allel in the respiratory sample, but in the literature it appears as the only microorganism isolated in culture that causes the disease. B. hinii also appears as a causal agent of bacteraemia in three cases described in the literature, with immunosuppression the common factor in two of them: one patient with myelodysplastic syndrome and another patient with HIV. Similarly, B. hinii also appeared as a causal agent of chronic cholangitis in a transplant patient on immunosuppressant treatment, so it is considered that it may have a potentially pathogenic role in immunocompromised persons. There were no findings in our patient to suggest immunosuppression. As in some previous publications, our patient had no known avian exposure, suggesting that the organism was obtained from another unidentified source. B. hinii is usually resistant to, or has intermediate resistance to ampicillin, cefuroxime, ceftriaxone, cefotaxime, ciprofloxacin and tobramycin, and is sensitive to imipenem, meropenem, gentamicin, amikacin and trimetoprim–sulfamethoxazol, which was similar to the antimicrobial sensitivity of our isolate, except as regards trimetoprim–sulfamethoxazol. Molecular identification using MALDI–TOF–MS and 16S sequencing, as was performed here, provides the correct microbiological diagnosis. The use of rapid techniques that increase the reliability and speed of identification of this microorganism could lead to clarifying its role as a coloniser and human pathogen.

References


PS are very rare. For Kohler, PS was first described by Rokitanski and Rektorzik in 1861, but it was not until 1946 when Pryce made it known as a clinical entity. In 1972, Felson et al. reported a case of bilateral PS confirmed by pathological examination. In 1977, Karp et al. described another case in a 13-year-old girl who presented a mass in the LLL, discovered after a chest radiograph. CT scanning and pulmonary arteriography were carried out, showing a left intralobar PS, already suspected, and a right PS that was not previously observed. A few cases have been published since then, similar in that they concern paediatric or adolescent patients. In 2009, Yamamura et al. explained the surgical procedure performed on a 44-year-old patient who presented bilateral intra- and extralobar PS. Wei and Li analysed 2625 cases of PS in 2011, among which only 3 were bilateral and 2 of these intralobar. The case presented is doubly unique in that this is an older patient with bilateral intralobar PS. He presented recurrent pneumonia with a changing aspect in the LLL, in which diagnostic tests were inconclusive. The possi-

Fig. 1. CT angiography. The feeding arteries of both sequestrations, subsidiaries of the thoracic artery, can be seen.
Obliterating Bronchiolitis in a Patient Treated With d-Penicillamine

Bronquiolitis obliterante en paciente tratado con d-penicilamina

To the Editor,

Various side effects during long-term penicillamine treatment have been described, especially dermatological reactions, such as exfoliative dermatitis, rash and alopecia. Other adverse events, such as vasculitis and thyroiditis, have also been reported. Respiratory effects have been documented, albeit less frequently, including cases of asthma, pulmonary fibrosis, interstitial pneumonitis and bronchiolitis obliterans.

We present the case of a male patient, 47 years of age, non-smoker, with a clinical history of hypertensive medical therapy, dyslipidemia on treatment with statins, Graves–Basedow disease treated 8 years ago with radioiodine (currently receiving replacement therapy) and Wilson’s disease diagnosed 20 years ago, initially treated with zinc and for the past 2 years with penicillamine. He had a work history as a salesman, with no contact with toxic substances or poultry. The patient consulted due to dyspnea on exertion. An initial lung function study was performed which showed severe airflow limitation, so treatment with inhaled corticosteroids was initiated. A computed tomography (CT) scan of the chest was requested, along with complete lung function testing, revealing severe irreversible obstructive changes with air trapping and reduced carbon monoxide diffusing capacity (DLCO). Discrete bronchiectasis and diffuse bronchiolitis were observed on the CT scan (Fig. 1). A sweat test to rule out cystic fibrosis was negative and an immunoglobulin study was normal. During the follow-up visit, respiratory failure was observed, so the decision was taken to admit the patient for further tests. On physical examination, oxygen saturation (breathing room air) was remarkable due to pulse oximetry of 88% without signs of labored breathing and normal pulmonary auscultation. The rest of the physical examination was normal. General clinical laboratory tests were unchanged and the chest CT was the same as before. In view of suspected drug-induced bronchiolitis, a biopsy was obtained from the lingula by anterior thoracotomy. After an uneventful post-operative period, the patient was discharged with domiciliary oxygen therapy. The final diagnosis from the pathology study was follicular bronchiolitis associated with constrictive bronchiolitis obliterans.

The decision was taken to discontinue penicillamine treatment and start treatment with bronchodilators and corticosteroids (methylprednisolone 40 mg/day), and the patient was referred to a reference center for evaluation for lung transplantation. In May 2012, a double-lung transplant was successfully performed.

Bronchiolitis obliterans is a non-specific disorder of the small airways (<2 mm in diameter). Its clinical presentation is very non-specific (cough and progressive dyspnea). Physical examination is unremarkable and signs of hyperinflation, prolonged expiration and non-specific breathing noises such as rhonchi, crackles or wheezing may be observed. A plain chest X-ray may be normal or show signs of air trapping. Additional information can be obtained from a high resolution CT scan during inspiration and expiration. Distinctive radiological signs in the expiration slices have been described on the CT scan which could guide diagnosis, whether direct (bronchiolar wall thickening with the typical “tree-in-bud” pattern, bronchiolitis and centrilobular nodules) or indirect (subsegmental atelectasis and signs of air trapping). This syndrome can be classified by etiology or pathology. From an etiological point of view, idiopathic cases must be distinguished from those caused by secondary factors. These latter cases are usually acute, the causative agent (primarily drugs or toxic substances) is known, and there is a good response to bronchodilator treatment. The following histopathological classifications have been described: constrictive, proliferative and follicular bronchiolitis, central interstitial fibrosis and diffuse panbronchiolitis. Bronchiolitis can be diagnosed from the clinical signs and symptoms and appropriate complementary tests, but the definitive diagnosis is obtained from lung biopsy. Treatment is based on corticosteroid therapy, bronchodilators, macrolide antibiotic treatment and in the case of rapid progression, lung transplant.

Our patient presented a case of constrictive bronchiolitis obliterans which has an uncommon histological pattern, characterized

References


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