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.

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.

*A*These two authors contributed equally to this work.

Schlüsselwörter
Entzündlicher myofibroblastischer Tumor · Brustkrebs

Zusammenfassung

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.

*These two authors contributed equally to this work.*
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.

Table 1. Immunohistochemical analysis of the resected breast lesions

<table>
<thead>
<tr>
<th>Molecule</th>
<th>Percentage</th>
<th>Molecule</th>
<th>Percentage</th>
</tr>
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<tbody>
<tr>
<td>VEGF</td>
<td>+</td>
<td>80%</td>
<td>AE1/AE3</td>
</tr>
<tr>
<td>FGFR</td>
<td>+</td>
<td>90%</td>
<td>ALK</td>
</tr>
<tr>
<td>ER</td>
<td>+</td>
<td>20%</td>
<td>CD34</td>
</tr>
<tr>
<td>PR</td>
<td>+</td>
<td>40%</td>
<td>CD31</td>
</tr>
<tr>
<td>C-erbB-2</td>
<td>++</td>
<td>80%</td>
<td>vimentin</td>
</tr>
<tr>
<td>p53</td>
<td>+</td>
<td>80%</td>
<td>desmin</td>
</tr>
<tr>
<td>MDR</td>
<td>–</td>
<td>N/A</td>
<td>actin</td>
</tr>
<tr>
<td>E-cadherin</td>
<td>+</td>
<td>N/A</td>
<td>ER</td>
</tr>
<tr>
<td>TOPII</td>
<td>–</td>
<td>N/A</td>
<td>PR</td>
</tr>
<tr>
<td>GST-N</td>
<td>–</td>
<td>N/A</td>
<td>S-100</td>
</tr>
<tr>
<td>SMA</td>
<td>–</td>
<td>N/A</td>
<td>LCA</td>
</tr>
<tr>
<td>TOPII</td>
<td>–</td>
<td>N/A</td>
<td>LCA</td>
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<tr>
<td>GST-N</td>
<td>–</td>
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</table>
| 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].
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 metastasis. 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.

References