

Imaging findings of inflammatory myofibroblastic tumor in breast

A case report

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Abstract

Rationale: Inflammatory myofibroblastic tumors (IMTs), particularly breast IMTs, are rare neoplastic lesions typically associated with a favorable prognosis. Breast IMTs are easily misdiagnosed as other types of malignant lesions, and therefore there is a significant unmet need for a better preoperational differential diagnosis based on imaging manifestations. Here, we report the imaging findings of a breast IMT and compare our findings with previously published features of breast IMTs.

Patient concerns: The patient, a 43-year-old female, reported the presence of a palpable lump within her left breast. An ultrasound examination revealed an irregular hypoechoic mass with unclear boundaries. Mammography demonstrated a mass of heterogeneous and striped density with granular calcification. Magnetic resonance image (MRI) inspection displayed an irregular tissue lump with an undistinguishable boundary and a further dynamic contrast-enhanced MRI disclosed an associated efflux change.

Diagnoses: Breast inflammatory myofibroblastic tumors.

Interventions: Breast needle biopsy and mammary resection were performed. Pathological staining of the bulk resected tumor after preoperative preparation revealed that the tumor-like tissue was enriched for spindle cells arranged in fascicular clusters. Histopathological diagnosis and immunohistochemistry confirmed the mass as being a breast IMT.

Outcomes: No metastatic recurrence was found during 6-month or 1-year follow-ups.

Lessons: Breast IMTs commonly develop in elderly women with atypical imaging features. They are primarily composed of lobular soft tissues infiltrated with an abundant focal blood supply and granular calcification. Development of breast IMTs is closely related to trauma. A preliminary diagnosis of such masses can be made based on combined manifestations of both clinical and imaging features, while a final confirmation still requires pathological staining. Imaging examinations are of value for such tumors to define the lesion edges and their associations with adjacent tissues.

Abbreviations: ALK = anaplastic lymphoma kinase, CDFI = color Doppler flow imaging, CT = computed tomography, DWI = diffusion-weighted image, IMT = inflammatory myofibroblastic tumor, MRI = magnetic resonance image, SMA = smooth muscle actin, WHO = World Health Organization.

Keywords: breast, inflammatory myofibroblastic tumor, mammography, MRI, ultrasound

1. Introduction

Inflammatory myofibroblastic tumors (IMTs), first reported by Brunn in 1939, are a rare benign tumor-like inflammatory lesion. They are infiltrated by myofibroblasts and inflammatory cells such as lymphocyte and plasmacytes.^[1] In 2002, WHO officially designated the IMT name and defined these tumors as intermediate, occasionally metastatic and locally recurrent

tumors. IMTs can develop in various tissue types at all ages, with the most common sites of localization including lung, liver, mesentery, omentum, and peritoneum. Previous research suggests that breast IMTs are particularly rare.^[2–4] Because breast IMTs lack typical clinical and imaging characteristics, they are easily to be misdiagnosed and confused with other breast disorders such as cancer or fibroadenoma. Through deep analysis of 1 breast IMT with complete pathological and imaging documentation in our hospital, we sought to shed new understanding on this rare tumor-like disorder.

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2. Consent

The clinical and imaging data were obtained with the patient's consent for purpose of scientific research publication.

3. Case Report

A 43-year-old female patient suffered from local dull pain and discomfort in her left upper breast without obvious inducement for approximately 6 months. The patient noted that the irregular pain and discomfort, which was aggravated during menstruation, had been exacerbated during the past 2 months. The patient had

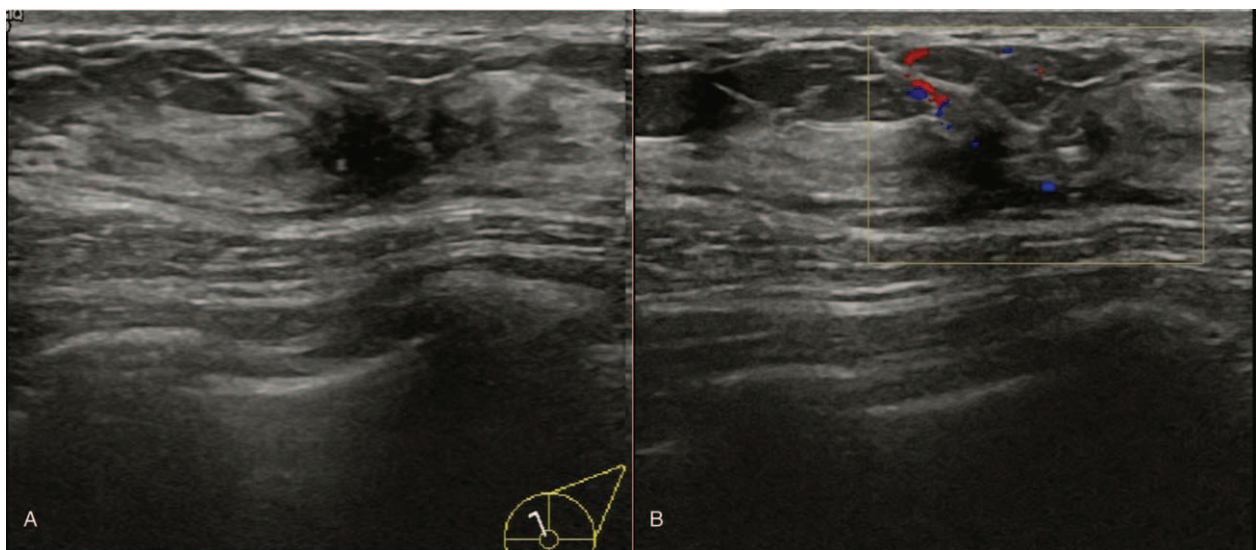


Figure 1. Ultrasound: a postoperative scar can be seen on the upper portion of the left breast and an irregular hypoecho was detected medially about 4.0 cm from the papilla. The scar was about 1.4 cm × 0.8 cm × 0.8 cm, with unclear boundaries and the internal echo was heterogeneous with an evident strong fleck echo (A). CDFI indicated limited blood flow signal within the hypoechoic region (B).

undergone a mass resection operation on her left breast one and half years prior, which had been pathologically diagnosed as breast fibroadenoma. Gynecological examination: by palpation, an approximately 1.0 cm × 0.8 cm mass, hard, with no clear boundary and poor activity but obvious haphalgnesia was evident on the upper quadrant of left breast. Ultrasonic examination (Fig. 1) showed that a vertical postoperative scar was detectable on the upper quadrant of the left breast. An irregular hypoecho, about 1.4 cm × 0.8 cm × 0.8 cm, with an unclear boundary was detected about 4.0 cm from the papilla. The internal echo was heterogeneous and a strong echo from flecks was evident. Color Doppler flow imaging (CDFI) indicated the presence of limited

blood flow within the hypoechoic area. A mammography check (Fig. 2) showed that the parenchymal density of the left breast was heterogeneous. Striped density increases and granular calcification could be seen without any associated detectable lump or dilated vessels. A dynamic contrast-enhanced breast magnetic resonance image (MRI) (Fig. 3) detected an irregular mass in the upper quadrant of the left breast, with a maximal cross section of about 1.1 cm × 1.2 cm and an unclear boundary. Lobulation and small sentus were apparent on the margin. The diffusion-weighted image (DWI) signal was elevated, and reinforcement was uneven. Dynamic contrast-enhanced imaging revealed an efflux change. A breast needle biopsy of the mass was conducted under ultrasound guidance, and this was followed by the expanded resection of the left breast (left mammary section resection). No recurrence or metastasis was found during a 6-month or 1-year follow-up visit.

Pathological examination (Fig. 4): even and uniform spindle cells arranged in bundles were abundant. Mitotic index phenomenon was rare, and inflammatory cells presence was limited. Immunohistochemistry staining results (Fig. 4): CK-, CK7-, CK5/6-, CK(HMW)-, P63-, CK8+, smooth muscle actin (SMA)+, Desmin+, 34βE12-, Vimentin+, S-100-, anaplastic lymphoma kinase (ALK)-, CD117-, CD23-; the Ki-67 score was about 10%. The final pathological diagnosis was IMT.

4. Discussions

IMT, characterized by low potential malignancy or borderline, is a rare type of benign tumor. Unlike malignant neoplasms, IMTs are more common in young people and primarily consist of spindle fibroblasts and myofibroblasts with inflammatory cell infiltration. IMTs were once known as inflammatory pseudotumors, plasma cell granulomas, and inflammatory fibrosarcomas due to their variable histomorphology. Recent molecular studies have discovered that 2p23 of the ALK genes of IMT patients were rearranged, therefore pointing to these masses being true cancerous lesions.^[5]

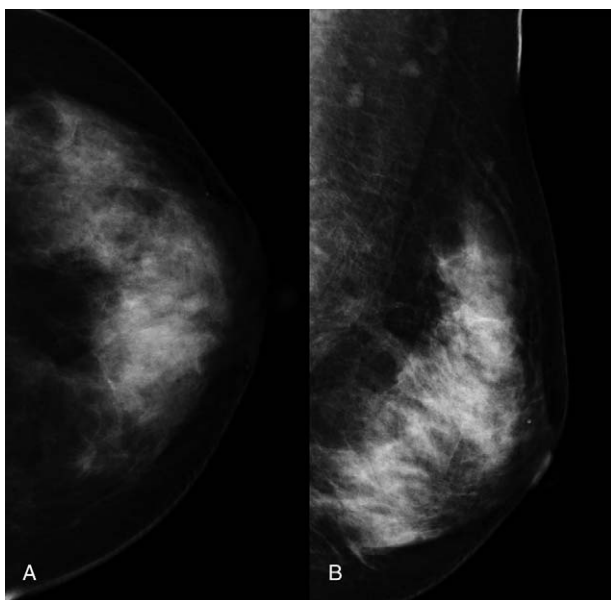


Figure 2. Mammography: substance density of the left breast was heterogeneous, and striped density increases and annular calcification can be seen.

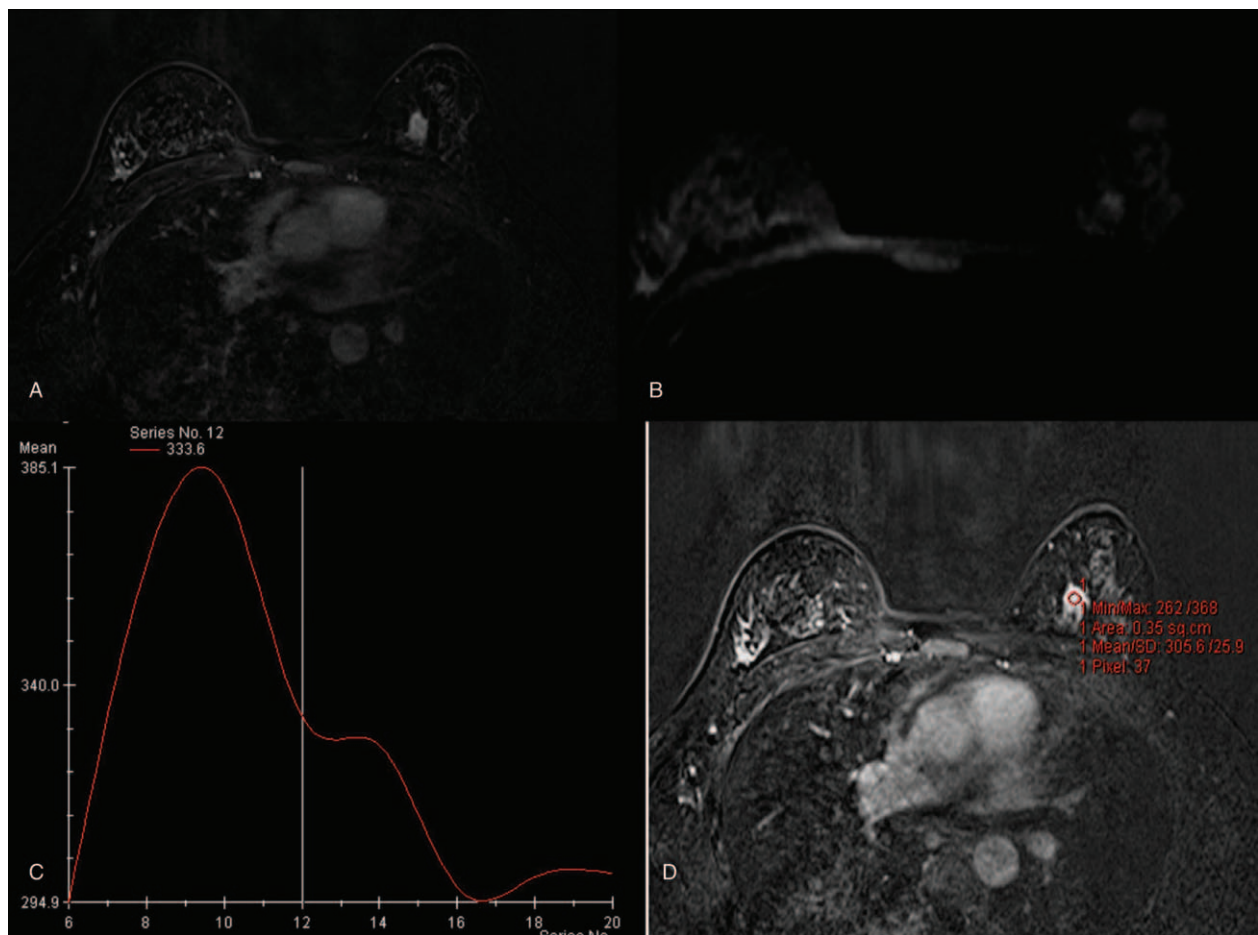


Figure 3. Dynamic contrast-enhanced MRI: irregular mass in the upper quadrant within the left breast, with a maximal cross-section of about 1.1 cm × 1.2 cm with unclear boundaries. Lobulation and small sentus can be seen on the margin (A). DWI was in the form of higher signal and reinforcement was uneven (B). Dynamic contrast-enhanced imaging represented an efflux change (C, D).

IMTs develop most often in the lung, whereas they rarely occur in other tissue types such as the breast. According to published studies, a total of 12 cases of breast IMTs (Table 1) have been reported to date.^[2-4,6-13] The average age of onset is 48.9 years old (range: 23–60). Unlike IMTs which develop in other organs of patients whose average age of onset was younger, most cases of breast IMTs occur in individuals >40 years of age.^[14] The patient in the present study was 43 years old, and was therefore in the appropriate age range for breast IMT development. The diameter of the 12 previously published masses ranged from 10 to 90 mm, with an average of 30.4 mm. Focal ultrasound examination in this case report demonstrated a solid irregular hypoechoic mass with unclear boundaries and a foliar margin periphery. Internal echo was uneven, and there was evidence of blood flow on the marginal area. Mammography characterized an irregular soft tissue mass with unclear boundaries, with some calcified focal flecks. Computed tomography (CT) examination indicated a morphologically irregular soft tissue mass with unclear boundaries. Breast MRI examination identified a morphologically irregular and uneven reinforced soft tissue mass with unclear boundaries and obvious reinforcement. Choi et al^[4] have previously reported that dynamic contrast-enhanced MRI imaging of breast IMTs reveals obviously even enforcement in early stage and no enforcement in the cystic area. Our imaging results were similar to those described for a typical IMT, and the

uneven mass enforcement and small sentus on the focal margin observed in the present case are believed to be caused by surgery-induced focal fibroplasia. Because of similar malignant morphology and hemodynamic characteristics, such as uneven enhancement of irregular lumps, rapid enhancement, and outflow curves, it is difficult to distinguish between breast cancer and IMT via imaging alone. Some believe that a clear IMT boundary may allow for a clearer diagnosis of IMT. However, the characteristics of certain low-grade malignant spindle cell tumors, such as desmomas, are similar to IMTs, further complicating such diagnostic imaging. In terms of localization, a desmoma is generally located outside the pectoral fascia, potentially allowing for better tumor identification on the basis of such localization. In general, breast IMTs, which lack representative imaging findings, should be considered if a patient is of an appropriate age and other diseases such as breast cancer have been excluded.

To date, the pathogenesis and mechanism governing IMT development remain to be clarified. Some researchers believe that IMTs are related to chromosome abnormalities, whereas others^[15] believe them to be tumorous lesions caused by hyperplasia of myofibroblasts with a proliferative advantage due to overreaction or trauma-induced inflammation. The IMT focal zone reported in our case was located near to the site of a surgical operation 18 months prior, leading us to speculate that

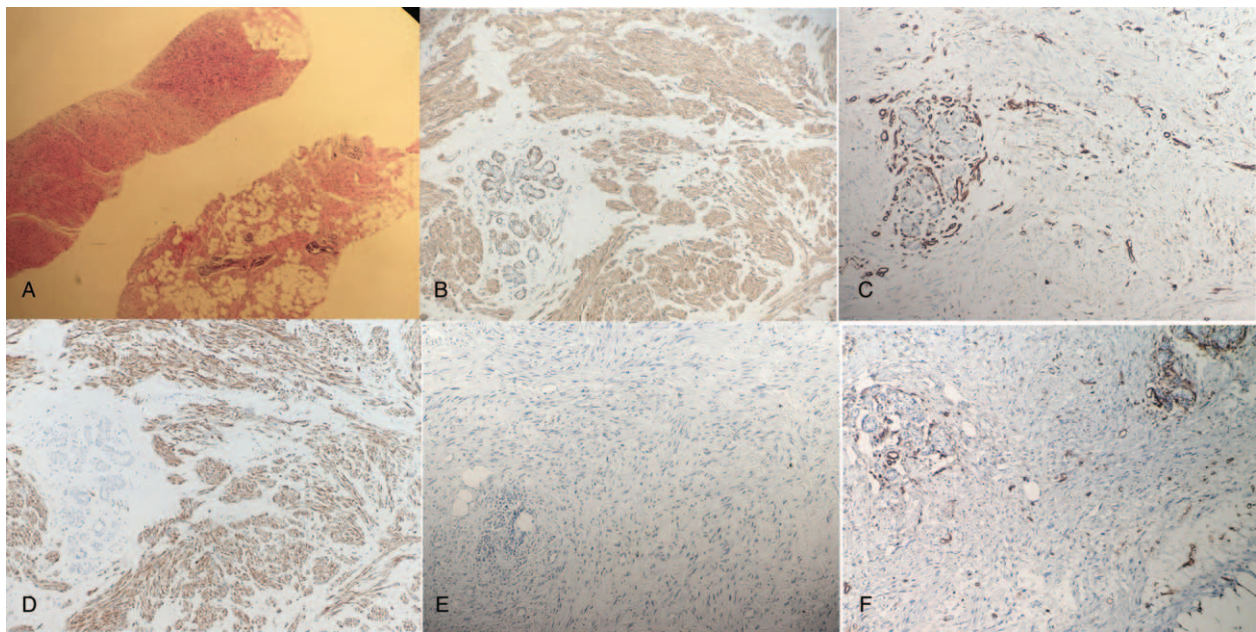


Figure 4. Histopathology and immunohistochemistry (A). Histological examination (original magnification 4×) showed an area of mucous edema, spindle cells showing no nuclear division and no necrosis, and inflammatory cells in dense areas, with obvious degeneration of the collagen fibers. The tumor cells stained diffusely positive for (B) SMA (original magnification, 10×), and focal expression of (C) vimentin and (D) desmin (original magnification, 10×), and (E) negative for S-100 (marker of vascular endothelial cells, original magnification, 10×), suggestive of an inflammatory myofibroblastic tumor. SMA = smooth muscle actin.

Table 1

Clinical information of breast inflammatory myofibroblastic tumor in the English literature.

Reference	Case number	Age	Sex	Location	Size, mm	X	U	CT	MRI
Zhao ^[6]	1	56	F	Right	40	+	+	+	
Choi ^[4]	1	27	F	Right	30	+	+		+
Xing ^[7]	1	56	F	Right	90			+	
Kim ^[8]	1	60	F	Left	15	+	+		
Park ^[9]	1	47	F	Right	30	+	+		
Bosse ^[2]	1	23	F	Left	20		+		+
Siraj ^[10]	1	60	M	Left	15		+		
Markopoulos ^[3]	1	67	F	Left	10	+	+		
Li ^[11]	1	39	F	Left	40			+	+
Li ^[11]	1	39	F	Left	25			+	+
Ilvan ^[12]	1	60	F	Right	10	+			
Hijil ^[13]	1	53	F	Right	40	+	+		

this IMT occurrence was closely related to operation-stimulated trauma.

Coffin^[15] classifies IMTs into 3 categories: (1) myxoid pattern, or angioid type, or nodular fasciitis type; (2) dense spindle cell type or fibrous histiocytoma pattern; (3) hypocellular fibrous pattern or desmoid fibromatosis. This second category is the most common form of IMT, and the category to which this case belonged. The diagnosis of IMT should be based on immunohistochemistry staining as follows: diffuse positive staining of Vimentin and myogenic markers such as Desmin, SMA, and MSA focal staining in the tumor specimen; approximately one-third of cases stain positive for focal keratin, with negative staining for S100, CD117, and CD23. In the present case, SMA+, Desmin+, and Vimentin+ were all positive, whereas S100, CD117, CD23, CD34, and ALK were negative, supporting the diagnosis of IMT.

In summary, breast IMT is a rare disease and develops most commonly in elderly women and presents without distinctive imaging signs. The focal zone, in most cases related to trauma, contains mainly a lobular soft tissues mass with an abundant blood supply and some granular calcification. Primary diagnosis can be made based on a combination of clinical and imaging features, whereas a confirmed diagnosis requires pathological indexes. Imaging examinations are of great value in specifying the focal range of these tumors and their relations to adjacent tissues.

Author contributions

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Supervision: Xljin Mao.

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Visualization: Ning Yu.

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