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Non-tumoral metastatic lesions as debut of primary biliary cholangitis and concomitant sarcoidosis

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Dear Editor,

Sarcoidosis has a special involvement at the pulmonary level in 90% of cases, with granulomatous expression in any organ system including the skin, nervous, digestive,

ocular or cardiac systems[1] and remains a diagnostic challenge[2-3].

We report the case of a 51-year-old patient with no relevant personal or family history who consulted for frank haemoptysis, with computed tomography (CT) showing the presence of right pulmonary haemorrhage, multiple adenopathies, and right lower bronchial occlusion.

The bronchial histological study was not conclusive for tumor lesions. A new CT scan was performed, revealing images suggestive of a hepatosplenic metastatic neoplastic process, lymphadenopathy and likely a primary right lower hilar lesion.

Within the differential diagnosis there were ruled out the coexistence of lymphoproliferative processes, storage diseases or diseases of uncertain etiology such as IgG4 syndrome, mycobacterial infections, myeloma and other processes with systemic expression. However, from the analytical point of view were highlighted cholestasis with mixed hyperbilirubinemia, positive antinuclear antibodies (ANA) titer 1/160, positive cytoplasmic pattern and antimitochondrial antibodies (M2), and elevated angiotensin-converting enzyme (ACE).

Given the progression of the cholestasis pattern, hepatic magnetic resonance imaging (MRI) was performed. No relevant findings were revealed. A diffuse parenchymal liver biopsy was carried out whose histology findings were found compatible with chronic hepatitis of likely autoimmune aetiology in the exacerbation phase with detection of granulomas in the portal space (Figure 1. A). Positron electron tomography (PET) revealed multiple hypermetabolic mediastinal lymph nodes in a lambda pattern suggestive of systemic granulomatous disease such as sarcoidosis, and multiple hypermetabolic bone foci, especially in the iliac and sacral bones (Figure 1. B).

A diagnosis of chronic diffuse liver disease was established due to primary biliary cholangitis in an exacerbation phase associated with sarcoidosis, the latter also justifying

the lung and bone involvement.

The debut of sarcoidosis with hemoptysis, hepatic and axial bone involvement simulating metastatic lesions in these organ locations is exceptional. The association of primary biliary cholangitis with sarcoidosis has a high-incidence rate, and some studies even recommend its screening in patients who have already been diagnosed with sarcoidosis[3]. Regarding the clinical presentation in our patient, there are few cases described in the literature, where hemoptysis associated with sarcoidosis[4] is observed, with axial bone involvement and the presence of sarcoid lesions simulating metastasis being exceptional[5], as in this case. This is a diagnostic challenge and involves complex management that requires multidisciplinary collaboration to minimise progression to advanced stages.

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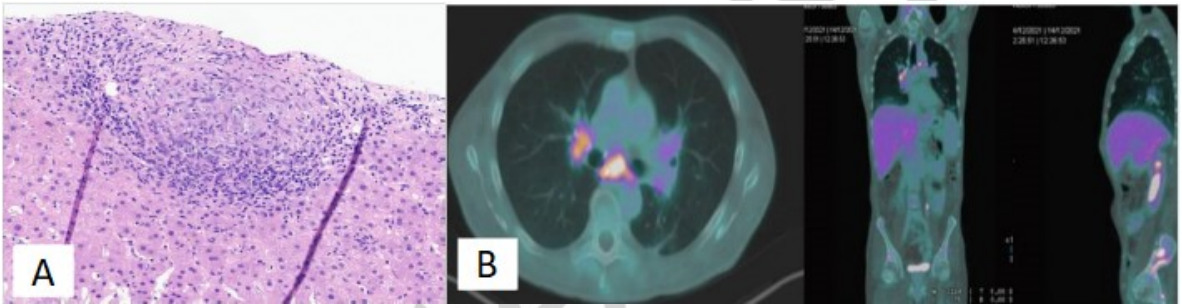
FIGURE 1



A) Liver biopsy with granuloma in the portal tract (haematoxilin-eosin stain).

B) Metabolic hyperuptake in PET-CT images