Tracheobronchitis in a Patient with Crohn’s Disease

Takashi Asami1, Shinichiro Koyama2, Yasutaka Watanabe2, Chihiro Miwa2, Shinya Ushimaru1, Yoshiyuki Nakashima3 and Mitsuhiro Nokubi4

Abstract

A 19-year-old Japanese man who had been diagnosed with Crohn’s disease (CD) suffered from dry cough and fever over 38°C for three days prior to hospitalization. On admission, his colonic CD condition was stable, neither active nor in remission. Computed tomography of the chest showed small elevated tracheobronchial lesions. Bronchoscopy showed diffuse whitish granular lesions in the trachea and bronchi. The pathological findings in the biopsy showed inflammatory infiltration suggesting Crohn’s tracheobronchitis. Thereafter, he was treated with inhaled Fluticasone propionate 400 μg/day. After one week his dry cough improved, and after two weeks bronchoscopic findings were improved.

Key words: Crohn’s disease, tracheobronchitis, inhaled corticosteroid therapy

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Introduction

The inflammatory bowel diseases (IBD), Crohn’s disease (CD) and ulcerative colitis (UC), are recognized disorders of the gastrointestinal (GI) tract that may have a variety of extraintestinal manifestations. These include pyoderma gangrenosum, erythema nodosum, uveitis, episcleritis, sclerosing cholangitis, hemolytic anemia, arthritis, and pulmonary disease of various types (1-3). Although tracheobronchitis associated with CD is rare (2, 4), it is another of the pulmonary complications of CD such as chronic bronchitis, bronchiectasis, granulomatous interstitial pneumonitis, bronchiolitis obliterans, fibrosis, alveolitis, or pleuritis. We report herein a young Japanese man with tracheobronchial inflammation accompanying CD, who improved significantly with inhaled corticosteroid (ICS) therapy.

Case Report

A 19-year-old Japanese man with Crohn’s disease (CD), who had been treated with nutrition therapy and mesalazine since May 2005, was admitted to our hospital in June 2006 for prolonged symptoms of general malaise, fever and non-productive cough.

On admission, blood pressure was 121/81 mmHg, pulse rate 111/min, temperature 38.7°C, and SpO2 was 97% on room air. Physical examination showed reddish tonsils and eruptions on his face, neck, and chest regarded as steroid acne. He was a never-smoker. Laboratory data on admission included the following: white blood cell count 10,020/μL, plt 44.9×104/μL, TP 9.1 mg/dL, CRP 11.7 mg/dL, IgE 1,030 IU/mL, IgG 1,890 IU/mL, CH50 53.1 U/mL and ESR 73 mm/h. Beta D-glucan and tests for allergy were negative. Serological tests for Mycoplasma pneumoniae and Chlamydia pneumoniae were negative. Pulmonary function tests were normal (VC 3.66 L, %VC 83.4%, FEV1 3.50 L, FEV1% 91.4%). On admission CDAI (Crohn’s Disease Assessment Index) was 160.6, IOIBD (The International Organization for the Study of Inflammatory Bowel Diseases) assessment score 1/10, CRP positive and ESR normal. As a result, his CD on admission was regarded as neither active nor in remission.

The patient was initially treated with antibiotics for 18 days (PAPM/BP, 1 g/day, 3days, AZM, 500 mg/day, 3days, CFPM, 2 g/day, 12 days), along with codeine phosphate, so
that the dry cough was gradually relieved; however fever and dry cough were not resolved completely. The chest computed tomography (CT) performed for exploring the cause of symptoms revealed an irregular surface on the membranous portion of the tracheal wall (Fig. 1). Flexible fiberoptic bronchoscopy (FFB) showed diffuse whitish granular lesions on the membranous portion of his trachea and both main bronchi (Fig. 2). Tracheal biopsy showed mixed histiocytic and neutrophilic inflammation. The histiocytes formed epithelioid cell granulomas, as vaguely circumscribed as the granulomas in his colonic mucosa of the previous study (Figs. 3A, 3B). Furthermore, non-tuberculous mycobacterial infection in a specimen taking by FFB was negative due to germ culture. It was thought that the airway lesions were caused by CD, because the pathological finding resembled that of the colon. ICS (Fluticasone propionate 400 microgram/day) was prescribed because the patient rejected oral administration of steroids.

A few days later he became afebrile, and after one week his dry cough was relieved. Furthermore, after two weeks, FFB revealed marked improvement in tracheobronchial lesions (Fig. 4). However, two months later, he stopped ICS and the symptom of dry cough and FFB findings recurred.

**Discussion**

We evaluated the effect of ICS therapy by comparing the before and after appearance of tracheobronchial mucosal lining observed by FFB. We were not able to find any previous reports that evaluated the treatment effect of ICS therapy with direct observation by bronchoscopy. To our knowledge, this is the first report to do so.

Tracheobronchial involvement with CD is rare (1), having been reported since 1976 (5) in only 12 cases (1-7) including the present case (7 men and 5 women, ages 19-59 year, average 29). In IBD in general, many forms of bronchial involvement typically develop after colectomy, and this feature is especially true in patients with UC (2). Upper airway involvement tends to occur more frequently in association with active IBD than inactive IBD, though upper airway and GI disease may be closely (1) related. Therefore, the inflammation of the mucosa in these upper airway diseases may be produced by the same mechanism as mucosal inflammation of the GI tract, associated with T-cell lymphocyte and macrophage activation. The possibility exists that common pathogenic antigens that might be present in both GI and tracheobronchial mucosa bring about mucosal immune responses.

On the other hand, it is said that the clinical courses between upper airway and GI disease are not entirely parallel. Although the trigger of the immune response may be essentially the same between the upper airway and large intestine, amplification of the inflammatory process might be different, resulting in unparallel clinical courses between upper
Figure 3A. High magnification micrograph of tracheal biopsy specimen, showing loosely formed and poorly circumscribed granuloma, mixed with neutrophilic infiltrate (arrows).

Figure 3B. Epithelioid cell granulomas in colonic mucosa (circle).

Figure 4. Flexible fiberoptic bronchoscopy findings after treatment show marked improvement. A: trachea, B: main carina

It is reported that the response to both inhaled and oral corticosteroid therapy of upper airway disease in CD was effective. All cases were treated with oral steroids (5 cases), inhaled steroids (2 cases), oral and inhaled (4 cases), and other (1 case) (1-7). All cases treated with steroids experienced an improvement in both their respiratory and colon symptoms. In the present case we used ICS therapy. Because the patient rejected oral administration of steroids, the patient was treated with Fluticasone propionate inhalant (400 μg/day) from the outset. After a week, his dry cough improved, and after two weeks FFB findings showed marked improvement. There remained only small whitish granular lesions on the membranous portions of the trachea (Fig. 4). Accordingly, we have confirmed ICS therapy as effective for inflammatory mucosa of the tracheal wall in CD. Furthermore, it is necessary to continue the ICS therapy for this tracheobronchitis with Crohn’s disease, because in this case his symptoms recurred after the ICS had been stopped for two months.

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References
