Clinical Rheumatology (2020) 39:2017–2018
https://doi.org/10.1007/s10067-020-05096-2

CLINICAL IMAGE

# Multiple fungus balls in rheumatoid arthritis–associated honeycomb lung

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Received: 21 February 2020 / Revised: 28 March 2020 / Accepted: 8 April 2020 / Published online: 28 April 2020
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## Presentation

A 78-year-old woman presented to the respiratory disease department with exertional dyspnea. Three years previously, she was diagnosed with seropositive rheumatoid arthritis (RA) with concomitant usual interstitial pneumonia (UIP) in a different hospital and had been on a regimen of glucocorticoids and tacrolimus hydrate (Fig. 1a). Bilateral fine crackles were heard on chest auscultation. Chest computed tomography revealed bilateral honeycombing with basal predominance, progression of fibrosis in the left lower pulmonary lobe, and multiple recently developed nodules in the clustered cystic airspaces in the right lower pulmonary lobe (Fig. 1b). Although the sputum culture result was negative for mycosis, *Aspergillus* galactomannan antigen value was 3.5 (cutoff index, < 0.5). Therefore, chronic pulmonary aspergillosis with concomitant RA-associated interstitial pneumonia was diagnosed. Despite treatment with antifungal agents, including voriconazole and itraconazole, and reversal of tacrolimus, she died 20 months after clinical presentation due to progression of aspergillosis.

## Discussion

Chronic pulmonary aspergillosis is a well-recognized complication of immunosuppression, and *Aspergillus* species grow and multiply in preexisting cavities or cysts in patients with pulmonary disease, including chronic obstructive pulmonary disease or sarcoidosis [1]. However, few cases of honeycomb cysts with aspergillomas have been reported, and the etiologies underlying UIP are usually idiopathic pulmonary fibrosis (IPF) or systemic sclerosis [2–4]. Although aspergillosis in patients with RA has been reported, to our best knowledge, multiple mycetoma in the lungs with honeycombed interstitial fibrosis associated with RA has not been reported. Patients with RA and UIP have a poor prognosis [5]. Since *Aspergillus* species usually form in preexisting lung lesions, we believe existing interstitial lung disease was more involved than steroids or immunosuppressive agents in the onset of pulmonary aspergillosis in this patient. When treating patients with RA and comorbid lung disease, clinicians should pay attention to lung infections, especially when administering steroids and immunosuppressive agents.

**Acknowledgments** We thank Shigehiko Karino (Department of General Medicine & Infectious Disease, Koseiren Takaoka Hospital, Takaoka, Toyama, Japan) for the valuable advice on antifungal treatment.

## Compliance with ethical standards

**Disclosures** None.

**Ethical approval** The patient has passed away. Thus, informed consent for publication and related images has been obtained from patient's relatives.

## References

1. Limper AH, Knox KS, Sarosi GA, Ampel NM, Bennett JE, Catanzaro A, Davies SF, Dismukes WE, Hage CA, Marr KA, Mody CH, Perfect JR, Stevens DA, American Thoracic Society Fungal Working Group (2011) An official American Thoracic Society statement: treatment of fungal infections in adult pulmonary and critical care patients. Am J Respir Crit Care Med 183:96–128
2. Louza GF, Zanetti G, Marchiori E (2018) Aspergilloma in honeycomb cysts and paraseptal emphysema: an unusual association. Arch Bronconeumol 54:110–111

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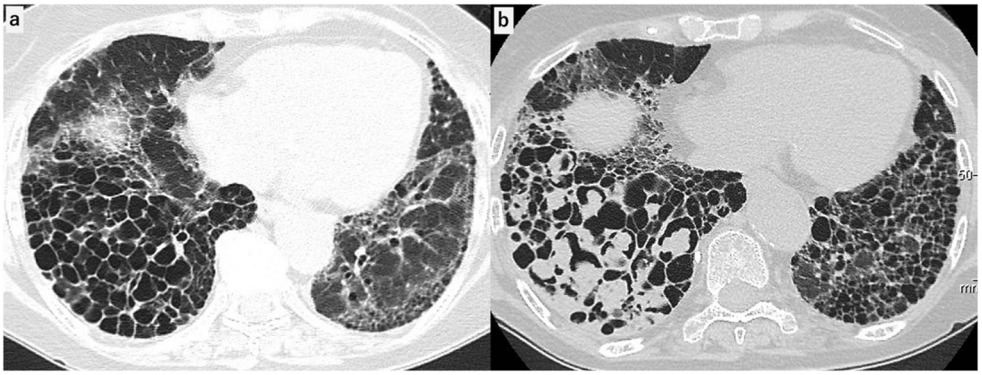


**Fig. 1** **a** Computed tomography 3 years before presentation, revealing bilateral honeycombing with basal predominance. The radiological pattern was usual interstitial pneumonia. **b** Computed tomography revealed multiple recently developed nodules in the clustered cystic airspaces in the right lower pulmonary lobe and progression of fibrosis in the left lower pulmonary lobe

3. Nandi S, Santra A, Ghoshal L, Kundu S (2015) Interstitial lung disease in systemic scleroderma, complicated with bilateral pulmonary aspergilloma: an unusual association. J Clin Diagn Res 9:11–13
4. Kumar N, Mishra M, Singhal A, Kaur J, Tripathi V (2013) Aspergilloma coexisting with idiopathic pulmonary fibrosis: a rare occurrence. J Postgrad Med 59:145–148
5. Kim EJ, Elicker BM, Maldonado F, Webb WR, Ryu JH, van Uden JH, Lee JS, King TE, Collard HR (2010) Usual interstitial pneumonia in rheumatoid arthritis-associated interstitial lung disease. Eur Respir J 35:1322–1328

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