

Which Is The More Advisable Treatment For Recurrent Inflammatory Pseudotumour Of The Liver? A Case Report.

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Abstract

In this report the authors describe the case of a recurrency of a liver pseudotumor previously described from the same authors, characterised by an hepatic mass in a woman aged 62. The infiltrative pattern of the mass at the US scan suggested a malignant hepatic neoplasm. The histologic examination of the needle biopsy of the liver suggested the diagnosis of an inflammatory lesion of the liver having the features of an inflammatory pseudotumour. The diagnostic and therapeutic implications of this rare pathology are here considered.

INTRODUCTION

As previously described in a prior paper by the same authors (1), the inflammatory pseudotumor of the liver is a heterogeneous group of disease, a complex of lesions of unknown aetiology and pathogenesis which occupy space and mimic a neoplasm. This quite rare lesion (lesser few than 140 cases reported in the world literature) can frequently arise in subjects affected by systemic diseases such as rheumatoid arthritis, or in subjects who use drugs.

Asia is the geographical center for this pathology, with prevalence for male gender. Usually this condition onset with dyspeptic symptoms and laboratory findings of increased ESR, increased CRP, leucocytosis, slightly elevated activities of the aminotransferase, increased bilirubin and variable alterations of the electrophoretic pattern.

The inflammatory pseudotumor shows nodular lesions characterised by wide sclero-hyaline bands, sometimes in vorticoïd structures, and an extremely variable inflammatory infiltrate (2).

Sometimes the histologic differential diagnosis may be quite difficult especially in discriminating pathologies such as chronic granulomatous hepatitis, sarcoidosis and tuberculosis affecting the liver (3).

Although rare, recurrences of this pathology are reported. These recurrences may either occur in operated patients or in patients treated by drugs administration.

CASE REPORT

A 62 years old woman was admitted to the General Surgery Department of Policlinico of Bari because of well defined abdominal pains in the right hypocondrium, digestive disorders and moderate elevation of the body temperature (37.4 - 37.6 C°) during the night.

The past medical history showed a serious form of long standing scoliosis, and allergic asthma lasting 15 years. She was also treated with specific drugs for rheumatoid arthritis for three years. Ten years before being hospitalised, she underwent to hysteroadnexectomy, because of an acute metrorrhagia due to uterine leiomyoma, six years later she underwent cholecystectomy for cholelithiasis. Two years ago she underwent to atypical bisegmentectomy of the 5th and 6th hepatic segments because an inflammatory pseudotumor of the liver.

The patient underwent laboratory tests, which revealed increased ESR (1st h =23) and increased hepatic function activities (ALT=63 U/l; AST=100 U/l; -GT=87 U/l; tot. Bil.=1.1 mg/dl). An ultrasound scan of the upper abdomen showed an increase of hepatic volume and the presence of an echographically heterogeneous area measuring about 2.5 cm in diameter in the right lobe of the liver. Colonoscopy and skin tests for tuberculosis were negative.

By using contrast, an abdominal CT scan confirmed the presence, in the residual right liver, of a heterogeneous hypodense expansive mass with a diameter of 4 cm, the dilatation of the extra-hepatic biliary tract, the enlargement

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of the pancreas head, and a “minus” image (reduction of lumen diameter) of the second segment of the duodenum. In addition, several intra-hepatic lesion, very little in diameter, were observed. The kidneys, the spleen and the other intraperitoneal organs appeared normal. The enlargement of the pancreas had an inflammatory origin, probably.

After hospital admission she underwent other clinical tests to evaluate some viral and bacterial markers (HBV, HCV, CMV, HSV1-2, S. tiphy, etc.) and the tumoral markers of the digestive tract (-FP, CEA, TPA, CA19.9), which proved to be negative, excepted S. tiphy.

Under US guidance was performed a needle biopsy of the main lesion. Hepatic tissue and liquefactive material was obtained.

The samples, fixed in 10% buffered neutral formalin, were paraffin embedded and the sections were stained for H/E and Alcian blue/PAS at pH 2.5.

Histologically focal areas of colliquative necrosis were detected, which accounted for the main lesion observed at US and CT scans. Little foci of chronic inflammation with fibrous bands, giant and granulomatous epithelioid cells were found in the surrounding normal tissue. The inflammatory cells were mainly lymphocytes, plasma cells and foamy histiocytes.

However, a lymphoid inflammatory infiltrate of the portal tract and around the bile ducts was observed.

This pattern and the past medical history of the patient are consistent with the hypothesis of a recurrent inflammatory pseudotumor of the liver.

High doses of co-trimoxazole (Bactrim™, 20 mg/kg, per os every 6 hours) were administered for 7 days, until dosage was reduced to 600 mg/qd, per os. The body temperature became normal after 3 days.

After 20 days the patient was discharged from hospital and the treatment was again continued for 25 days.

During the treatment were performed four US scans at the inpatient, and three at the outpatient. Two US scans were performed two months and five months after the end of the treatment.

The imaging study by US, during and after the treatment,

demonstrated a slow and continuous regression of the lesions of the liver. These lesions have completely disappeared after three months from the onset of the therapy

DISCUSSION

The inflammatory pseudotumor of the liver is a condition of great interest, because it maybe easily confused with other pathologies such as granulomatous hepatitis, sarcoidosis, hepatic tuberculosis or hepatocarcinoma.

Though the inflammatory pseudotumor onset as clinically silent hepatitis, it could insidiously mimic a malignant neoplasm (sudden and progressive weight loss, elevated activities of the aminotransferase, increase of CRP, of ESR and leucocytosis) that, if confirmed by the radiodiagnostic exams, may lead to an excessively aggressive therapeutic treatment (4).

Otherwise, the negativity of tumoral markers can lead to the diagnosis of a non-neoplastic condition, which may have relief - in any case - from the surgical removal of the mass that acts as a further irritative stimulus for the surrounding parenchyma, and can cause an extended fibrosis of the organ, which determine a progressive hepatic failure and the death of the patient (5-6). It represents therefore a pathology which is quite difficult to describe clinically, whose nature can be exclusively revealed by histologic study after mass removal. Liver biopsy could be useful for diagnosis in compliant patients, avoiding major surgery.

According to some studies (7) this pathology is due to bacteria from foods or chronic appendicitis or cholecystitis whose would seed in the hepatic parenchyma through the bloodstream of the portal vein, subsequently eliciting inflammatory lesions with obliterating phlebitis and granulomatous inflammation. Some Authors support the hypothesis of an EBV infection.

Optimum management of this disease has not yet been standardized. The majority of patients are treated by hepatic resection, although spontaneous regression has also been described.

According to the world literature, the best therapeutic option was been liver resection in 93 cases, medical therapy in 16 cases and a “watchful waiting” in the other cases. In fact, in patients whose underwent to surgical liver resection were reported 6 deaths after 72 hours and 17 after 60 months from the surgical intervention. The patients of the other two groups had high rate of mortality (totally 8 deaths due to

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hepatic failure) and morbidity (persistence and/or progression of the disease).

The reported case, considering that at follow up four years after surgery the patient is healthy, confirms the utility and necessity of surgery in individuals presenting large-sized lesions (more than 5 cm in diameter) and suggests the usefulness of antibiotic therapy in patients with smaller lesions (lesser than 5 cm in diameter) and histology confirmation of the diagnosis.

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