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Case report

Pleuroparenchymal sarcoidosis - A recognised but rare manifestation of disease

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A B S T R A C T

Pleural involvement is rare in sarcoidosis. The presence of a large symptomatic effusion in a patient with sarcoidosis should therefore prompt further investigation for an alternate aetiology. Here we present a case of confirmed pleuro-parenchymal sarcoidosis. We discuss the important differential diagnoses and review the current literature.

1. Case report

A 67-year-old Caucasian non-smoker female with a past medical history of hypertension and hypothyroidism presented to our clinic with a 12-month history of exertional dyspnoea and dry cough. She was otherwise well with no systemic, nasal or gastro-oesophageal symptoms. Her prescribed medications were levothyroxine, aspirin and losartan and these had not changed in recent years. She denied atopy, exposure to previous TB or recent travel and systems enquiry did not reveal any eye, genitourinary or gastrointestinal symptoms. She had worked throughout her career in administration.

Vital signs were recorded as pulse 80 bpm, blood pressure 130/50 mmHg, respiratory rate 17 and oxygen saturations of 97% air. Cardiorespiratory examination was normal. Lung function revealed: FEV1 1.14 (57% predicted), FVC 1.60 (67% predicted), FEV1/FVC ratio 71%, TLCO 3.30 (71% predicted), TL_{CO} 5.48 (79% predicted) and KCO 1.95 (131% predicted). ECG showed sinus rhythm with no evidence of heart block. Chest X-ray showed blunting of the costophrenic angles with scattered peribronchial thickening predominantly within the mid and lower zones (Fig. 1a). High resolution computed tomography (HRCT)-Chest demonstrated bilateral hilar and central mediastinal nodal calcification, in the absence of significant lymphadenopathy, with perilymphatic nodularity in the upper and mid zones of the lungs and bilateral pleural effusions, left larger than right (Fig. 1bi). Echocardiogram was normal.

Laboratory investigations revealed a mildly elevated ACE level (summarised in Fig. 2). Left sided chest ultrasound demonstrated an anechoic effusion, with an exudative lymphocytic yellow aspirate (lymphocytes 70%), with a normal adenosine deaminase (ADA) level that was negative to TB culture (summarised in Fig. 2).

Given the suspicion of pulmonary sarcoidosis, flexible bronchoscopy was undertaken to obtain a tissue diagnosis. Bronchoalveolar lavage samples demonstrated 90% macrophages and 10% lymphocytes. Cultures were negative for TB and fungi. Endobronchial biopsies (Fig. 1 ci) demonstrated discrete non-necrotising epithelioid granulomas consistent with sarcoidosis. Transbronchial biopsies were non-diagnostic.

Sarcoidosis was considered the most likely overarching diagnosis and thus the patient was commenced on a tapering course of oral prednisolone therapy over the following 9 months. Initial clinical improvement was followed by progressive breathlessness. HRCT demonstrated stable parenchymal appearances but worsening right effusion (Fig. 1 a and bii). MRI excluded cardiac sarcoidosis, demonstrating mild diastolic dysfunction only. Attempted diuresis, a repeat trial of tapering doses of prednisolone then introduction of methotrexate (12.5mg weekly) in conjunction with prednisolone 10mg had no significant benefit, with gradual worsening of bilateral effusions and symptoms over the subsequent 12 months (Fig. 1a). In light of this deterioration a definitive diagnosis was sought. Medical thoracoscopy was not technically feasible and thus she proceeded to right-sided video-assisted thoracoscopic (VATS) wedge lung biopsy and pleural biopsy with planned therapeutic drainage and talc pleurodesis.

Pleural biopsies showed features of chronic pleuritis with associated granulomata (Fig. 1c ii-iii). The lung VATS biopsy sections (Fig. 1 c iv) showed pleural and subpleural granulomatous inflammation. A small interstitial granuloma was also noted. Ziehl-Neelsen stain was negative. Overall, these features are most suggestive of pleuro-parenchymal sarcoidosis.

A further trial of combination therapy (hydroxychloroquine (200mg bd), azathioprine (150mg daily) and prednisolone (10mg)) was attempted, stabilising the parenchymal disease over a 6-month period,

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Abbreviations used

FEV1	Forced expiratory volume in 1 second
FVC	Forced vital capacity
TLCO	Transfer factor for carbon monoxide
KCO	Transfer coefficient
ECG	Electrocardiogram
HRCT	High Resolution Computed Tomography
PE	Pleural effusion
ACE	Angiotensin converting enzyme
MRI	Magnetic resonance imaging

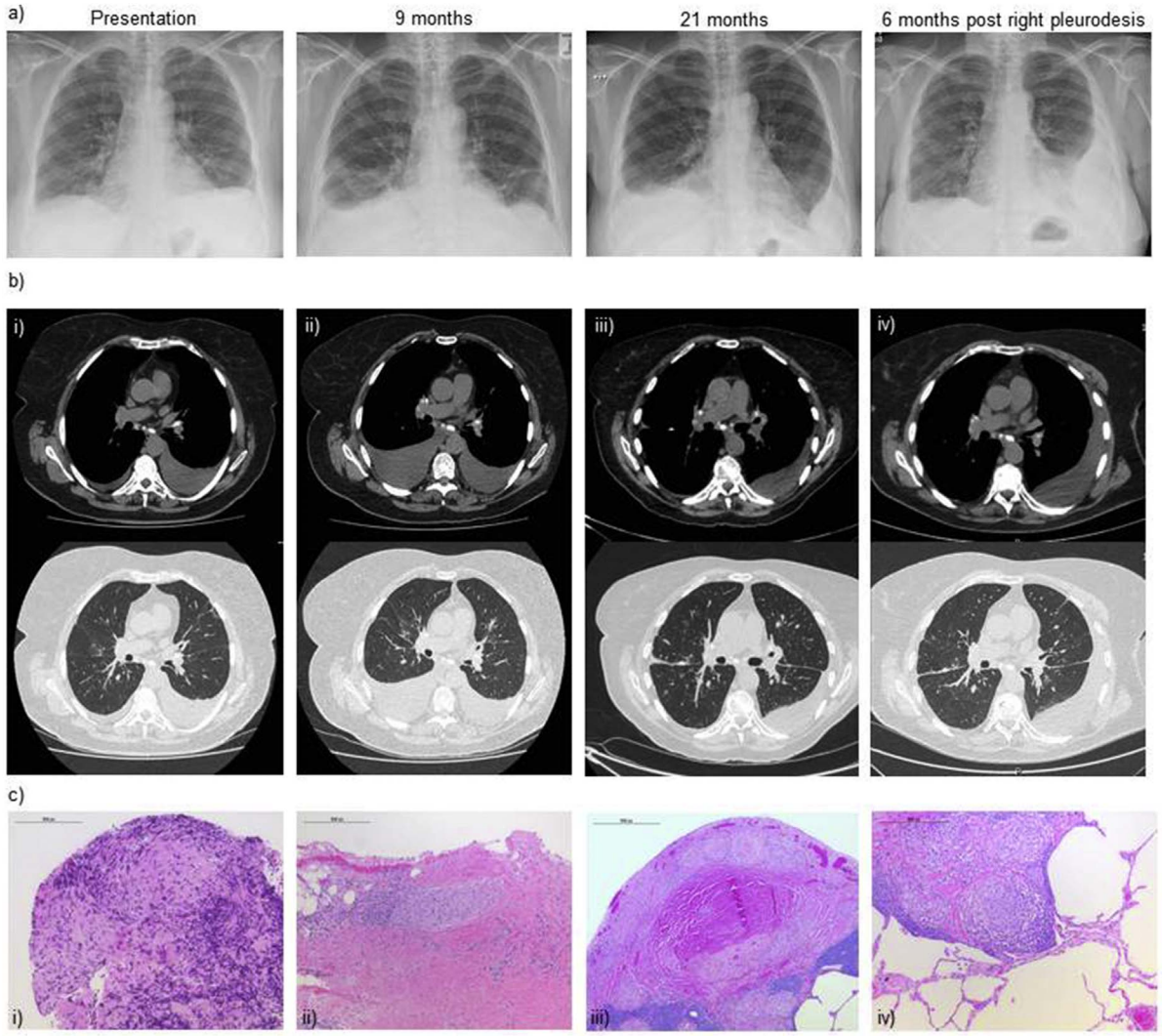

Fig. 1. a) CXR and representative HRCT images taken at presentation, 9 months and 21 months and 6 months post right talc pleurodesis. (i) CXR at presentation demonstrates blunting of the costophrenic angles with scattered peribronchial thickening in the mid and lower zones. (ii) CXR 9 months later following a tapering course of prednisolone therapy and maintenance of 10mg daily, demonstrating blunting of the costophrenic angles with worsening of the right sided effusion. iii) CXR at 21 months following repeated trials of tapering prednisolone, attempted diuresis and trial of methotrexate therapy in conjunction with 10mg prednisolone: CXR demonstrates persistent pleural effusions with apparent worsening of both right and left effusions. iv) CXR 6 months' post pleurodesis on prednisolone 10mg once daily, hydroxychloroquine 200mg twice daily and azathioprine 150mg once daily. No reaccumulation of right effusion. b) Representative HRCT images taken at presentation and 9 months later: Soft tissue windows above and lung windows below of i) HRCT images taken at presentation demonstrating mediastinal and hilar nodal calcification, perilymphatic nodularity and bilateral pleural effusions, left larger than right. ii) Nine months later following a tapering course of prednisolone therapy and maintenance of 10mg daily. Stable mediastinal and hilar nodal calcification and perilymphatic nodularity with bilateral pleural effusions, worsening on the right. iii) Four weeks post right talc-pleurodesis. Worsening nodularity in the context of slight increase of left sided-effusion. iv) Six months after right-talc pleurodesis. Stable appearances of lung nodularity in the context of worsening left effusion on prednisolone, hydroxychloroquine and azathioprine. c) Histological analysis i) Endobronchial biopsy demonstrates non-caseating granuloma. ii) Right sided VATS pleural biopsy demonstrating pleural and iii) subpleural based non-caseating granulomatous inflammation with surrounding lymphocytic infiltration and accompanying fibrosis. iv) Concurrent VATS lung wedge biopsy demonstrates an additional interstitial sarcoidal granuloma. ZN and Wade-Fite stains were negative excluding tuberculosis. No malignancy was seen. Scale bar indicates 500 μm.

but was associated with continued pleural fluid accumulation on the left, in the absence of re-accumulation on the right. The patient was subsequently referred for left-sided talc pleurodesis with a proposed plan to seek funding for anti-Tumour Necrosis Factor-α biological therapy if this failed.

2. Discussion

Sarcoidosis is a multisystem disease characterised by non-caseating granulomas. Whilst it most frequently affects the lungs and lymph nodes, extrapulmonary presentations of the skin and eyes are also common. The differential diagnosis for nodal calcification and

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