Inflammatory Myofibroblastic Tumor of the Breast Coexisting with Breast Cancer: A Case Report

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Keywords

Inflammatory myofibroblastic tumor · Breast cancer

Summary

Background: The inflammatory myofibroblastic tumor (IMT) is an uncommon low-risk lesion with only a few cases described in the literature. **Case Report:** Here, we report a unique case of an IMT coexisting with breast cancer. Modified radical mastectomy was performed, followed by TAC chemotherapy (taxotere, adriamycin and cyclophosphamide). At the 2-year follow-up, the patient continues to be disease free. **Conclusion:** At the preoperative stage, definitive diagnoses of masses are extremely difficult; surgery is advised only after the diagnosis is confirmed by pathological examination.

Schlüsselwörter

Entzündlicher myofibroblastischer Tumor · Brustkrebs

Zusammenfassung

Hintergrund: Der entzündliche myofibroblastische Tumor (IMT) ist eine ungewöhnliche, mit niedrigem Risiko behaftete Läsion, von der nur wenige Fälle in der Literatur beschrieben sind. **Fallbericht:** Wir berichten hier von einem einzigartigen Fall eines IMT, der zusammen mit Brustkrebs auftrat. Eine modifizierte radikale Mastektomie wurde durchgeführt, gefolgt von einer TAC-Chemotherapie (Taxotere, Adriamycin und Cyclophosphamid). Bei der 2-Jahres-Nachuntersuchung war die Patientin weiterhin krankheitsfrei. **Schlussfolgerung:** In der präoperativen Phase ist eine definitive Diagnose der Tumoren extrem schwierig; eine Operation ist nur dann angeraten, wenn die Diagnose durch eine pathologische Untersuchung bestätigt werden konnte.

Introduction

The inflammatory myofibroblastic tumor (IMT) is a rare lesion that can affect any part of the body. Its occurrence in the breast is rare, and only a few cases have been described according to reports. Herein, we identify, for the first time, a unique case of an IMT coexisting with breast cancer.

Case Report

A 39-year-old woman presented in the Jiangsu cancer hospital with 2 lumps in the left breast. The mass in the upper inner quadrant of the breast was hard and mobile, with a size of 4.0 × 4.5 cm. The second mass, in the upper outer quadrant, had less well-defined boundaries and mobility and was approximately 2.5 × 2.0 cm in size. Contrast-enhanced computed tomography (CT) and a magnetic resonance imaging (MRI) scan further identified the 2 masses (fig. 1).

We performed a surgical excision of the 2 masses. Pathologic analysis revealed that the small mass was an invasive ductal carcinoma and the large mass was an IMT (fig. 2). Therefore, a follow-up modified radical mastectomy of the left breast was performed with adjuvant TAC chemotherapy (taxotere, adriamycin and cyclophosphamide). Immunohistochemical analysis was performed on both resected tissues (table 1). As would be expected for an IMT, anaplastic lymphoma kinase (ALK), vimentin, and smooth muscle actin (SMA) were positively detected, whereas the detection of desmin was negative, which is an atypical result for an IMT.

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Fig. 1. Imaging diagnostics of the breast masses. **(A)** A contrast-enhanced CT scan revealed 2 masses within the left breast, measuring 3.0×5.0 cm and 1.5×1.0 cm, respectively. **(B)** MRI of the lesion showed a mass of 3.4×4.8 cm with clear boundaries in the upper inner quadrant of the left breast and a mass of 1.6×2.4 cm with dense shadow and less well-defined outlines in the upper outer quadrant of the left breast.

Fig. 2. Hematoxylin and eosin staining of the resected breast masses. **(A)** Invasive ductal carcinoma, **(B)** IMT, characterized by spindle cell proliferation and collagen deposits.

Table 1. Immunohistochemical analysis of the resected breast lesions

Breast cancer			IMT		
Molecule		Percentage	Molecule		Percentage
VEGF	+	80%	AE1/AE3	–	N/A
FGFR	+	90%	ALK	+	2%
ER	+	20%	CD34	+	tumor vessels
PR	+	40%	CD31	–	N/A
C-erbB-2	++	80%	vimentin	+	N/A
p53	+	80%	desmin	–	N/A
MDR	–	N/A	actin	+	tumor vessels
E-cadherin	+	N/A	ER	–	N/A
TOPII	–	N/A	PR	–	N/A
GST-N	–	N/A	S-100	+	N/A
			SMA	+	N/A
			LCA	+	lymphocytes

VEGF = Vascular endothelial growth factor, FGFR = fibroblast growth factor receptor, ER = estrogen receptor, PR = progesterone receptor, MDR = multi drug resistance, TOPII = topoisomerase II, GST-N = glutathion-S-transferase-N, AE1/3 = cytokeratin AE1/3, ALK = anaplastic lymphoma kinase, SMA = smooth muscle actin, LCA = leukocyte common antigen, + = detectable, – = not detectable, N/A = not applicable.

Discussion

The IMT is an uncommon low-grade lesion composed of spindle cells interwoven with mature plasma cells and other inflammatory cells. IMTs were widely considered to be benign growths, until the early 1990s when Meis and Enzinger [1] published a series of 38 cases of follow-up from patients with IMT. These data showed a significant rate of aggressive, neoplastic-like IMT growth. Over the next several years, the identification of recurrent clonal rearrangements involving chromosome 2p provided additional support that the IMT was a neoplasm [2]. It is now known that chromosome 2p rearrangements involve the ALK locus [3, 4]. Although the pathogenesis of IMTs is still controversial, clinical observations and recent molecular data indicate that at least some IMTs are true neoplasms with low malignant potential [5–8].

Patients with IMTs generally present with a mass or otherwise nonspecific symptoms, due to involvement of the organs that contain the IMT. A constitutional syndrome consisting of fever, weight loss, and malaise is seen in 15–30% of the

patients [9]. Patients with IMT often present with 1 mass; however, multiple masses have also been observed [10], and an IMT can coexist with malignant cancer [11]. However, to our knowledge, the presence of an IMT coexisting with breast cancer has not been previously reported.

IMTs are diagnosed based on their pathological appearance [12]. The following guidelines should be considered for defining an IMT: (1) diffuse inflammatory infiltrate with prominent plasma cells, (2) mild nuclear atypia, including scattered ganglion-like cells, (3) a low mitotic rate without atypical forms, and (4) ALK positivity by immunohistochemistry or ALK gene rearrangement. In our experience, IMTs are more commonly misdiagnosed. Care should be taken to distinguish IMTs from other benign and malignant lesions and to avoid overtreatment.

Histologically, IMTs are classified into 3 basic histological patterns, which can be seen in combination within the same tumor: (1) a myxoid/vascular pattern, (2) a compact spindle cell pattern, and/or (3) a hypocellular fibrous (fibromatosis-like) pattern [13]. The myxoid/vascular pattern presents with a fasciitis-like appearance and with loosely arranged, plump

spindle cells in an edematous or myxoid stroma. The compact spindle cell pattern is distinguished by a cellular proliferation of spindle cells with fascicular or storiform architecture in a collagenous stroma. Compared to the other two patterns, the fibromatosis-like pattern is relatively hypocellular, with elongated, rather than plump, spindle cells in a densely collagenous background containing scattered lymphocytes, plasma cells, and eosinophils.

IMTs are classified as tumors of intermediate risk, due to a small risk for local recurrence and distant metastasis. In consideration of its pathologic features and clinical manifestations, complete surgical resection of IMTs is mandatory. Based on our experience, the therapeutic effect of surgical excision in these cases is uncertain. To patients with an IMT of the breast, a wide local excision or simple mastectomy is recommended. If a metastasis of the axillary lymph node is suspected, then a wide local excision with sentinel lymph node biopsy or modified radical mastectomy should be performed. The prognosis of an IMT is generally considered to be favorable if complete surgical resection is performed, with only a rare incidence of malignant transformation and distant metas-

tasis. Some reports show that treatment with corticosteroids improves the outcome [14]; however, these results are still under discussion, and adjunctive therapy after surgery needs further clinical investigation.

In the case presented here, the IMT coexisted with breast cancer, so a modified radical mastectomy was performed, followed by 6 courses of TAC chemotherapy. Subsequently, oral administration of tamoxifen (10 mg) was taken twice daily. After 2 years, the patient remains disease free.

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Disclosure Statement

The authors declare no conflict of interest.

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