

Case Report

Inflammatory Myofibroblastic Tumor of the Breast

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Inflammatory myofibroblastic tumors (IMTs) of the breast represent extremely rare lesions. Due to the scarcity of reports, their natural history, recurrence, and metastatic potential remain poorly defined. We report on a case of a primary breast IMT in a postmenopausal female patient treated successfully with breast conserving surgery and review the literature pertaining to this rare entity.

1. Introduction

Inflammatory myofibroblastic tumors have largely been considered as a subgroup of inflammatory pseudotumors [1] and have been encountered in various anatomical locations [2–7]. Interestingly, mammary IMTs have scarcely been reported. These lesions demonstrate variable clinical features and their neoplastic nature is ill-defined to date. Recent studies on IMTs have identified clonal abnormalities of the anaplastic lymphoma kinase (ALK) gene, but the impact of this feature on the neoplastic behavior of the tumor is not clarified [8]. We herein report on a rare case of a primary breast IMT in a postmenopausal woman and review the literature regarding the clinicopathological characteristics of these extremely rare lesions.

2. Case Presentation

A 67-year-old female patient was admitted to our breast unit for management of a recently palpated lump located on the upper outer quadrant of her left breast. Past medical history was significant for a total abdominal hysterectomy at the age of 51 due to multiple, large fibroids. She had no family history of breast cancer and previous screening mammograms were normal. Physical examination revealed a nontender, firm mass in the upper outer quadrant of the left breast with

moderate skin dimpling. Examination of the contralateral breast and both axillas was unremarkable. Bilateral diagnostic mammogram (Figures 1 and 2) demonstrated a roughly round-shaped mass with ill-defined margins measuring $1 \times 0.8 \times 0.7$ centimeters, situated on the upper outer quadrant of the left breast, without any other abnormality of the contralateral breast or both axillary regions. The lesion was reported as BIRADS IV; dedicated breast ultrasound confirmed a heterogeneous oval mass with echogenic rim, reported as BI-RADS ACR4 (Figure 3).

The patient underwent an excision biopsy under general anesthesia; on frozen section the tumor was highly suspicious for lymphatic or myofibroblastic neoplasia. Surgical margins of the specimen were free of disease; in view of the frozen section evaluation, planned sentinel lymph node biopsy was not performed.

Pathology examination of paraffin embedded sections of the tumor exhibited tuberous aggregations of lymphocytes without any blastic centers, with concurrent invasion by numerous plasma cells, histiocytes and gigantic cells, probably myofibroblasts, and scarce polymorphonuclear and eosinophilic cells. At immunohistochemistry the lesion stained positive for vimentin, actin, plasmacytoma CD138, histiocyte CD68, Ki67 (5%), and lymphocytes CD20, CD3, and CD5, while it was negative for desmin, anaplastic lymphoma kinase (ALK), pankeratin, keratin 34BE12, CD15, and CD30.

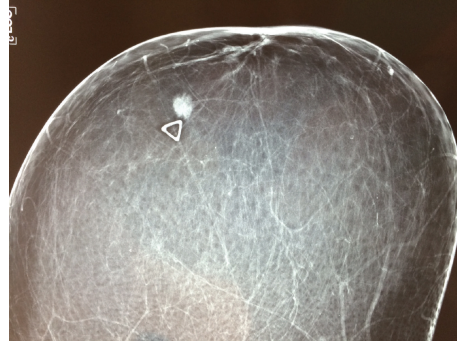


FIGURE 1: Left craniocaudal (CC) mammographic view: round centimetric mass with indistinct margin (arrow); BI-RADS 4.



FIGURE 2: Left mediolateral oblique mammographic view: the mass appears superficial.

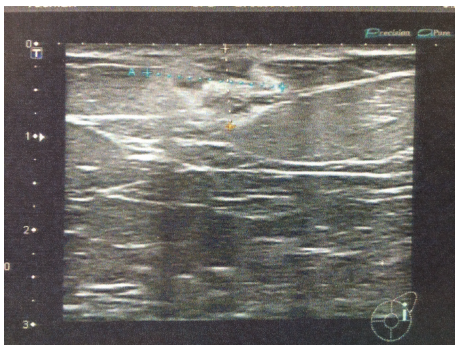


FIGURE 3: Ultrasound: heterogeneous oval mass with echogenic rim; BI-RADS ACR4.

Pathology concluded for an anaplastic lymphoma kinase-(ALK-) negative inflammatory myofibroblastic tumor of the breast (Figures 4(a)–4(f)).

Full staging investigations were negative for systemic disease and the patient received no further treatment. Six months after surgery, the patient is well and free of disease.

3. Discussion

The World Health Organization (WHO) classifies inflammatory myofibroblastic tumors (IMTs) as distinct borderline lesions, while the physical history of these lesions can variably range from reactive to truly neoplastic [1]. Inflammatory myofibroblastic tumor of the breast is an extremely rare lesion. Generally, IMTs are roughly considered as a subset of inflammatory pseudotumors (IPTs); yet the interchangeable use of either term in the literature can generate some confusion; purportedly, these two entities (IMT and IPT) share common morphology although they demonstrate different clinicopathological features [9].

IMTs have been observed predominantly in young patients and have been encountered virtually in any anatomical location such as the lungs [2], mesentery [4], omentum [3], retroperitoneum [5], extremities [6], head [7], liver [10], spleen [11], thyroid [12], and urinary bladder [13]. Notably, reported cases on IMTs/IPTs of the breast are very scarce [1, 9, 14–34].

Microscopically, IMTs usually are characterized by the presence of spindle cell proliferation and inflammatory infiltrates by lymphocytes, plasma cells, histiocytes, or, less frequently, large vacuolated cells [15, 16]. The pathogenesis of IMTs is largely undetermined. While they were initially believed to generate upon an inflammatory or infectious stimulus, recent studies on these rare entities have revealed aberrations located in chromosomes 2 and 9 [35]. Furthermore, nearly half of IMT cases reportedly exhibit mutations involving the anaplastic lymphoma receptor tyrosine kinase gene (ALK) at 2p23. This feature has favored a neoplastic nature of these tumors, as clonal abnormalities of the ALK gene were first described in anaplastic large cell lymphoma (ALCL), which is a true neoplasia [1, 36, 37]. In ALCL patients though, ALK-positivity is reportedly accompanied by a less aggressive clinical course. Whether there is a more favorable outcome of patients with an ALK-positive IMT is yet to be defined [38, 39].

Since an IMT of the breast can mimic or even behave as true malignancy, wide excision with negative margins is highly recommended [1, 37]. As this is a very rare lesion to be encountered in the breast, data on malignant course, recurrence, and metastasis are scarce. Zhao et al. reported

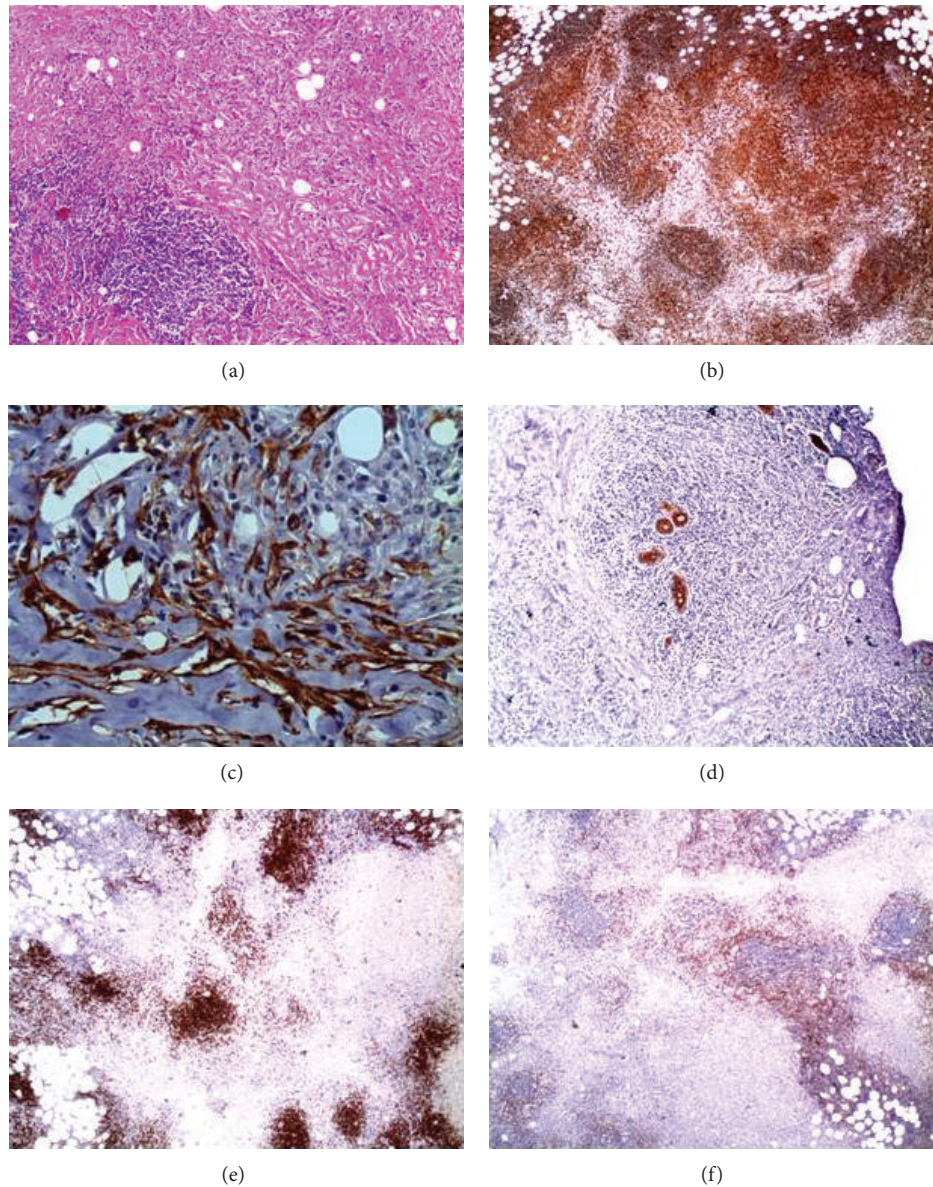


FIGURE 4: (a) Hematoxylin-eosin (H&E) staining of the lesion (magnification $\times 10$): significant inflammatory invasion by lymphocytes and plasma cells; presence of stacks of fusiform myofibroblasts and few gigantic cells. (b) Vimentin staining of the lesion (magnification $\times 10$): presence of myofibroblasts and lymphocytes. (c) Actin staining of the lesion (magnification $\times 40$): presence of myofibroblasts which stain positive for actin. (d) Keratin 34BE12 staining (magnification $\times 10$): myofibroblasts immunonegative to keratin 34BE12. (e) CD20 staining (magnification $\times 4$): CD20 positive B-lymphocytes. (f) CD3 staining (magnification $\times 4$): CD3 positive T-lymphocytes.

one patient who had local recurrence and metastasis to the left groin area 3, 7, and 10 months after initial surgery for an IMT of the breast [14]. Moreover, studies have reported on recurrence rates up to 25% for IMTs on other anatomical locations [37].

Interestingly, axillary management in cases of breast IMT is yet undefined. In none of the case reports so far did the authors encounter any axillary involvement in the setting of a primary IMT of the breast. Furthermore, the sarcomatous component of this lesion could raise a premise that lymphatic spread is unusual and hence routine SNLB is unnecessary.

Still, larger series in the future could probably clarify this matter.

Chemotherapy, radiotherapy, and immunomodulation have not been reported to be consistently effective against IMTs, albeit sporadic cases of treatment with chemotherapy or anti-inflammatory agents have been reported [40, 41]. Wide excision with free margin remains the treatment of choice for patients with IMTs and is adequate in most circumscribed tumors. However, tumors with ill-defined morphology and/or incomplete resection have been associated with higher recurrence rates [42].

4. Conclusion

IMT of the breast represents a very rare entity with intermediate clinical behavior. Although reportedly infrequent, recurrence and metastatic potential exist and, as such, wide surgical resection with close clinical mammographic follow-up is recommended.

Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

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