Unusual association of diseases/symptoms

Giant tuberculoma in an adolescent: atypical form of tuberculosis

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Summary

A tuberculoma is a rare form of presentation of tuberculosis (TB) in children. We describe the case of a 13-year-old girl, with 3 weeks of progressive tiredness and asthenia and a 48 h fever and cough. Physical examination revealed diminished pulmonary sounds in the left lower hemithorax. A chest radiograph showed an oval hypotransparency image in this location. The TB skin test was anergic and sputum was negative for acid-fast bacilli (AFB). The thoracic CT revealed a cystic mass in the left lower half hemithorax, compressing the adjacent pulmonary lobe, with double non-calcificated membrane. The exeresis of the pulmonary mass was performed and the anatomopathological study revealed a tuberculoma with AFB. TB treatment was established with a favourable clinical and radiological evolution. TB is a prevalent disease around the world. In this case, due to the mass dimensions and adjacent-organ compression, surgery was essential for a favourable clinical evolution.

BACKGROUND

Tuberculosis (TB), an airborne infection caused by *Mycobacterium tuberculosis*, is still a major cause of morbidity and mortality. Patients with an active infection may be asymptomatic, may have mild symptoms or severe disease. If left untreated, a patient with active TB infects approximately 10–15 other people per year.¹

Although the progression from TB infection to disease is uncommon (5–10%), children have a higher risk as well as an increased risk of disseminated infection.² Adolescents may have clinical manifestations similar to adults (secondary disease), including weight loss, fever, productive cough, haemoptysis and night sweats. Pulmonary TB represents 80–90% of all TB infection in immune competent patients.

Pulmonary tuberculoma is uncommon.^{2–3} It is a well-circumscribed nodule or mass located in the lungs. It is a caseous mass encapsulated by multiple concentric layers of connective tissue without surrounding inflammation or spread. Tuberculoma can be the manifestation of both primary and postprimary TB and it was reported to be manifested in 6–95% of postprimary TB. Without treatment, the natural course of pulmonary tuberculoma can be classified as: progressive, regressive and stationary (30–50% of patients).⁴

A pulmonary tuberculosis lesion can be mistaken for lesions of another nature, namely bacterial or parasitic, neoplastic or autoimmune.

We present a case of a giant tuberculoma as a form of presentation of postprimary pulmonary TB, the diagnosis and treatment of which was a challenge.

CASE PRESENTATION

The patient is a 13-year-old girl, born in Luanda, Angola, and resident in Portugal since the age of 3 years. She had no notable personal or familiar history and no known contact

with the illness. She annually travelled to Angola and the last trip had taken place 2 years earlier. She was fully immunised according to the Portuguese immunisation programme, including bacillus Calmette-Guerin vaccination.

Three weeks before admission she complained of asthenia and tiredness for mild efforts. She was evaluated by her general physician, who diagnosed a ferropenic anaemia; oral iron was started with the symptom's persistence. She came to our hospital complaining of a dry cough, myalgia and a 48 h duration mild fever.

At admission, she was in a reasonable state of health, eupneic, breathing without effort, pale mucosa and diminished breath sounds in the lower half of the left hemithorax.

INVESTIGATIONS

The thoracic radiography (figure 1) showed an oval lesion, with regular borders occupying the left lower hemithorax and adjacent parenchyma consolidation. Laboratory tests revealed a microcytic anaemia (haemoglobin (Hb) 7.6 g/dl, Htc 24.9%, mean corpuscular value 60.1 fl, mean corpuscular Hb 18.4 pg), 10 700/μl leucocytes with 65.2% neutrophils, C reactive protein 7.37 mg/dl and an elevated erythrocyte sedimentation rate (100 mm/h), coagulation tests with elevated prothrombin and partial thromboplastin time. The tuberculosis skin test (TST) was anergic and the search for acid-fast bacilli (AFB) in sputum was negative (three separate samples). Endovenous antibiotic therapy was started.

The thoracic CT (figure 2) scan showed a large size (11×10×8.5 cm) cystic lesion with a double membrane without calcifications, in the lower half of the left hemithorax, compressing the left lower lobe and causing right deviation of the heart. The imagiological characteristics (cystic lesion and double membrane) were highly

Figure 1 Thoracic radiography at admission: presence of an oval lesion occupying the lower half of the left hemithorax.

suggestive of hydatid cyst and albendazol (15 mg/kg/day) was added to the therapy.

Immunoglobulin E was normal for the age group and all serology studies were negative (including echinococcus antibodies). No similar lesions were found in other organs (including the central nervous system and liver).

She had no fever from day 7 of admission, but tiredness and a mild left toracalgia remained, without breathing distress. The laboratory tests showed a progressive reduction of infection parameters.

At day 15 she repeated TST, which was positive with a 15 mm induration, suggesting that the pulmonary lesion could be a manifestation of the tuberculosis disease. To confirm this result an interferon- γ released assay (IGRA) was performed.

TREATMENT

Due to the dimensions of the lesions, which was compressing of the left lower pulmonary lobe, exeresis of pulmonary mass was performed by thoracotomy. The histological exam performed during surgery revealed aspects of an active granulomatosis necrotising pleuritis. After surgery, the IGRA result was known and it was positive.

The anatomic-pathologic exam confirmed the diagnosis of tuberculoma with identification of AFB (figure 3) and isolation of *M. tuberculosis* in the intralesion aspirate. TB

treatment (TBT) was established with isoniazid (10 mg/kg/day), rifampicin (15 mg/kg/day), pirazinamide (30 mg/kg/day) and etambutol (20 mg/kg/day). The AFB identified was sensitive to the treatment regimen prescribed.

She had daily chest physiotherapy.

Nine months of anti-TBT was completed with a favourable clinical evolution.

OUTCOME AND FOLLOW-UP

At present, a year after the diagnosis, she is asymptomatic with global expansion of the left lung expanded and the laboratory tests are normal for age group. She was tested for primary and secondary immunodeficiency but investigation was negative. Her close familiar contacts were screened for TB-infection but the results were negative and no index-case was found.

DISCUSSION

In this case, mainly due to its imagiological characteristics and an anergic TST, echinococcosis was the first diagnostic hypothesis. In pulmonary echinococcosis, only 30% of cases have a positive specific serology and lesions are similar to the one found in our patient; a cystic lesion with double membrane. The laboratory results were also compatible with a chronic inflammatory process, which occurs both in tuberculosis and echinococcosis. It is for this reason that the antiparasitic treatment was started.

The surgery was performed due to the compression effect of the pulmonary lesion but also to confirm the diagnosis. TB is a high prevalent and potentially serious disease, especially in certain age groups. It is crucial to investigate the tuberculosis in pulmonary lesions in patients living in or travelling from endemic areas.

A tuberculoma, although a rare pulmonary tuberculosis presentation, is one of the most common benign pulmonary nodules. It represents 5–24% of resected solitary pulmonary nodules and its size varies from 1 to 10 cm in diameter.³ It is a cavity or calcification lesion and its edges are usually smooth and sharp. Their usual location is in the upper lobes.

The diagnosis of pulmonary tuberculoma remains a challenge not only because it usually requires an invasive procedure such as percutaneous needle aspiration or open thoracotomy but also because it can be accompanied by malignant lesions.^{3 4}

Treatment is based on anti-TB medication, generally complemented with surgery due to the lesion's dimensions.^{4 5} The response to medical treatment alone is

Figure 2 Thoracic CT with giant cyst lesion compressing the left lower lobe.

Figure 3 Anatomic-pathological exam; left-piece with an epithelioid granuloma with Langerhans cells; right a bacilli identified.

strongly related to the nodule dimensions and number; in larger nodules the use of isolated anti-TB medication rarely causes reduction of a significant size and in most cases there is a need to combine medical treatment with surgery (total resection).^{5 6} In our case, the treatment was started only after surgery and the confirmation of the diagnosis.

In some cases of atypical presentation of TB, as in our case, the combination of surgery with medical treatment is the best approach. The benefits of resection include conclusive differential diagnosis, determination of future therapeutic strategies and reduction of the dosage and duration of anti-TBT.

Learning points/take home messages

- ▶ We present a case of an atypical form of presentation of tuberculosis.
- ▶ Solitary lung lesions can be secondary to a wide range of different causes; an accurate patient history and clinical investigation is needed to achieve a correct diagnosis.
- ▶ The final diagnosis was reached after surgical resection of the mass revealing a tuberculoma.
- ▶ In this case, the surgical approach combined with medical treatment was crucial for the clinical recovery and cure.

Competing interests None.

Patient consent Obtained.

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