Respiratory Medicine Case Reports 20 (2017) 31–33

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr

Necrotizing tracheobronchitis associated with rheumatoid arthritis

Shigehisa Kajikawa [a, *], Kazushi Noda [b], Yasuhiro Nozaki [b]

[a] Department of Respiratory Medicine, Tokoname Municipal Hospital, Japan
[b] Department of Respiratory Medicine, Chukyo Hospital, Japan

ARTICLE INFO

Article history:
Received 17 October 2016
Received in revised form
8 November 2016
Accepted 8 November 2016

Keywords:
Necrotizing tracheobronchitis
Rheumatoid arthritis
Bronchoscopy

ABSTRACT

We report a case of necrotizing tracheobronchitis with rheumatoid arthritis. A 64 year-old-man presented with dry cough and was initially diagnosed with community-acquired pneumonia. The patient was admitted; he received initial antibiotic treatment. The treatment was effective and the symptoms worsened. Bronchoscopy was performed for more thorough examination. It showed that white and soft tissues were on the trachea-bronchus. Transbronchial biopsy of the tracheal lesions revealed necrotic tissue with squamous metaplasia and inflammatory cells. Whereas, symmetrical arthralgia of multiple joints of the limbs was noted and rheumatoid factor and anti-cyclic citrullinated peptide antibody of levels were high. According to these results, the patient was diagnosed with rheumatoid arthritis. In this case, necrotizing tracheobronchitis occurred as a result of systemic inflammation associated with rheumatoid arthritis. An acute exacerbation of the patient's respiratory condition was treated with steroid therapy. Tracheal findings and respiratory symptoms were improved by steroid therapy.

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 infiltration of the both lung fields (Fig. 1a). Chest computed tomography revealed the non-segmental patchy shadow and 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 pazufloxacin. 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

* Corresponding author. Respiratory Medicine of Tokoname Municipal Hospital, 479-8510, 3-3-3 Asukadai, Tokoname, Aichi, Japan.
E-mail address: shigeokazy@yahoo.co.jp (S. Kajikawa).

http://dx.doi.org/10.1016/j.rmcr.2016.11.006

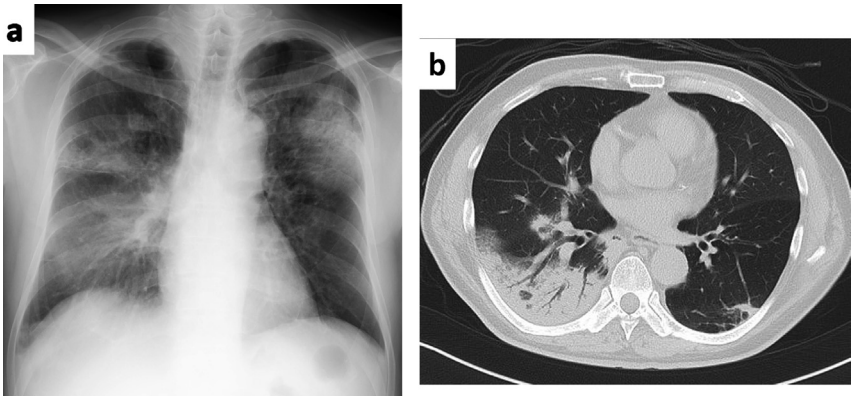

Fig. 1. a) Chest X-ray showed bilateral infiltration without pleural effusion and cardiomegaly. b) Chest computed tomography indicated non-segmental patchy shadow and consolidation with air bronchogram.

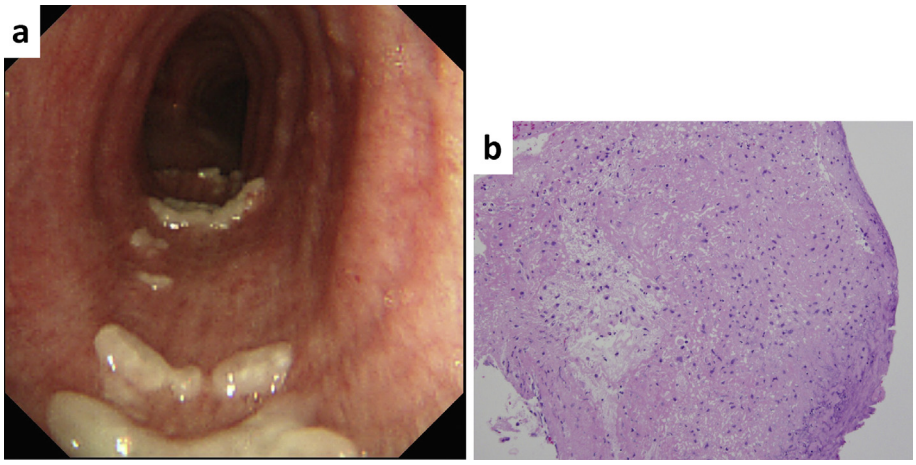

Fig. 2. a) Bronchoscopic finding showed that white and soft tissue were presented on the trachea discontinuity. b) Transbronchial biopsy revealed necrotic tissue with squamous metaplasia and inflammatory cells (Hematoxylin and Eosin staining ×40).

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.

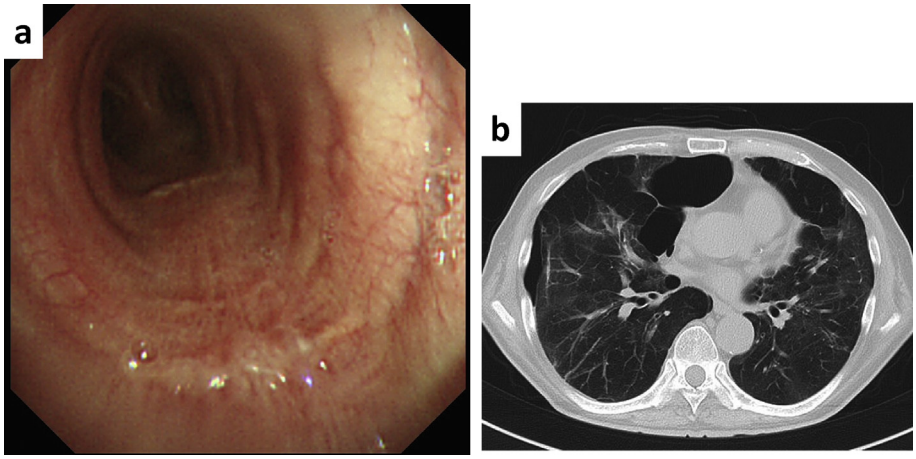

Fig. 3. a) Trachea, after steroid pulse therapy indicated vanishing of the lesions. b) Bilateral consolidation had been improved significantly, and it changed to lung cysts.

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 pneumatocel 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.

References

[1] C. Bartels, C. Bell, A. Rosenthal, K. Shinki, A. Bridges, Decline in rheumatoid vasculitis prevalence among US veterans: a retrospective cross-sectional study, Arthritis Rheum. 60 (9) (2009) 2553–2557.
[2] C.M. Bartels, C.L. Bell, K. Shinki, A. Rosenthal, A.J. Bridges, Changing trends in serious extra-articular manifestations of rheumatoid arthritis among United State veterans over 20 years, Rheumatology 49 (9) (2010 Sep) 1670–1675.
[3] Y. Koseki, C. Terai, M. Moriguchi, M. Uesato, N. Kamatani, A prospective study of renal disease in patients with early rheumatoid arthritis, Ann. Rheum. Dis. 60 (4) (2001 Apr) 327–331.
[4] C. Hoshino, N. Satoh, M. Narita, A. Kikuchi, M. Inoue, Organising pneumonia as the first manifestation of rheumatoid arthritis, BMJ Case Rep. 24 (2011), 2011 Mar.
[5] M. Artifoni, P.R. Rothschild, A. Brézin, L. Guillevin, X. Puéchal, Ocular inflammatory diseases associated with rheumatoid arthritis, Nat. Rev. Rheumatol. 10 (2) (2014 Feb) 108–116.
[6] D. Aletaha, T. Neogi, A.J. Silman, et al., Rheumatoid arthritis classification criteria: an American college of rheumatology/European league against rheumatism collaborative initiative, Arthritis Rheum. 62 (9) (2010) 2569–2581, 2010 Sep.
[7] C. Gaugler, D. Astruc, L. Donato, S. Rivera, C. Langlet, J. Messer, Neonatal necrotizing tracheobronchitis: three case reports, J. Perinatol. 24 (4) (2004 Apr) 259–260.
[8] V. Chechani, V.P. Vasudevan, S.L. Kamholz, Necrotizing tracheobronchitis: complication of mechanical ventilation in an adult, South Med. J. 84 (2) (1991 Feb) 271–273.
[9] J.B. Putnam Jr., C. Dignani, R.C. Mehra, E.J. Anaisse, R.C. Morice, H.I. Libshitz, Acute airway obstruction and necrotizing tracheobronchitis from invasive mycosis, Chest 106 (4) (1994 Oct) 1265–1267.
[10] M. Hiyoshi, K. Kawai, M. Shibuya, T. Ozawa, J. Kishikawa, T. Nirei, et al., Tracheobronchitis with dyspnea in a patient with ulcerative colitis, Intern Med. 54 (7) (2015) 749–753.
[11] H. Matsushima, N. Takayanagi, K. Kurashima, D. Tokunaga, M. Ubukata, et al., Multiple tracheobronchial mucosal lesions in two cases of Churg-Strauss syndrome, Respirology 11 (1) (2006 Jan) 109–112.
[12] Ryou SH, Bae JW, Baek HJ, Lee DH, Lee SW, Choi GH, et al., Pulmonary pneumatocele in a pneumonia patient infected with extended-spectrum β-lactamase producing proteus mirabilis.
[13] M.J. Quigley, R.S. Fraser, Pulmonary pneumatocele: pathology and pathogenesis, Am. J. Roentgenol. 150 (6) (1998 Jun) 1275–1277.
[14] E.J. Kim, H.R. Collard, T.E. King Jr., Rheumatoid arthritis-associated interstitial lung disease: the relevance of histopathologic and radiographic pattern, Chest 136 (5) (2009 Nov) 1397–1405.
[15] N. Tanaka, J.S. Kim, J.D. Newell, K.K. Brown, C.D. Cool, R. Meehan, et al., Rheumatoid arthritis-related lung diseases: CT findings, Radiology 232 (1) (2004 Jul) 81–89.
[16] J.M. Anaya, L. Diethelm, L.A. Ortiz, M. Gutierrez, G. Citera, R.A. Welsh, et al., Pulmonary involvement in rheumatoid arthritis, Semin. Arthritis Rheum. 24 (4) (1995 Feb) 242–254.
[17] M.S. Ip, M.P. Wong, K.L. Wong, Rheumatoid nodules in the trachea, Chest 103 (1) (1993 Jan) 301–303.
[18] R. Koide, A. Isoo, K. Ishii, A. Uruha, M. Bandoh, Rheumatoid leptomeningitis: rare complication of rheumatoid arthritis, Clin. Rheumatol. 28 (9) (2009 Sep) 1117–1119.