Case Report

Inflammatory Myofibroblastic Tumor of Abdomen: Computerized Tomographic (CT) and Pathological Findings

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Inflammatory Myofibroblastic Tumor (IMT) is the recent name of Inflammatory pseudotumor which was in intermediate group of fibrous-origin tumor. The authors retrospectively reviewed such cases in Ramathibodi Hospital from January 2001 to December 2005. There were 115 cases of fibrous-origin tumor, which was IMT in 17 cases. Nine cases occurred in the abdomen and only 3 of these had complete computerized tomographic (CT) imaging. One was hypodense liver mass with thick rim enhancement. Another one in the liver presented as a liver abscess which appeared as multiloculated hypodense mass with enhanced septum. The third case was a large malignant-looking retroperitoneal mass and having a small accompanying hepatic lesion which rapidly grew in the follow up study at nine months.

IMT in the abdomen was scanty. The diagnosis was done with difficulty because of different signs and symptoms such as fever and palpable abdominal mass. The laboratory findings were nonspecific or within normal limits. Tissue biopsy was the way of definite diagnosis. We reported 3 cases of abdominal IMT with variable imaging findings that may lead to inappropriate treatment. Recognization of such findings will help achieve correct diagnosis.

Keywords: Inflammatory myofibroblastic tumor, Inflammatory pseudotumor, Liver, Retroperitoneum, Computerized tomography

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Fibrous-origin tumor is one category of soft tissue tumor that is composed entirely of fibrocytes, fibroblasts, myofibroblasts, or mixtures of these three components. It was classified into benign, intermediate, and malignant tumors histologically⁽¹⁾. The tumors in the intermediate group may behave aggressively mimicking malignancy, causing confusion in the imaging studies. The majority of fibrous-origin tumors were found in the extremities (60%), in the trunk and retroperitoneum $(30\%)^{(2)}$. In the abdomen, a painless mass is the most common sign and symptom and prompts the patients to request investigation.

Inflammatory myofibroblastic tumor (IMT) is a relatively new histopathologic term for an entity previously known as inflammatory pseudotumor, which is in the intermediate group of fibrous-origin tumors. It is a rare pseudosarcomatous inflammatory lesion that occurs in the soft tissue of young adults^(3,4). The exact pathogenesis of IMTs remains unclear, although various allergic, immunologic, and infectious mechanisms have been postulated⁽⁵⁾. The lesion manifests macroscopically as a tumorlike mass, and a mixed inflammatory infiltrate is revealed at microscopic inspection⁽⁶⁾. Definite diagnosis is usually based on histological and cytological examinations.

In the present study, the authors reported 3 cases of abdominal IMT in Ramathibodi Hospital, concerning the CT and pathological findings.

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Material and Method

The protocol of the present study was approved by the Internal Review Board (IRB) and Ethics Committee (EC) of Faculty of Medicine, Ramathibodi Hospital, Mahidol University, Bangkok, Thailand.

Patients

The cases of fibrous-origin tumor were retrieved from surgical pathology reports in the archives of the Department of Pathology, Ramathibodi Hospital, between January 2001 and December 2005. The reports were classified according to classification of the fibrous-origin tumor of the Armed Forces Institute of Pathology (AFIP) in 2001⁽¹⁾.

CT scan

The CT scans were performed by using either the 4-slice Multidetector CT (LightSpeed plus; GE Medical System, Milwaukee, WI, USA) or the 64-slice Multidetector CT (Somatom Sensation Cardiac 64; Siemens, Germany).

A board certified radiologist and a resident in Radiology reviewed all the images using e-film workstation, version 2.1 (Merge E-film Medical System, USA) on the Window XP operating system (Microsoft Cooperation, USA), with consensus agreement.

Pathology

The reports and microscopic slides of the selected cases were reviewed by a senior musculoskeletal pathologist who was an expert in soft tissue tumors in Ramathibodi Hospital.

Results

There were 115 cases of fibrous-origin tumor in the surgical pathology report of the Department of Pathology, which consisted of intermediate and malignant groups. The intermediate group was desmoid and inflammatory pseudotumor (the name at the time of report, newly named inflammatory myofibroblastic tumor [IMT]). The desmoid tumors (n = 63) were found in the head and neck (n = 2), extremities (n = 32), and trunk and abdomen (n = 29). The IMTs (n = 17) were found in the head and neck (n = 5), abdomen (n = 9), and extremities (n = 3).

Among nine cases of abdominal IMTs, only three cases had imaging studies and pathology slides available for review. There were two liver masses and one retroperitoneal lesion

Case 1

A 40-year-old male with a history of acute cholecystitis and CBD stone, status post cholecystectomy and explored CBD for 6 months. He presented with a high-grade fever with chill and jaundice. The symptoms and laboratory data suggested active inflammation of the liver.

The CT images revealed a solitary lesion in the right hepatic lobe, which measured about 12.5 x 9.6 x 12.5 cm in AP, transverse and vertical dimensions, respectively. The mass was large, multiloculated and hypodense, involved hepatic segment 6 and 8. After administration of the contrast medium, the lesion showed enhancement of the internal septae (Fig. 1). Drainage was performed followed by antibiotics. The second CT scan at 1 month revealed decreased size of



Fig. 1 Case 1; A 40-year-old male with hepatic IMT: Nonenhanced (A) and portovenous-phase enhanced (B) axial CT scans of the liver reveals a large multiloculated hypodense mass in the right hepatic lobe with enhancing septae

the lesion to be 6.0 x 5.7 x 5.7 cm. The antibiotics were continued. The third CT scan at 2 months revealed slightly decreased size of the lesion, measured 5.4 x 5.3 x 4.2 cm. The patient, therefore, underwent wedge resection. The gross specimen showed a single illdefined, firm, inhomogeneous yellowish-tan mass. The histologic study revealed that the lesion was made up of plump spindle cells running in interlacing bundles and occasionally storiform pattern intermingling with various types of inflammatory cells ranging from lymphocytes, plasma cells, eosinophils, and polymorphonuclear leucocytes (Fig. 2). The spindle cells showed bland, vesicular, round to oval shaped nuclei and possessed abundant eosinophilic cytoplasms imparting myofibroblasts. Mitosis and atypia were not recognized.

When correlating the CT features with the histological findings, the central hypoattenuation in the mass corresponded to the abundant thick bands of collagen, and the enhancing septae corresponded to reactive area. There was no evidence of purulent material or acute inflammatory cells such as neutrophils within the lesion.

The patient's last follow up was at 1 year after the first CT scan. He was clinically asymptomatic with normal liver function test.

Case 2

A 68-year-old male presented with low-grade fever. The laboratory investigation revealed leucocytosis and suggested active inflammation of the liver. The serum level of α -fetoprotein, carcinoembryonic antigen (CEA), CA19-9, and *E. Histolitica* antibody were within normal range.

The CT scan of the upper abdomen revealed a round, well-defined hypoattenuating mass with central fluid density at hepatic segment 8, with a thick rim of enhancement after administration of the contrast medium (Fig.3). The lesion measured about 5.3 x 4.7 x 3.6 cm, and it was reported as a thick wall abscess or tumor with central necrosis. The histology obtained from fine needle aspiration revealed inflammatory pseudotumor. The histologic findings resembled that shown in Fig. 2, consisted of chronic inflammatory process with histiocyte, plasma cell, and lymphocyte infiltration. There was no atypia. Minimal fibrosis was also presented. After appropriate antibiotics, the patient was clinically asymptomatic 1 month after starting treatment. Follow up CT scan at 2 months revealed the decreased size of the lesion to be 3.5 x 2.2 x 1.5 cm.



Fig. 2 Photomicrograph of the lesion showing plump spindle cells of myofibroblasts having vesicular, round to oval nuclei running in interlacing bundles and interspersing with numerous lymphocytes, plasma cells, eosinophils and polymorphonuclear leucocytes (H&E; x200)

The patient's last follow up was at 1 year after starting treatment. Abdominal ultrasound was performed and showed that the mass had reduced in size to be 1.7×1.3 cm.

There was no lymphadenopathy, metastasis, or ascites in two cases of hepatic IMT.

Case 3

A 39-year-old female, known case of HIV infection, presented with a palpable mass at the left upper quadrant of the abdomen with low-grade fever. The laboratory findings were unremarkable. Her sonogram revealed a large heterogeneous retroperitoneal mass, which measured about 14.0 cm in greatest dimension.

The CT scan of the whole abdomen revealed a huge, rather well-defined hypo- to isodense retroperitoneal mass with internal hypodensity and small tiny calcifications. It measured about 8.5 x 10.5 x 13.0 cm in AP, transverse and vertical dimensions, respectively (Fig. 4 A). The mass showed inhomogeneous enhancement after contrast medium injection. The mass was located in the left anterior pararenal space and caused pressure effect to the adjacent structures without demonstrable invasion. There were a few enlarged lymph nodes, subcentimeter in size. No ascites was noted. There was an accompanying 1.0-cm non-enhancing hypodense lesion in the hepatic segment 2 (Fig. 4 B).

She underwent explore laparotomy for total tumor removal with distal pancreatectomy and



Fig. 3 Case 2; A 68-year-old male with hepatic IMT: Nonenhanced (A) and portovenous-phase enhanced (B) axial CT scans of the liver reveals a well-defined hypodense mass in hepatic segment 8 with central fluid density. After administration of contrast medium, there is peripheral thick wall enhancement

splenectomy. Grossly, the specimen consisted of an oval, lobulated, firm, pale-tan mass with attached intact purple spleen and part of the pancreas, weighted 1,100 grams in total. The cut surface showed an encapsulated, rubbery to firm, pale-tan and yellow lesions with scattering orange areas. There was one small necrotic area without hemorrhage. The scattering nodules surrounding the large mass showed homogenous grayish yellow appearance. Microscopically, this lesion consisted of reactive fibrosis with multiple areas of abscess formation seen as accumulation of neutrophils. An area of necrosis was also noted. The immunochemistry was positive only for vimentin. The



Fig. 4 Case 3; A 39-year-old female with retroperitoneal IMT and accompanying lesion in the liver Enhanced axial CT scan of abdomen at level of renal lower pole reveals an ill-defined heterogeneous-enhancing hypodense left-sided retroperitoneal mass (A). Focal spot of hemorrhage, necrosis and calcification are also noted. Enhanced scan at upper level (B) reveals a focal hypodense lesion with minimal enhancement in the hepatic segment 2. Axial CT scans at 9 months after total removal of the tumor (C) reveals recurrence of the retroperitoneal mass. The lesion in hepatic segment 2 is progressively enlarged, with enhancing pattern resembling the retroperitoneal mass. It is likely to be another focus of IMT, but it has not been pathologically proven

pathology report concluded as inflammatory myofibroblastic tumor. The lymph nodes at peripancreatic and splenic hilar areas were reported as metastatic inflammatory myofibroblastic tumor. The pancreas, spleen, and lymph nodes at gastric area were not remarkable.

Fine needle aspiration for the hepatic lesion was performed and revealed necrosis without malignancy.

Follow-up CT scan at 8 months after surgery revealed local tumor recurrence (Fig. 4C). The hepatic lesion also increased in size and showed CT findings resembling the hepatic IMT in Case 1 (Fig. 1). This patient was treated by *chemotherapy*. The CT scan at completion of chemotherapy revealed decreased size of the lesions.

Discussion

Inflammatory myofibroblastic tumor (IMT) is a rare soft tissue lesion in young adults. It was first described in the lung in 1937 by Philips⁽⁷⁾ and later in the liver by Pack and Baker⁽⁸⁾. It has subsequently been documented in the central nervous system, breast, pancreas, spleen, lymph nodes, skin, and soft tissues⁽⁴⁾. Its previous synonyms included xanthogranuloma, fibrous xanthoma, plasma cell granuloma, histiocytoma, pseudolymphoma, and plasmacytoma. IMT could be defined as a localized mass consisting of a fibrous stroma and chronic inflammation, infiltrated with a predominance of plasma cells or histiocytes and an absence of anaplasia. Based on predominant cell type, IMT was classified into xanthogranuloma type, plasma cell granuloma type and hyalinized sclerosing type⁽⁹⁾.

The pathogenesis of IMT is still unclear. Some authors have suggested an infectious origin, but few cases have positive cultures or microorganisms isolated from tissue sections⁽¹⁰⁾. It was suggested that the chronic xanthogranulomatous pattern was a response to previous infectious process. IMT can occur after surgery, trauma, ventriculo-peritoneal shunts, radiotherapy, steroids, and in association with infectious agents such as *Mycobacterium avium intracellulare*, *Cornyebacterium equi*, *Campyrobacter jejuni*, *Bacillus sphaericus*, *Coxiella burnetti*, Ebstein Barr virus, *Escherichia coli*, and Actinomycosis⁽¹⁰⁻¹²⁾. Case 1 in the present study also occurred after surgery (cholecystectomy).

IMT associates with high recurrent rate. Meis and Enzinger^(13,14) have reported metastasis in retroperitoneal IMT. Case 3 in the present study had local tumor recurrence at 9 months after removal of the primary lesion. The accompanying lesion in the left lobe of liver that was enlarged at the time of retroperitoneal recurrence, may be another focus of IMT or being metastatic deposits since the beginning.

Hepatic location is usually solitary. However, they have been reported to be multiple and synchronous, and are in the same lobe or in both lobes of the liver⁽⁴⁾. Radiographic findings of hepatic IMT have a non-specific morphologic appearance^(10,15-17). In the study of Nam and Yoon^(15,16), two different appearances of the hepatic IMT were observed. First, enhancement is broad and ill-defined at the periphery of the mass, while multiple low attenuation or nonenhancement is another feature. In correlation with the histopathologic findings, lower attenuation found in contrast-enhanced CT corresponds to areas of concentrated fibrous tissue. while the hyperattenuating area is corresponding to the predominantly cellular infiltration with foamy histiocytes, plasma cells, and lymphocytes. These images and pathological results corresponded to the findings in this present study.

Retroperitoneal location is rarely found, although some case reports have been described^(10,11,18-23). The patients presented with intraabdominal mass with inflammatory feature. The CT images showed welldefined heterogeneous-enhancing hypo- to isodense retroperitoneal mass with internal hypodensity and small calcifications. Mali⁽¹⁰⁾ reported a homogeneous mass without calcification or contrast enhancement, whereas Case 3 in the present study was inhomogeneous and showed tiny calcification. These might support the concept that CT findings of the IMT were nonspecific. Case 3 in the present study revealed an accompanying lesion in the liver, presenting an identical picture with IMT of the retroperitoneum. The authors believed that it was another focus of IMT. Unfortunately, there was no histology conformation from the liver lesion.

There were 14 case reports of HIV with IMT, located in the spleen, liver, lung, testis, tongue, lymph nodes, bowel, and intraocular region⁽²³⁻³¹⁾. Although the association between the HIV infection and IMT cannot be proven, some authors have suggested that IMT might be attributable to the underlying disease that could have affected the immunological response of the host, or due to secondary infections by other viruses already mentioned such as Epstein-Barr virus or Herpesvirus-8⁽²⁵⁾.

In general, for evaluation of the IMT, magnetic resonance imaging (MRI) is better than CT both before and after surgery, because of its superior soft tissue resolution. However, there are many imaging features in this group of lesion that could mimic many diseases. Presentation as a large mass, infiltrative pattern, necrosis, or hemorrhage can be found, and may mislead diagnosis. Therefore, it is still difficult to distinguish between IMT and malignant tumor even using MRI.

There were certain limitations in the present study. First, the authors excluded more than half of the cases (6 from 9) because of incomplete images or pathological slices. Second, Case 2 had only incisional biopsy, thus the authors cannot correlate the exact CT findings with histologic components. Third, there is only one retroperitoneal IMT in the present study. This entity is exceedingly rare and meaningful analysis is difficult.

In conclusion, inflammatory myofibroblastic tumor (IMT), categorized in the intermediate group of fibrous-origin tumors, is a rare pseudosarcomatous inflammatory lesion. In the abdomen, they manifest as (a) solitary masslike lesion in the liver, either multiloculated & hypodense with enhancing septae, or hypodense with thick rim of enhancement and (b) inhomogenous mass in retroperitoneum, may or may not be accompanied by a hepatic lesion. Considerable similarities between IMT and malignant tumor make the differentiation difficult, and, hence, histologic examination is necessary.

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Inflammatory myofibroblastic tumor ในช่องท้อง: ภาพตรวจพบจากเครื่อง CT scan และลักษณะ ทางพยาธิวิทยา

วิชชุดา แย้มรับบุญ, สิทธิ์ พงษ์กิจการุณ, สุภนีวรรณ เชาว์วิศิษฐ, วรชัย ศิริกุลชยานนท์, เจียมจิตร ตปนียากร, สุวิภาภรณ์ ศิริพรพิทักษ์

Inflammatory myofibroblastic tumor (IMT) เป็นชื่อเรียกใหม่ของ inflammatory pseudotumor ซึ่งจัดอยู่ ในกลุ่มเนื้องอกที่เกิดจากเนื้อเยื่อพังผืด (fibrous-origin tumor) ได้ทำการศึกษาย้อนหลังตั้งแต่เดือนมกราคม พ.ศ. 2544 ถึง เดือนธันวาคม พ.ศ. 2548 พบว่ามีผลทางพยาธิวิทยาที่เป็นเนื้องอกที่เกิดจากเนื้อเยื่อพังผืด 115 ราย เป็น IMT 17 ราย ในช่องท้องพบ 9 รายซึ่งมีเพียง 3 รายที่มีข้อมูลการตรวจทางรังสีครบถ้วน พบลักษณะเป็นก้อนในตับ 1 ราย เหมือนฝีหนองในตับ 1 ราย รายที่ 3 เป็นก้อนขนาดใหญ่ใน retroperitoneum ซึ่งมีลักษณะเหมือนเนื้อร้ายร่วมกับ มีรอยโรคขนาดเล็กในตับซึ่งโตขึ้นอย่างมากในการติดตามผลที่ 9 เดือน

IMT ในช่องท้องพบได้น้อย เมื่อพบมักจะวินิจฉัยได้ยาก เพราะผู้ป่วยอาจมาพบแพทย์ด้วยอาการต่างกัน เช่น อาการไข้ หรือคลำได้ก้อนในท้อง ผลการตรวจทางห้องปฏิบัติการจะพบความผิดปกติแบบไม่เฉพาะเจาะจง (nonspecific) หรือ อาจอยู่ในเกณฑ์ปกติ การตรวจทางรังสีอาจพบลักษณะแตกต่างกันได้หลายแบบ ซึ่งอาจชี้นำการรักษา ไปในทางที่ผิด การวินิจฉัยที่ถูกต้องได้จากการตรวจชิ้นเนื้อ (biopsy)