Bilateral spontaneous pneumothorax in a patient with longstanding ankylosing spondylitis

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Ankylosing spondylitis (AS) is a chronic inflammatory disease and pulmonary involvement is usually asymptomatic in the early disease^{1,2}. Pneumothorax is defined as accumulation of air or gas in the pleural cavity. Spontaneous pneumothoraces (SP), which occur in the absence of thoracic trauma, are classified as primary (without underlying pulmonary disease) or secondary (with underlying pulmonary disease). Spontaneous pneumothoraces in AS are seldom rare³⁻⁵. We herein report a male patient with longstanding AS who developed spontaneous pneumothorax which resolved without intervention.

CASE

Fifty-five-year old male with long standing AS attended to our clinic with persisted cough and mild dispnea. He was diagnosed with AS nearly 30 years ago. He was slim, had typical stroop posture and chest expansion was highly restricted (1 cm). He had not regularly used disease modifying anti-rheumatic drugs but only used non-steroid anti-inflammatory drugs on demand and verapamil for the treatment of hypertension. He had no history for smoking and blunt trauma.

On physical examination; he had respiratory rate of 23/min, pulse rate of 80 /min, and blood pressure of 140/96 mmHg. On chest auscultation there was diffuse wheezing and diminished breath sounds and increased resonance particularly on the middle and lower zones of both lungs. Erythrocyte sedimentation rate was 35 mm/h, C-reactive proteine (CRP) level was 13.5 mg/l and complete blood count was normal. The patient was consulted at the department of chest diseases. Chest X-ray revealed suspicious pneumothorax and thoracic computerized tomography (CT) was taken immediately. Pulmonary function tests revealed restricti-

ve pattern. Thoracic CT demonstrated bilateral pneumothorax, ground-glass densities, bronchiectasis, parenchymal-bands and honeycomb appearance (Figure 1). The patient refused any intervention. Therefore followed and discharged after the control chest X-ray which did not reveal any progression. After one month, he did not have cough or dispnea and spontaneous resolution of pneumothorax was confirmed on chest X-ray. The patient was followed for one year without any recurrence of the pneumothorax.

DISCUSSION

Pulmonary involvement in AS is usually asymptomatic and mostly manifested with abnormalities of the thoracic cage and parenchyma of the lung. Thoracic CT findings usually reveal pleural thickening, interstitial lung disease, ground-glass appearance, bronchiectasis, emphysema, thickening of the bronchial wall, subpleural and parenchymal bands or blebs^{1,2}. Rosenow *et al* reported 2 cases of SP in 2080 patients with AS⁶. Lee *et al.* reported the incidence of SP as 0.29% (3/1028) in



FIGURE 1. Computerized tomography scan shows parenchymal bands, bronchiectasis and bilateral pneumothorax

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patients with AS, and suggested that this frequency was higher than the normal population⁵. Rupture of bullea has been considered as the cause of SP in all of these patients who had also underlying fibrobullous abnormalities of the lungs. These patients were treated with interventions like tube thoracotomy, video-assisted thoracoscopic surgery (VATS) or talc pleurodesis⁵.

One of the major contributors in the development of SP is slenderness⁷. Our patient had a thin stature (but neither Marfanoid nor had joint hypermobility). In small-size primary SP, close observation is the preferred management; however larger pneumothoraces need some interventions as recommended in guidelines^{8,9}. Other reported cases of SP associated with AS have been treated with tube thoracotomy, surgical interventions and chemical pleurodesis^{3,4}. Additionally, care should be taken in patients with AS while performing respiratory exercises¹⁰.

CONCLUSION

Although rare, spontaneous pneumothorax is likely in patients with AS. Patients with AS should be scrutinized for signs and symptoms of pneumothorax. In patients with spontaneous pneumothorax with underlying pulmonary involvement, possible respiratory insufficiency and high risk for recurrence could be expected.

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