A succesfully managed inflammatory pseudotumour of liver without surgery : report of a case

Onder Colakoglu[1], Belkis Unsal[1], Mehmet Haciyanli[2], Mine Tunakan[3], Zafer Buyrac[1], Gazi Yorukoglu[1], Nafi Yazicioglu[1], Hudai Genc[2]

Ataturk Training and Research Hospital, (1) Gastroenterology, (2) Surgery and (3) Pathology Clinics, Izmir-Turkey.

Abstract

The inflammatory pseudotumor (IPT) of the liver is an extremely rare focal lesion of the parenchyma with unknown aetiology. It has the appearance of a malignant tumor but has a benign histology and clinical course. Herein, we report a case of IPT in a 55 year-old woman. She had complained of right upper quadrant pain and subfebrile fever. Imaging studies revealed a heterogeneous, solitary mass in right lobe of liver which was 90 × 81 mm in size. Histopathological examination of ultrasonography-guided true-cut biopsy demonstrated IPT. After the initiation of the antibiotic and nonsteroidal anti-inflammatory drugs (NSAID), the size of hepatic pseudotumor decreased (20 × 20 mm) and then completely disappeared. To rule out a malignancy and to reach the diagnosis of IPT, biopsy from the mass is mandatory. After the diagnosis, IPT gave an excellent response to short course of antibiotic and NSAID therapy. To prevent unnecessary resective liver surgery, IPT should be kept in mind in the differential diagnosis of liver mass which can be successfully managed medically. (Acta gastroenterol. belg., **2005**, 68, **382-384**).

Key words : inflammatory, pseudotumor, liver.

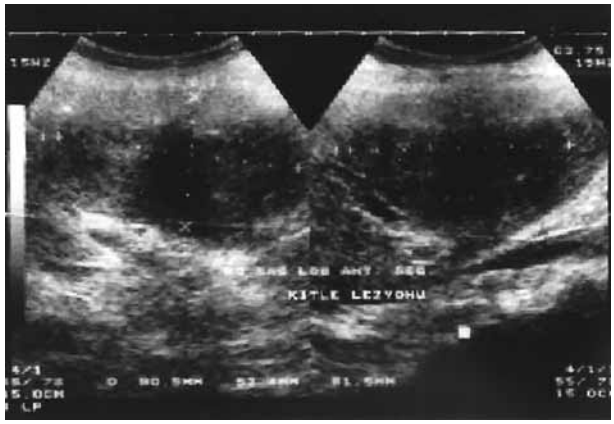

Fig. 1. — The lesion was located in the right lobe of liver anterior portion. In sonographic appearance of it was hypoechoic and heterogene. The mass had cystic and solitary components.

Introduction

Inflammatory pseudotumor of the liver (IPT) is one of the rare mass lesions of the liver. Since the clinical presentation and the morphological appearance are nonspecific, the diagnosis is often difficult despite the use of sophisticated imaging and laboratory techniques (1). Another major difficulty in diagnosis is that these masses often confuse with primary or metastatic liver malignancies so tissue sample of the mass is essential to reach a final histopathological diagnosis. The diagnosis of IPT is important to avoid unnecessary invasive therapy although the surgical resection has been the preferred treatment for IPT of the liver in reported cases (2). Herein, we report a successfully diagnosed and managed IPT without surgery.

Case report

A 62-year-old woman was admitted to the hospital with the complaints of an abdominal pain in the right upper quadrant and fever. She denied any weight loss or active infection recently. She had type II Diabetes Mellitus.

Her physical examination revealed nothing other than slight tenderness in the right upper abdomen and body temperature of 37.7˚C. Laboratory data included an elevated white blood cell count (12.200/mm^3), blood fasting glucose level (179 gr/dl), and erythrocyte sedimentation rate (58 mm/h). Abdominal ultrasonography (US) was performed and a liver mass was detected. The mass was located in the right lobe of liver and 90 × 81 × 53 mm in size. It was hypoechoic and hypovascular on Doppler US. The mass had mainly solitary but contained some cystic areas (Fig. 1). A stone, 15 mm in-diameter, was incidentally found in the gall bladder.

On magnetic resonance imaging (MRI), the mass was hypointense on T1-weighted images and hyperintense on T2-weighted images and distinctive from surrounding liver parenchyma with relatively regular and prominent boundary features. The central and peripheral portions demonstrate hyperintensity on T2-weighted images (Fig. 2).

She had a further investigation to exclude primary or metastatic liver tumor or hydatic liver disease. Serology for hepatitis and cyst hydatic disease was negative. The urinary and blood cultures were negative for microorganisms. The serum tumor markers (μ-FP, CEA, CA 19-9, CA 125) were within normal limits. Gastrointestinal

Correspondence to : Mehmet Haciyanli, M.D., 215 Sokak 27 / 7 Basin Sitesi, 35360 Izmir-Turkey. E-mail : mhaciyanli@hotmail.com.

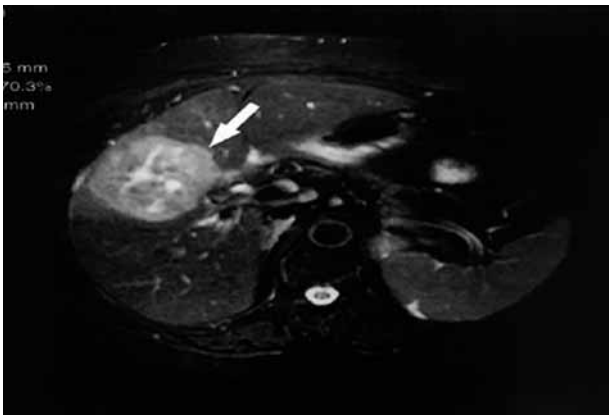

Fig. 2. — The lesion is located in anterior portion of the right lobe on T2-weighted images. The central and peripheral portions demonstrate hyperintensity on T2-weighted images.

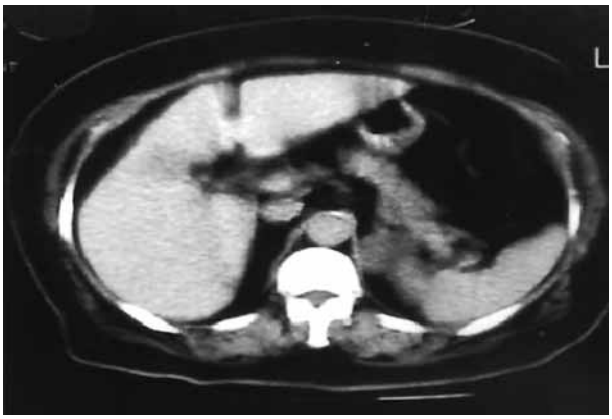

Fig. 4. — The mass in the liver regressed from 90 mm to 20 mm at 20th day.

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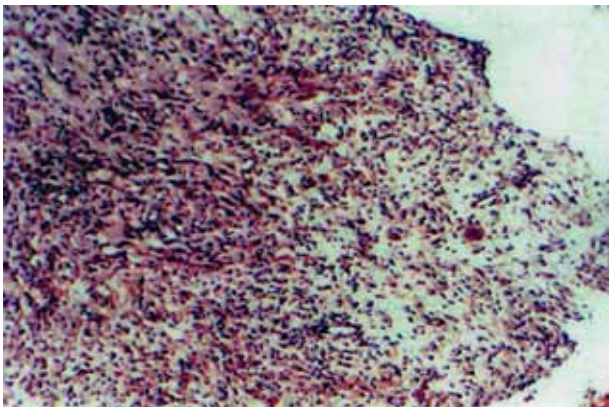

Fig. 3. — 110 × HE. Capillary vascular proliferation, mononuclear cell infiltration and degenerated hepotocytes.

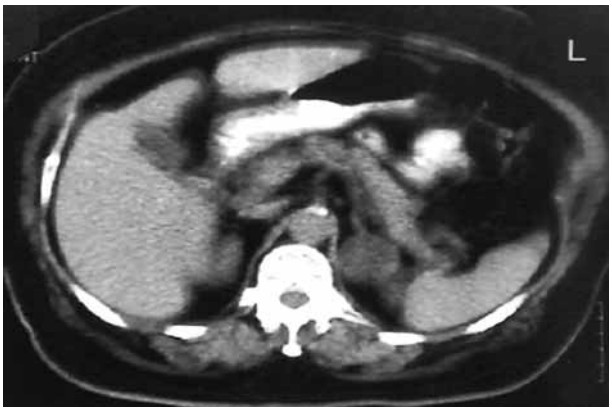

Fig. 5. — The mass completely disappeared on the 40th day

(GI) system endoscopic examination excluded any GI malignancy. Her breast examination and mammography were also normal.

US-guided percutaneous tru-cut biopsy of the mass was performed to reach a final diagnosis. The histological examination of the biopsy specimen revealed mononuclear cell histiocyte-like cells with a granular cytoplasm and a single small nucleus, multi-nuclear cells containing a yellowish bile-like material in a few fields (pseudo-bile duct proliferation), lymphocyte-like cells with undistinguishable cytoplasm which had a single hyperchromatic nucleus, plasma cells and rarely eosinophils (Fig. 3). These histopathological findings were consistent with the "inflammatory pseudotumor" of liver. She had given on an empiric oral antibiotic and NSAID treatment for 30 days. The serial follow-up CT images were obtained on 20th and 40th days of the initiation of treatment. The mass in the liver regressed from 90 mm to 20 mm (Fig. 4) and pain in the right upper quadrant resolved at 20th day. The mass completely disappeared on the 40th day (Fig. 5). She is quite well on control a year after the hospitalisation.

Discussion

IPT is a rare benign tumor of the liver and its aetiology and pathogenesis remain unclear. About a hundred cases of IPT of the liver have been reported in the English literature since its first description in 1953 (2). Synonyms of this condition include inflammatory myofibroblastic tumour, inflammatory myofibrohistiocytic proliferation, and plasma cell granuloma (3). IPT may also arise in a variety of tissues and organs other than liver including lungs, brain, prostate, pancreas and omentum. Although IPT is common among the middle-aged women (range 3-77 years), it can be seen in childhood (1,4).

The most common symptoms related to IPT are fever, epigastric pain and weight loss (5). Leukocytosis, elevated erythrocyte sedimentation rate and serum C-reactive protein levels are usually associated with the condition. A mild elevation in the liver transaminases and alkaline phosphatases may be observed.

Hypoechoic pseudotumours are frequently encountered in the forth segment of liver (6). Some of them are

inflammatory in nature. There are no specific findings on US and CT images. IPT lesions often appear hypoechoic on ultrasonographic images. Most of them are solitary. However these lesions in the liver may occasionally be encountered as cystic structures. Hypovascularity of the lesion on Doppler US urged us to think about a metastatic liver tumor since the primary liver tumours are usually hypervascular. MRI findings of IPT are variable and often reveal a lesion which is hyperintense on T2-weighted images and hypointense on T1-weighted images (7). Differential diagnosis includes hepatocellular carcinoma, focal nodular hyperplasia and other space-occupying lesions in the liver such as metastatic tumours. Because even routine imaging procedures usually fail to differentiate hepatic IPT from liver neoplasms, obtaining a tissue sample for histopathological diagnosis is usually needed (8). Like in this case, according to the radiological features a liver tumor was not ruled out so a tissue diagnosis was mandatory.

Diagnostic accuracy of fine needle aspiration cytology for IPT was low (42%) and cytomorphology is nonspecific (9). If only fine-needle aspirations are performed, a definitive diagnosis may be difficult to obtain because many of the benign entities have some overlapping histological features (10). So it is recommended that US- or CT-guided core biopsies be performed. Koea et al recently reported six patients with IPT of liver and in all cases, diagnosis was made on core biopsy of the liver lesions (accuracy rate was 100%) and all patients were managed non-operatively with complete resolution of the tumours (11). But it should be kept in mind that if a definitive pathological diagnosis still cannot be reached even after core biopsy, a surgical resection may be necessary.

Since IPT is a benign condition, its prognosis is favourable after the diagnosis. Most of the reported cases in the literature had the surgical resection of the mass (2) but few cases had been reported who were treated medically and complete disappearance of the mass occurred (11-16). However the response of IPT to medical treatment also remains unclear and there is no enough data in the literature in respect to which drugs should be used and how long should be the duration of therapy in the medical treatment of the lesion.

In this case we use an antibiotic and an anti-inflammatory drug and a partial response was obtained at the 20th day of the initiation of the therapy and complete resolution of the mass was reached at the 40th day of the initiation of the therapy. Her complaints were disappeared and she is free of symptoms and has normal liver images during the one year follow-up. We report an additional case of hepatic IPT with a favourable outcome under medical treatment (antibiotic and NSAID) without surgical resection and we documented the response to the therapy with both clinical examination and radiological images.

As a result, IPT of liver mimics the malignant lesions of the liver and tissue diagnosis is mandatory to rule out a malignancy and to establish a final diagnosis. Once the diagnosis is reached, prognosis is good, complete resolution of the mass and the symptoms can be reached with the medical treatment.

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