Inflammatory Myofibroblastic Tumor of the Soft Tissue — Sonographic and Computed Tomographic Features: A Case Report

Hui-Lun Zhan, Yi-Hong Chou, Chui-Mei Tiu, Hong-Jen Chiov, Jen-Dar Chen, Winby Chen, Cheng-Yen Chang and Chun Yu

Soft-tissue inflammatory myofibroblastic tumor (IMT) is a rare benign lesion. Only six cases of IMT of the skin and subcutaneous tissue have been reported recently. We report a rare case of soft-tissue IMT with multifocal recurrence after surgical excision. Sonographic features of these discrete lesions revealed heterogeneous hypoechoic masses with irregular margins and several echogenic foci in the lesion. Stranding of the adjacent fat and mild thickening of the overlying skin were also evident. Color Doppler ultrasound showed increased flow in some portions of the lesion. Computed tomography showed an ill-defined nodule at each site with homogeneous hypodensity but no obvious enhancement.

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KEY WORDS: • inflammatory myofibroblastic tumor • multifocal • soft-tissue tumor • ultrasound • computed tomography

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare benign lesion related to or identical to inflammatory pseudotumor or plasma cell granuloma [1]. Although IMT can occur in any part of the body, the lung is the most common site [2]. Soft-tissue IMT is especially rare. Only six cases of IMT of the skin and subcutaneous tissue have been reported recently [1]. It occurs more commonly in children and young adults, and occasionally in older adults [1]. IMT is characterized by inflammatory masses of uncertain etiology that simulate true neoplasm and, therefore, often result in a diagnostic dilemma [2]. Diagnostic uncertainty may lead to unnecessary radical surgery in some instances. We report a rare case of soft-tissue IMT with multifocal recurrence after initial surgical excision.

CASE REPORT

A 57-year-old female was admitted to our hospital in August 1996, complaining of painful palpable masses in the subcutaneous region of the right upper arm and right elbow for 2 months. Physical examination revealed two movable hard nodules in the right upper arm that measured about 2 × 1 cm and another nodule in the right elbow that measured...
about 0.5 × 0.5 cm. Excisional biopsy and histopathology showed a picture of IMT. However, several painful nodules in the right upper arm, right elbow and right upper back were noted in January 1997. Repeated excisional biopsies were performed in all the nodules. Histopathology was consistent with IMT. In July 2002, the patient found some rapidly growing nodules in the left upper abdominal wall and right thigh. Two months later, a small nodule was noted in the left breast. Sonographic examination of the left upper abdominal wall revealed a heterogeneous hypoechoic mass with an irregular margin and several echogenic foci in the lesion. Stranding of the adjacent fat and mild thickening of the overlying skin were also evident. Color Doppler ultrasound showed increased color flow signals in some portions of the lesion (Fig. 1). At the same time, one nodule in the left breast and one in the right thigh were observed by ultrasound; both showed the same sonographic characteristics as the nodules in the abdominal wall (Fig. 2). Computed tomography (CT) of the whole abdomen and the chest showed an ill-defined nodule at each site with homogeneous hypodensity but no obvious enhancement in the left anterior lower chest wall and left upper quadrant of the abdominal wall. There was thickening of the overlying skin and desmoplastic reaction in the peripheral fat (Fig. 3). Excisional biopsy of these two lesions showed a picture of IMT (Fig. 4). Culture of the biopsy specimens showed no evidence of bacterial growth.

DISCUSSION

IMT is a rare lesion that has been described in the literature under a number of names, including inflammatory pseudotumor, plasma cell granuloma, histiocytoma, pseudolymphoma, fibroxanthoma, and plasmacytoma [3]. It is mainly a tumor of young adults and children [1,4], but has been reported in infants as young as 9 months and in adults up to 83 years of age [4]. Our patient was an older adult, which is unusual. These tumors have been detected in the liver, small bowel mesentery, pancreas, kidney, spleen, bladder, stomach, and central nervous system [2]. The exact etiology and pathogenesis of these lesions are unknown. Trauma, surgery and infections, including Epstein Barr virus infection, have been implicated in the etiology in some cases [1]. IMTs are usually slow-growing lesions [2]. However, in our patient, the nodules that developed in 2002 were relatively rapid-growing, which could be exceptional. Recent studies in a limited number of cases have demonstrated the presence of clonal cytogenetic abnormalities and anaplastic lymphoma kinase expression similar to that in anaplastic large cell lymphoma [5,6]. This may explain why some patients have relatively rapid-growing tumors. We did not perform clonal cytogenetic studies in this patient to check if there was any abnormality. The incidence of local recurrence has been reported to be 25% [1]. Most tumors recur within 1 year after initial surgery.

Fig. 1. Gray-scale and color Doppler ultrasound of the left upper abdominal wall. (A) A hypoechoic lesion with an irregular margin and some echogenic foci in the subcutaneous region (arrows). Stranding of the adjacent fat is also evident. (B) Color Doppler ultrasound demonstrates color flow signals in the lesion (arrows).
Fig. 2. Ultrasound of the breast (A) and right thigh (B) shows hypoechoic nodules with irregular margins and stranding of the adjacent fat (arrows), similar to those on the chest wall.

Fig. 3. Computed tomography of the chest (A, B) and upper abdominal wall (C, D), pre-(A, C) and post-(B, D) intravenous contrast injection. (A, C) Ill-defined nodules with homogeneous hypodensity in the subcutaneous area of the left anterior lower chest wall and abdominal wall (arrows). Thickening of the overlying skin and desmoplastic reaction in the peripheral fat is also evident. (B, D) Post-contrast computed tomography shows no evidence of contrast enhancement in the lesions (arrows).
Distant metastasis is rare. It has been proposed that multifocal lesions are most likely, rather than metastatic spread [7]. Interestingly, sarcomatoid transformation from a typical IMT to undifferentiated sarcomatoid proliferation and blending with a histiocytoma-like pattern has also been reported. IMT is generally expected to have a benign course. However, there may be multiple local recurrences, distant metastases, sarcomatoid or malignant transformation, and even tumor-related deaths, which suggests that IMT represents the border between inflammation and malignant neoplasia [7].

Due to the need to evaluate all different areas of the tumor to make the diagnosis of IMT, needle biopsy is not a reliable diagnostic method. Complete surgical resection is the best treatment for IMT [7]. The ability of imaging studies to differentiate IMT from other diseases remains largely undefined. To our knowledge, no prototypical appearance has been described. Both macroscopic and microscopic variabilities have complicated radiologic evaluation. Another reason that IMT is confused with neoplasia is its capacity for local tissue infiltration [2]. The CT features of the lesion are variable, and no constant enhancement pattern appears. This is probably due to the variable vascularity of the lesion, which ranges from avascular to hypervascular [8]. The sonographic findings of IMT are also variable. In previous reports on sonographic features of IMTs, most lesions were described as well circumscribed nodules or masses, with either hypoechogenicity or hyperechogenicity [8]. In our case, the sonographic findings included a heterogeneous hypoechic mass with an irregular margin, fibrous stands or infiltration of the adjacent fat, mild thickening of the overlying skin, and several echogenic foci in the lesion. Color Doppler ultrasound showed increased color flow signals in some portions of the lesion. The echo pattern is not specific, but multiple firm nodules in the soft tissue raise the possibility of IMT.

REFERENCES