necrosis factor- α) after her colectomy. She was prescribed mesalamine suppositories for rectal bleeding from proctitis, but had discontinued the medication due to inconvenience of administration and lack of symptom improvement. She was not given any antibiotic until she presented with bronchitis-like symptoms, at which point she was prescribed azithromycin, which failed to improve her symptoms. She was subsequently started on levofloxacin due to treatment failure with azithromycin. This was followed by hospitalisation and treatment with piperacillin and tazobactam, which failed to improve her symptoms. She was subsequently treated with methylprednisolone, which improved her symptoms completely.

Our patient was also a non-smoker without any previous lung disease. The results of the biopsy, consistent with inflammatory findings similar to those of ulcerative confirm the diagnosis. The PFT patterns in our case are consistent with the obstructive

pattern shown in the previous reported cases of respiratory involvement of UC. Unlike asthma, there is no reversibility with bronchodilators.⁴

Our patient was treated with intravenous steroids followed by oral and then inhaled steroids. She showed prompt improvement of symptoms. Steroids are the most frequently reported treatment for pulmonary disease associated with IBD, but controlled trials confirming their efficacy are lacking possibly because of rarity of pulmonary involvement in IBD.⁴ It is important to rule out any underlying infectious pathology before starting steroid therapy. In cases of mild involvement inhaled steroids may be used. More aggressive airway inflammation will require nebulised steroids in conjunction with oral steroids. The dosage and duration of steroids is guided by symptomatic response.^{2 5} In refractory cases, regardless of the reason, a serial bronchial lavage via a fiberoptic bronchoscope has been proposed to achieve higher steroid doses locally.⁵ There was one case report where the patient required dilation of the trachea and eventually died due to tracheal rupture.¹⁰

Learning points

- ▶ Respiratory involvement is an uncommon extraintestinal manifestation of ulcerative colitis (UC).
- ▶ A high index of suspicion for pulmonary involvement should be kept in all patients with UC who present with pulmonary symptoms.
- ▶ Respiratory involvement responds well to immunosuppression.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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