Necrotizing tracheobronchitis associated with rheumatoid arthritis

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1. Introduction

Clinically, extra-articular lesions in patients with rheumatoid arthritis (RA) are well known, which may be renal, dermatologic, pulmonary or vascular [1–3]. Occasionally, extra-articular manifestations appear prior to and coincident with RA [4,5]. Localized necrotizing lesions in patients with RA are rare. Here, we report a case of necrotizing tracheobronchitis (NTB) in a patient with RA.

2. Case report

A 64 year-old-man was referred to the hospital because of dry cough since 2 weeks. The patient was an ex-smoker, smoking 2 packs per day for 40 years. His medical history revealed hypertension treated with anti-hypertensive agents. The patient was conscious and alert, with body temperature 36.5 °C, heart rate 122 bpm, and blood pressure 147/88 mmHg. Oxygen saturation was 93% in room air. Coarse crackles were detected bilaterally on chest auscultation. He had felt multiple arthralgias on the limbs symmetrically without swelling or deformation. Chest X-ray showed consolidation on both sides (Fig. 1b). The white blood cell count was 17,500 cells/µL and C-reactive protein level was 26.5 mg/dL. The patient was diagnosed with community-acquired pneumonia, and was subsequently treated with ampicillin/sulbactam and pefoxacin. Blood and sputum cultures were negative, whereas anti-cyclic citrullinated peptide antibody (anti-CCP Ab) was elevated significantly. Antibiotic treatment was ineffective. Bronchoscopy, performed for more thorough examination, revealed white and soft tissues scattered in the trachea that could not be removed by suction or forceps (Fig. 2a). Purulent sputum was absent. Pathological and bacteriological evaluation of the tracheal lesions and lung was performed. Specimens of tracheal mucosa revealed findings consistent with NTB indicated by necrotic tissue with squamous metaplasia and inflammatory cells of tracheal tissue (Fig. 2b). However, the findings did not confirm chondritis. No specific pathogen was identified with PAS or Grocott and Ziehl-Neelsen staining. Lung biopsy indicated organizing pneumonia (OP), whereas eosinophilic leukocytosis and vasculitis were not observed. The patient was diagnosed with NTB and OP. Moreover, diagnosis of RA was determined results of rheumatoid factor and anti-CCP Ab tests, and evaluation of joints' manifestations by orthopedist [6]. After the bronchoscopy, an acute exacerbation of the respiratory condition developed. Immediately, steroid pulse therapy and oxygen therapy were started. Then the both clinical condition were gotten stability. Repeat bronchoscopy to observe...
the tracheal lesion indicated resolution of scattered lesions without scarring; while, re-biopsy samples of trachea showed persistent necrosis. Subsequent, chest radiography indicated improvement (Fig. 3a and b).

3. Discussion

We report a case of NTB as a complication of an exacerbation period of RA, which showed immediate response to steroid therapy.
NTB is a rare disease that occur as a complication of mechanical ventilation in infants and infectious lung disease [7–9]. NTB is usually diagnosed by pathological examination necrotic tissue in the trachea and/or bronchus. The treatment is based on the cause of NTB.

A few case reports have described that NTB occurred by systemic inflammatory disease [10,11], with lesions such as ulcer-like, swelling and irregular changes of tracheal mucosa [10,11]. In contrast, the present case showed scattered lesions in the trachea with intact intraluminal membranes. It is likely that these scattered lesions were an early tracheal manifestation of NTB and therefore, confined locally existed on the trachea. Hiyoshi reported NTB as a complication of ulcerative colitis (UC) that corticosteroids improved both respiratory status and symptoms of UC [10]. Similarly, our patient showed an improvement on bronchoscopic findings, and decreased respiratory and joint-related manifestations in response to steroid therapy. Cystic changes observed on chest radiography were suggestive of pneumatocele due to severe lung inflammation [12,13].

RA, a collagen disease associated with systemic inflammation, may first be identified due to antecedent manifestation of extra-articular such as ocular and pulmonary [4,5]. Although various pulmonary complications of RA have been encountered usually [14–16]. Otherwise, endobronchial lesions related with RA are rare. Ip MS reported that followed-up RA patient occurred to rheumatoid nodules in the trachea respond to steroid therapy. However, the timing of occurrence of endobronchial lesions was unknown [17]. In our case, NTB was possibly a manifestation of RA symptoms. Previously, some extra-articular lesions with necrotizing change were reported [5,18]. Local necrotizing changes with RA are related to the associated sever systemic inflammation. Therefore, steroid therapy can improve these lesions.

4. Conclusion

This is the first report of NTB associated with RA, to our knowledge. This case indicated the steroid therapy improved radiographic and endoscopic findings. In patients diagnosed with NTB, physicians should be aware of underlying systemic inflammatory diseases, especially during the exacerbation period.

Conflict of interest

The authors have no conflict of interest.

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