CASE REPORT

Sarcoid lung

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SUMMARY

Sarcoidosis is a multisystem disease with a varied clinical presentation. The lung disease is common and is responsible for most of the morbidity and mortality associated with sarcoidosis. Tuberculosis remains a prevalent disease in some countries, such as Portugal. There are reports of sequential occurrence of these identities. The authors present a case of a patient with lymphatic tuberculosis who, some years after treatment of this infection, was diagnosed with sarcoidosis in an atypical pseudotumoural presentation.

BACKGROUND

Sarcoidosis is a chronic inflammatory granulomatous disease characterised by granuloma without caseum, which more frequently affects the lung, although multiorgan involvement is common. Changes in chest imaging are described at some stage of the disease in 90% of patients, more common as bilateral hilar lymphadenopathy and interstitial lung disease. Atypical manifestations, such as pseudotumours, can also occur.¹

The aetiology of sarcoidosis is not fully understood; the association between sarcoidosis and tuberculosis remains controversial.²

CASE PRESENTATION

A 24-year-old previously healthy man presented a bilateral panuveitis, complicated with retinal vasculitis and retinal haemorrhage, associated with inflammatory arthralgia in the metacarpophalangeal and interphalangeal joints. There were no other symptoms and there was no epidemiological context.

At this point, the main differential diagnoses were infectious (including mycobacteria and fungi). Investigation revealed mild thrombocytopaenia, elevated C reactive protein (27.7 mg/L), erythrocyte sedimentation rate (67 mm/1sth) and adenosine deaminase (34/L). Immunological study showed normal immunoglobulins (IgG 1280 mg/dL, IgA 185 mg/dL and IgM 281 mg/dL), negative antineutrophil cytoplasmic antibody, antinuclear antibodies, European Nucleotide Archives, double-stranded DNA <10 IU/mL, RR 9.81 (normal <30 IU/mL) and circulating immune complexes >100 g/mL. Immunophenotyping of blood presented an increase in B lymphocytes (19%), increase in T lymphocyte TCR γ/δ (23.3%) and the presence of a polyclonal B-cell population. Serology test for HIV, syphilis, toxoplasmosis, herpes 1 and 2, Epstein-Barr virus, cytomegalovirus and *Coxiella burnetii* were negative. Mantoux test and *Interferon-Gamma Release Assay* were positive. A chest CT scan was performed and presented a subcarinal mediastinal mass

(figure 1A, B) and bilateral pulmonary micronodules (3–4 mm; figure 2). At this time, a bronchoscopy was performed; immunophenotyping of bronchoalveolar lavage (BAL) lymphocytes revealed lymphocytic alveolitis, mild eosinophilia (3.2×10^5 cells/mL, 69.4% lymphocytes and 1.6% eosinophils), with a CD4 predominance (88.2%) and a high CD4/CD8 ratio (9.3). Caseum was collected from transtracheal aspiration, which showed a small cluster of epithelioid cells with granulomatous design. Culture tests (blood, sputum, urine and BAL) were negative.

Lymphatic tuberculosis was diagnosed and anti-tuberculosis drugs were initiated and continued for 18 months, along with systemic corticosteroid therapy for ocular involvement, with resolution of symptoms.

Five years later, the patient experienced exertional dyspnoea, fatigue, worsening of arthralgia associated with articular swelling and emergence of

Figure 1 (A) Transversal view of chest CT scan with subcarinal mediastinal mass, lateralised to the right, with 82×70 mm, suggestive of adenopathy, without enhancement after contrast effect. (B) Coronal view of chest CT scan with subcarinal mediastinal mass, lateralised to the right, with 82×70 mm, suggesting adenopathy, without enhancement after contrast effect.

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Figure 2 Chest CT scan (in lung window) with 3–4 mm bilateral pulmonary micronodules, with the largest, 16 mm, in the right anterior segment of the lower lobe, with irregular borders and infiltration of surrounding parenchyma.

nodular skin lesions on the elbows and face (figure 3A, B). A new chest CT scan showed a pulmonary nodule on the lower right lobe (69×54 mm; figure 4A, B) and nodular thickening of fissures. A transthoracic biopsy of the pulmonary nodule revealed a chronic inflammatory, granulomatous process with multinucleated giant cells; mycobacteriological examination and *Mycobacterium tuberculosis* (MTB) PCR were negative. Sarcoidosis with pulmonary, ocular, cutaneous and articular involvement was diagnosed.

DIFFERENTIAL DIAGNOSIS

With such a prominent mediastinal lymphadenopathy, the most probable diagnoses were lymphoproliferative diseases, and

Figure 3 (A) Nodular and erythematous skin lesions on the face. (B) Nodular and erythematous skin lesions on the elbows.

autoimmune and infectious diseases (particularly by mycobacteria or fungi). A supplementary study excluded most of these pathologies and made the diagnosis of sarcoidosis with multisystemic involvement.

OUTCOME AND FOLLOW-UP

Treatment with corticosteroids (1 mg/kg) was carried out for 6 months, with clinical improvement. Subsequently, slow and progressive corticosteroid weaning over 1 year was performed, with recurrence of symptoms. Hydroxychloroquine was ineffective and methotrexate (MTX; 10 mg/week) was initiated, with resolution of skin, articular and pulmonary nodule symptoms. The patient has been on MTX for 1 year, with regular follow-up, and is asymptomatic. Pulmonary changes in thoracic CT disappeared completely; some small calcified mediastinal ganglia remained, however, the skin lesions disappeared.

DISCUSSION

Sarcoidosis is a chronic granulomatous disease that can affect multiple organs; pulmonary involvement is frequent but usually not with this presentation. In about 1% to 4% of cases, coalescence of interstitial granulomas lead to formation of masses, which are pseudotumoural forms of sarcoidosis; these should be considered in the differential diagnosis of pulmonary nodules and masses.^{1–4}

Some authors contend that MTB may play a role in the aetiology of sarcoidosis, due to the similar clinical and histopathological outcomes.⁵ In recent decades, studies have been

Figure 4 (A) Chest CT scan (lung window) with pulmonary nodule on lower right lobe (69×54 mm). (B) Chest CT scan (mediastinal window) with pulmonary nodule on lower right lobe (69×54 mm).

published documenting the presence of MTB DNA in patients with sarcoidosis.⁶ Similarly, there are reports of sequential occurrence of sarcoidosis and tuberculosis.^{7 8} Currently, some authors propose that MTB antigens can induce sarcoidosis in genetically predisposed individuals.^{9 10} In this case, it is possible that the diagnosis of sarcoidosis and tuberculosis (pulmonary and lymphatic) coexist, despite microbiological isolation of MTB not having been documented.

The goals of sarcoidosis management are to prevent or control organ damage, relieve symptoms and improve the patient's quality of life. While a significant percentage of sarcoidosis patients never need therapy, there are several individuals who do require treatment. Despite the fact that hydroxychloroquine is effective in patients with dermatological involvement and joint

manifestations, this was not so in our case. Methotrexate is one of the most commonly used corticosteroid-sparing therapies for sarcoidosis, due to its effectiveness and, at the dosages used to treat sarcoidosis, relatively low risk of side effects.

Contributors AM was responsible for the diagnostic and treatment decision and contributed to the writing of the article.

Competing interests None declared.

Patient consent Obtained.

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Learning points

- ▶ Sarcoidosis is a chronic inflammatory granulomatous disease that more frequently affects the lung, although multiorgan involvement is common; the diagnosis of sarcoidosis can never be assured; it is a diagnosis of exclusion, which sometimes can be problematic.
- ▶ Pseudotumoural forms of sarcoidosis should be considered in the differential diagnosis of pulmonary nodules and masses.
- ▶ The possible role of *Mycobacterium tuberculosis* in the aetiology of sarcoidosis is currently under discussion.
- ▶ The goals of sarcoidosis management are to prevent or control organ damage, relieve symptoms and improve the patient's quality of life.

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