

Inflammatory Pseudotumor of the Liver: A Therapeutic Dilemma

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Key words: inflammatory pseudotumor, liver, treatment

IMAJ 2005;7:472–473

Inflammatory pseudotumor, first described in the liver by Pack and Baker in 1953, is an uncommon benign lesion occurring in various organs. The hepatic lesions are mostly focal and affect predominantly males. Patients have varying degrees of non-specific symptoms such as fever, abdominal pain, weight loss and malaise [1]. There are no specific laboratory or radiologic findings, and in most cases inflammatory pseudotumor is diagnosed after surgical removal of the lesion, which is usually malignant preoperatively.

Since inflammatory pseudotumor is thought to be a benign lesion and in view of reports of spontaneous regression under conservative treatment, the treatment is controversial when the diagnosis is made before surgery. We present a case of progression of the pseudotumor under conservative treatment, which emphasizes the need for early surgical removal despite the fact that this is a benign lesion.

Patient Description

A 59 year old woman was referred to our hospital for investigation of low grade fever, right upper quadrant abdominal discomfort, weight loss of 5 kg, and night sweats for 5 weeks. Her medical history included bronchial asthma treated with inhaled salbutamol on an as-needed basis, multinodular non-toxic goiter, and endometrial atypia that was treated several years earlier with hysterectomy and bilateral salpingoophorectomy. Physical examination revealed no pathologic findings except for temperature of 38°C. Blood analysis demonstrated: erythrocyte sedimentation rate 120 mm/hour, C-reactive

protein 159 mg/L (reference range 0–0.5), hemoglobin 9.8 g/dl, mean corpuscular volume 79.8 fl, alkaline phosphatase 148 U/L (reference range 70–115), albumin 3.3 g/dl (reference range 3.5–5.5), globulin 4.4 g/dl, fibrinogen 8,050 mg/L (reference range 2,000–4,000), iron 190 µg/L, and transferrin 1,870 mg/L (reference range 2,000–3,600). Prothrombin time, partial thromboplastin time, glucose, electrolytes, autoantibodies, serology for viral hepatitis, and tumoral markers (alpha-fetoprotein, carcinoembryonic antigen 15-3, CA 19-9) were all normal.

Abdominal ultrasonography revealed a normal-sized liver with a 5 cm hypoechoic lesion surrounded by a hyperechoic ring. The other abdominal organs were normal. Computed tomography of the upper abdomen showed a 5 cm mass in the right lobe of the liver. Injection of contrast material caused intense enhancement of the lesion, with a peripheral

non-enhanced halo surrounding the lesion [Figure].

Liver scan with labeled erythrocytes showed no evidence of hemangioma. Biopsy of the liver mass demonstrated replacement of the hepatic parenchyma by non-specific inflammatory cells, mainly plasma cells, lymphocytes and young fibroblasts. Specific stains showed polyclonality for lambda and delta chains and hypervascularization. These features were diagnostic of inflammatory pseudotumor.

The patient was treated initially with ofloxacin and metronidazole for 4 weeks. Since her clinical condition did not improve, 60 mg prednisone once daily was added and resulted in temporary amelioration of the symptoms and a reduction of the lesion size to 4 cm. Two months later, however, the patient's symptoms worsened, and repeated ultrasonography and CT showed lesion enlargement to 5.5 cm. We recommended excision of the tumor mass, and a right hepatic lobectomy was performed. The pathologic findings of the excised tumor confirmed the diagnosis of inflammatory pseudotumor. On follow-up, she was asymptomatic with no laboratory evidence of inflammatory disease.

Comment

The treatment of inflammatory pseudotumor is controversial. It is known that some inflammatory pseudotumors actually regress and completely resolve without treatment [1]. However, inflammatory pseudotumors can recur or metastasize, leading to death in some patients [2]. Since most documented patients underwent surgery due to



CT of the upper abdomen showing the liver lesion. On post-intravenous administration of contrast material, an intense enhancement is noted, with a peripheral non-enhanced halo surrounding the lesion.

presumed diagnosis of a malignant lesion, there are few reports of patients treated conservatively with antibiotics, non-steroidal anti-inflammatory agents or steroids, or of patients who were even cured without treatment [3]. In a recent publication, a series of six patients with inflammatory pseudotumor of the liver showed complete resolution of the tumor under conservative treatment [4]. Regression generally occurred within 6 months [3], though it was occasionally slower, up to 30 months. In one case, the lesion showed no change but the patient became asymptomatic. Success is not always achieved by conservative therapy and immediate surgical strategy is sometimes the best option [5].

Pathogenesis of this hepatic lesion is still unknown. Microorganisms that gain access through the portal system have been proposed as a possible etiology, probably by producing an inflammatory reaction within the liver parenchyma [3]. Another possibility is hepatic bleeding and necrosis [1]. A cytogenetic analysis of the inflammatory pseudotumor specimens demonstrated chromosomal abnormality and monoclonality of the cells, suggesting that pseudotumor is not a

tissue reaction to inflammation. Biselli et al. [2] found aneuploidy (hyperploidy) in five of nine liver specimens of pediatric patients with liver inflammatory pseudotumor. Three of them had local recurrence, and two died of the disease. Chromosomal abnormalities were demonstrated as well, including translocations and partial chromosomal deletion. No correlation was found between the histopathologic appearance and the incidence of recurrence or locally aggressive pseudotumor. Our patient was treated with antibiotic and steroids without success, and the lesion was excised surgically.

Inflammatory pseudotumors are biologically heterogeneous and include a wide range of lesions, from those that may regress to those that may recur or metastasize, and there are no accurate predicted prognostic factors. Based on our experience with this patient, the relevant literature, and the lack of useful markers predicting the progression of the lesion, we believe that a rapid and accurate preliminary diagnosis is crucial to avoid unnecessary major surgery. However, surgical excision of liver inflammatory pseudotumor should be considered as a

therapeutic option early in the treatment course.

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