Necrotizing pseudotumoral hepatic brucelloma: Imaging-pathologic correlation

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KEYWORDS
Brucelloma; Liver abscess; Calcification

Observation

A 62-year old Algerian patient with no prior medical history was admitted to the emergency unit for upper right quadrant pain after deterioration of his general condition for 2 months. Clinical examination was normal. Biological tests showed elevated C-reactive protein without hyperleucocytosis. Liver tests showed moderate cholestasis without jaundice or associated cytolysis. Liver ultrasound showed a large heterogeneous 11 cm mass in the right liver with a central macrocalcification. Abdominopelvic CT examination showed a hypodense mass with irregular borders and a calcification in the centre, strongly suggesting an infectious lesion (Fig. 1). Liver MRI (Fig. 2) showed a multilocular lesion with numerous fluid signal compartments and restricted diffusion. Microbiological agglutination tests (Wright and Rose Bengal reaction) were positive and suggested brucellosis. Fine needle ultrasound-guided biopsy of the lesion revealed pus that was shown to be due to Gram-negative bacteria that was further confirmed as Brucella melitensis after 5 days of incubation. A Brucella liver abscess was
caseous necrosis surrounded by a granuloma composed of histiocyte cells arranged in palisade (Fig. 3). Several weeks later, despite continued antibiotics, there was still no improvement. The patient’s clinical condition continued to worsen and postoperative liver failure developed, then hepatorenal syndrome, so that emergency liver transplantation was decided. Histological assessment of the explanted liver showed severe sepsis on long-term cholestasis with ‘‘young’’ F2 fibrosis according to the Metavir score and diffuse nodular regenerative hyperplasia. The early post-transplantation course was associated with numerous infectious complications, such as recurrent pneumonopathies. Twelve months after transplantation, the patient was in good general condition, and serology for brucellosis was negative on PCR.

**Discussion**

Brucellosis is a cosmopolitan anthropozoonosis whose incidence has markedly decreased in Northern Europe, but which is still frequent in the Mediterranean basin. It is due to the Gram-negative bacteria (*B. melitensis*). Human contamination is direct by cutaneous-mucosal route from animal reservoirs or indirect by contamination from unpasteurized milk products. The germ is disseminated by the lymphatic system or blood vessels to reach the cells of the reticuloendothelial system. Primary infection is characterized by a long lasting fever, multiple adenopathies and hepatosplenomegaly. The usual biological tests do not confirm the disease, and serodiagnostic tests are not specific. The

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**Figure 1.** CT scan in the axial plane during the portal phase shows hypoattenuating. Multilocular lesion with irregular borders. Round macrocalcification is visible inside the lesion.

**Figure 2.** Liver MRI, Axial T1 (a), T2 SPAIR (b), diffusion b800 (c), ADC mapping (d), contrast-enhanced 3D T1-EG during the arterial phase (e) and delayed phase 3 min (f). Multilocular lesion with a hypointense signal of contents on T1 and hyperintense signal on T2 images, liquid type, restricted diffusion (ADC = 0.8 × 10⁻³ mm²/s). Thick hyperintense walls on T2-weighted images with an increased ADC coefficient, with contrast enhancement during arterial and delayed phases. Intense enhancement of the adjacent liver during the arterial phase. Central calcification, which is seen as a low signal intensity lesion on all sequences.
diagnosis is confirmed by Brucella culture or hemoculture during the acute phase, or organ tissue samples [1].

The development of secondary locations, in particular in the liver, are characteristic of the focal complications of brucellosis during the subacute or chronic stage. The liver is one of the prefered targets of Brucella, because of its intracellular tropism parasites Kupffer cells. Liver involvement is always granulomatous, but can have two types of anatomoclinical presentations. This most frequent is a generally asymptomatic hepatic granulomatosis that includes lymphohystiomonic granulomas which usually lack giant cells. The second type, which corresponds to our case study, is a pseudotumoral necrotizing granulomatosis, commonly known as bruceloma, which is very rare, with only thirty cases or so in the literature. Bruceloma is secondary to an undiagnosed liver brucellosis. It is due to the fusion of granulomatous lesions resulting in the development of caseous necrosis and leading to a true abscess. The pathologic assessment shows large areas of focal caseous necrosis surrounded by an epitheloid granuloma arranged in a palisade. Reports of fibrosis of the liver parenchyma have also been described in the literature, although the pathogenic role of brucellosis in the development liver fibrosis and the progression to cirrhosis is still controversial [2].

Imaging plays a fundamental role in identifying the liver lesion usually some time after the primary infection and in a clinical context that is generally poor. The morphological features on CT scan are very specific showing a single pseudotumoral lesion, which is often found in the right liver, including compartments of thick liquid with thick walls that are enhanced after injection of a contrast medium and containing a focal, central or marginal calcification [3–5]. This intrasosional calcification was nearly always reported in the few published case studies and is considered to be specific for the diagnosis. This focal calcification is large or round. It corresponds to a calcified granuloma and is a sign of chronic disease. Liver MRI is not essential to the diagnosis. It shows elements similar to pyogenic liver abscess with a multilocular lesion including a group of cystic lesions of various sizes with a hypointense signal on T1-weighted images and a hyperintense signal on T2-weighted images [6]. Enhancement of the walls during the arterial phase, persists during the delayed phase as well as enhancement of the adjacent liver parenchyma during the arterial phase because of inflammation. Diffusion-weighted imaging can further confirm the diagnosis of an abscess rather than tumoral necrosis by showing restricted diffusion of the purulent content.

The treatment of bruceloma is systematically based on double antibiotics, which are usually doxycycline combined with rifampin or an aminoglycoside for at least 3 months [1]. Percutaneous radiological drainage should be considered immediately because of the large size of the lesion at diagnosis, with a success rate that varies in the literature [7]. The semi-solid consistency of the lesion is in fact the greatest difficulty of drainage. In case of failure, surgical treatment is indicated. Liver transplantation should be considered only when the progression of the disease is extremely severe.

In conclusion, bruceloma is an extremely rare liver disease. Its diagnosis is based on typical imaging features, isolation of the Brucella germ in culture and histology showing a granuloma.
Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References