Letters to the Editor

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Spontaneous pneumothorax: an unusual presentation of rheumatoid arthritis

Sir, We read with interest the article by Gotsman et al. [1] describing a case of spontaneous pneumothorax in a rheumatoid arthritis (RA) patient treated with methotrexate. Spontaneous pneumothorax secondary to pulmonary rheumatoid nodules is an uncommon complication of RA. Cases described so far have been male patients with longstanding RA [2]. We describe a female patient who developed a spontaneous right-sided pneumothorax before developing arthritic symptoms.

A 40-yr-old lady developed dyspnoea on minimal exertion following a heavy cold. She also complained of right-sided pleuritic chest pain radiating to her back. There was no significant past medical history and she was not taking any medication. She worked as a cook in a residential home and was a smoker of 5 cigarettes per day. On examination, she was found to have clinical features of a right-sided pneumothorax, which was confirmed by chest X-ray (Fig. 1). The eosinophil count was slightly raised at 0.6 x 10⁹/l (normal range 0.04–0.4 x 10⁹/l); the full blood count, liver function tests and urea and electrolytes were unremarkable. The pneumothorax failed to resolve with needle aspiration and chest tube insertion. Subsequently she underwent right side video assisted thoracoscopic surgery (R VATS) apical bullectomy, apical pleurectomy and pleural abrasion. She made an uneventful recovery. The histology of the excised tissue showed granulomatous pleural inflammation, fibrous exudate, palisading histiocytes and scattered multinucleated giant cells, consistent with a rheumatoid nodule (Fig. 2). Staining for acid fast Bacilli (AFB) was negative.

One week after surgery, the patient developed acute onset of pain and swelling in her left knee and the small joints of her hands. She also complained of painful feet and early-morning stiffness lasting more than 30 min. On examination, there were no subcutaneous nodules. Cardiovascular and respiratory examinations were unremarkable. There was active synovitis affecting a number of proximal interphalangeal joints and the second and third metacarpophalangeal joints bilaterally. There was synovitis of the left knee with 15° fixed flexion deformity. Blood tests showed a slightly elevated erythrocyte sedimentation rate (ESR; 21 mm/h), and she was positive for rheumatoid factor, with a titre of 1:320. She was negative for antineutrophil cytoplasmic antibodies and the angiotensin-converting enzyme level was <25 U/l (normal range 0–55 U/l). X-ray of the hands and feet showed no erosions. She responded well to intra-articular steroid injections and sulphasalazine 1 g twice daily.

Pulmonary manifestations of RA include pleural disease, pulmonary infections, arteritis with pulmonary hypertension, obliterator bronchiolitis, bronchiectasis, nodules and Caplan’s syndrome [3]. Pulmonary necrobiotic nodules are a rare manifestation of RA, usually associated with the presence of subcutaneous nodules. They are seen mainly in men with longstanding seropositive RA. They may vary in

Fig. 1. Histology of the excised tissue showed granulomatous pleural inflammation, fibrous exudate, palisading histiocytes and scattered multinucleated giant cells, consistent with a rheumatoid nodule.

Fig. 2. Chest X-ray showing right-sided pneumothorax.
size. The nodules can cause haemoptysis, pneumothorax or bronchopleural fistula depending on their location. Pneumothorax in RA may be associated with eosinophilia, high ESR and other pulmonary manifestations of RA, such as pulmonary fibrosis and vasculitis [4]. Our patient also had eosinophilia at the time of presentation.

Anecdotal reports suggest that methotrexate might exacerbate subcutaneous nodulosis in RA [1]. Gotsman et al. [1] has described a case of spontaneous pneumothorax developing in an RA patient treated with methotrexate. As in other cases of pneumothorax secondary to pulmonary rheumatoid nodules, their patient was a male with longstanding RA. In view of these reports, we avoided disease-modifying treatment with methotrexate in our patient because of concern that this might aggravate pulmonary nodulosis.

To our knowledge this is the first described case of a patient with spontaneous pneumothorax due to a pulmonary rheumatoid nodule which preceded the development of RA.

The authors have declared no conflicts of interest.

S. Saravana, T. Gilloch, F. Abourawi, M. Peters, A. Campbell, S. Griffith

Department of Rheumatology, Department of Endocrinology and Diabetes, Department of Pathology, Diana Princess of Wales Hospital, Grimsby DN33 2BA, UK;

Department of Cellular Pathology, Hull Royal Infirmary, Hull, UK and Department of Cardiothoracic Surgery, Castle Hill Hospital, Cottingham, UK

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Correspondence to: S. Saravana, Department of Rheumatology, Diana Princess of Wales Hospital, Grimsby DN33 2BA, UK. E-mail: adersh555saravana@hotmail.com


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Renal microinfarction in Behc¸et’s disease

Sir, We describe a patient with Behc¸et’s disease and renal cortical microinfarct whose renal impairment stabilized following anticoagulation. Behc¸et’s disease is a chronic inflammatory vascular disorder of unknown aetiology. Vasculitis, thrombosis and infarction have been reported in a variety of organs with varying frequency. Renal involvement in Behc¸et’s disease is uncommon and includes amyloidosis, crescentic glomerulonephritis and IgA nephropathy. In the literature there is only one report of acute renal infarction due to Behc¸et’s disease [1]. We describe a patient with Behc¸et’s disease with bilateral renal microinfarcts confirmed by computed tomography (CT) and DMSA renal perfusion scans. The patient gave his informed consent for publication.

A 38-yr-old West African man developed sore throats, headaches, swelling fevers and joint pains mainly in the right wrist, which was hot, tender and swollen. During the pyrexial episodes he noted macroscopic haematuria. He then developed oral and genital ulcers, sterile pustules and a symmetrical peripheral neuropathy. Investigations showed negativity for antinuclear antibodies, anti-extractable nuclear antigen, anti-DNA and antiperoxygenin antibodies, lupus anticoagulant and antineutrophil cytoplasmic antibodies, and normal serum complement levels. Renal function was abnormal: urea 12.4 mmol/l, serum creatinine 155 mol/l. The erythrocyte sedimentation rate (ESR) was 96 mm in the 1st hour and C-reactive protein (CRP) was 70 mg/l. Radiographs of the chest, hands and feet were normal. A formal pathergy test was negative but he frequently noted pustules developing at venepuncture sites. Abdominal CT scan and an intravenous urogram showed bilateral kidney scarring. A renal biopsy showed no evidence of a thrombotic microangiopathy and skin biopsy showed a neutrophil-rich perivascular infiltrate in the mid-dermis. He was diagnosed with Behc¸et’s disease complicated by renal microinftarction. HLA B51 was negative. He responded well to three 500 mg methyl-prednisolone pulses and 20 mg oral prednisolone reducing to 12.5 mg daily and colchicine 0.5 mg twice daily. However, he continued to have flares of Behc¸et’s disease every 6 weeks with episodes of fever, oral, nasal and genital ulcers, skin lesions and arthralgia. Azathioprine was commenced but was stopped after 6 months because of neutropenia. On methotrexate 10 mg weekly he improved, with normalization of his ESR and CRP levels. Five years after presentation, he developed hypertension. His creatinine clearance was 74 ml/min and there were no casts in his urine. He responded to lisinopril and nifedipine.

Two years later, whilst his disease was still in remission, he was investigated following an episode of macroscopic haematuria. His radioisotope glomerular filtration rate was 64 ml/min. Cystoscopy was normal and a DMSA scan showed bilateral shrunken scarred kidneys with an equal division of glomerular filtration rate. Anti-phospholipid antibodies remained persistently negative and a full thrombophilia screen was negative. He had never had any urinary tract infections and the most likely diagnosis was bilateral renal cortical