

Case report:

Inflammatory Myofibroblastic Tumor of the Breast: A Case Report

Ahmet Salih Karaoglu¹, Mahmut Hudai Demir¹, Abdullah Ayaz¹, Hamza Uysal¹, Tuçe Soylemez², Tunc Eren³, Abdullah Aydin², Orhan Alimoglu³

Abstract:

Inflammatory myofibroblastic tumors (IMTs) of the breast are extremely uncommon lesions, usually labelled as a subgroup of inflammatory pseudotumors. They are composed of inflammatory cells and bland spindle cells without nuclear atypia. Nearly half of all IMTs of the breast include clonal translocation of the anaplastic lymphoma kinase (ALK) gene, located at the chromosome band 2p23, and generally present with a palpable lump, swelling, and pain. Herein, we present a 66-year-old female patient with pain, swelling and a palpable lump in her right breast. A 40×26 mm sized T1A hypointense and T2A hyperintense mass with slightly lobulated margins was detected at breast magnetic resonance imaging. A mass associated with an intramammary lymph node was evaluated in the upper inner quadrant of the right breast via mammography. The results were considered as BI-RADS 3. Breast-conserving surgery with sentinel lymph node biopsy was performed. During pathological evaluation, cytoplasm with poorly-defined margins, and large-nucleoli tumor cells with benign ducts between these tumor cells, were observed. Intensive inflammatory cell infiltration and sclerotic changes in different areas were also noted. The lesion stained positive for caldesmon, smooth muscle actin, vimentin, CD10, and S100; however, it was negative for ALK on immunohistochemistry. The patient remained disease-free after the surgical procedure.

Keywords: Inflammatory pseudotumor, inflammatory myofibroblastic tumor, breast, anaplastic lymphoma kinase.

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Introduction

Inflammatory myofibroblastic tumors (IMTs) are usually labelled as a subgroup of inflammatory pseudotumors.¹ IMTs are comprised of bland spindle cells without nuclear atypia and inflammatory cells such as plasma cells, lymphocytes and eosinophils.² They can be found in various anatomic locations such as the lungs, kidneys and breasts.³⁻¹⁰ Inflammatory myofibroblastic tumors are extremely rare lesions, even rarer in the breast compared to the other parts of body.^{3,4,7,10} IMTs are mostly presented with a palpable lump, swelling, and pain. Nearly half of all IMTs of the breast include clonal translocation of the anaplastic lymphoma kinase (ALK) gene located at the chromosome band 2p23.⁵ In this article, we aimed emphasize this rare tumor type

and provide a substantial addition to the literature.

Case report

A 66-year-old female patient who had a family history of breast cancer presented with pain, swelling, and a palpable lump in the upper inner quadrant of her right breast. In her past medical history, she had previously undergone appendectomy, tonsillectomy, and thyroidectomy. Moreover, she had been diagnosed with hypertension, diabetes mellitus type 2, and asthma. Upon physical examination, a mass with a size of approximately 4×4 cm was palpated in the upper inner quadrant of her right breast. The examination of her bilateral axillae was unremarkable. Breast ultrasonography (US) revealed a 39×31 mm lesion including microcalcification with rough and lobulated margins. Furthermore, a breast magnetic

1. Faculty of Medicine, Istanbul Medeniyet University, Istanbul, Turkey.
2. Department of Pathology, Faculty of Medicine, Istanbul Medeniyet University, Istanbul, Turkey.
3. Department of General Surgery, Faculty of Medicine, Istanbul Medeniyet University, Istanbul, Turkey.

Correspondence to: Prof. Dr. Orhan Alimoglu, Department of General Surgery, Faculty of Medicine, Istanbul Medeniyet University Goztepe Training & Research Hospital, Dr. Erkin Street, Goztepe, 34722, Kadikoy, Istanbul, Turkey. E-mail: orhan.alimoglu@medeniyet.edu.tr

resonance imaging (MRI) showed a 40×26 mm, T1A hypointense and T2A hyperintense mass with slightly lobulated margins in the upper inner quadrant of her right breast (Figure 1 and 2). A mass associated with an intramammary lymph node was evaluated in the upper inner quadrant of her right breast. The results were considered BI-RADS 3.

Breast-conserving surgery with sentinel lymph node biopsy was performed as follows. Periareolar methylene blue was injected under general anesthesia. Two lymph nodes were excised and sent to frozen examination. A batwing incision was created in the upper inner quadrant of the right breast and a wide excision was performed, including the tumor, to the boundary of the major pectoral muscle. The specimen was sent for frozen section examination. No metastasis was detected via sentinel lymph node biopsy, the surgical margins of the specimen were intact, and the operation was terminated.

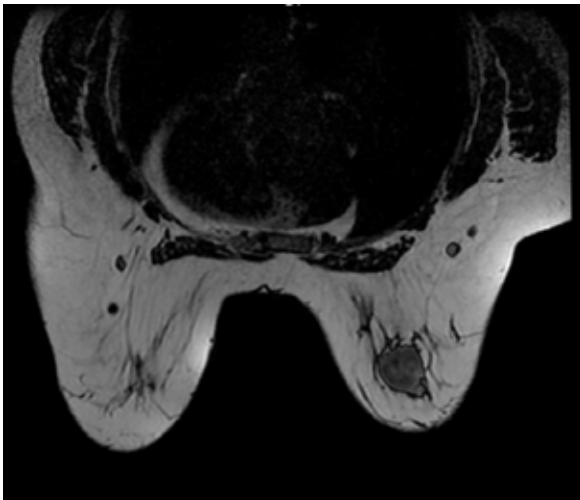


Figure 1: Breast MRI (T1A and T2A).

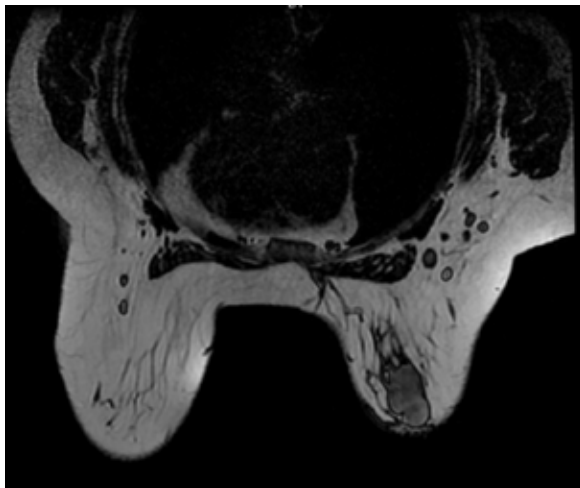


Figure 2: Breast MRI (T1A and T2A).

Discussion

In 1939, the IMT was described in the lungs for the first time in medical literature.⁶ IMTs are infrequent tumors that can be found in different anatomic locations including the lungs, omentum, retroperitoneum, mesentery, extremities, liver, spleen, head, thyroid, and urinary bladder.^{3,4,7,10} IMTs present with a firm palpable mass. In our case, the patient presented with a palpable breast lump, and pain. IMTs are microscopically characterized by the proliferation of spindle cells and inflammatory cells such as the lymphocytes, plasma cells, and histiocytes.^{8,9} In our case, large-nucleoli tumor cells with benign ducts between these tumor cells were observed. Intensive inflammatory cell infiltration and sclerotic changes in different areas were also noted (Figure 3). Despite numerous hypotheses, the pathogenesis of IMTs has not truly been understood. However, the most appropriate hypothesis is that the inflammatory process is due to either infectious or non-infectious stimuli. Aberration of the long arm of chromosome 2 and the short arm of chromosome 9 has been reported in recent studies.¹⁰ In approximately half of IMT cases, ALK gene mutation at 2p23 has been noted.^{1,2,11} The lesion stained positive for caldesmon, smooth muscle actin, vimentin, CD10, and S100; however, it was negative for ALK on immunohistochemistry in our case.

From our observations of what has been reported in literature, the Mediterranean climate region and Asian populations have shown a higher frequency of IMTs of the breast when compared to other populations. We investigated 32 IMTs of the breast in PubMed, and 13 such cases were associated with Asian populations, 13 were associated with the Mediterranean climate region while the remaining six were associated with other regions. Our patient was from a region with a Mediterranean climate. According to this data, we consider that IMTs of the breast may be correlated with specific geographical regions.

Our patient remained disease-free after the surgical procedure. We would like to emphasize the importance of keeping this rare condition in mind as a differential diagnosis during the clinical evaluation of a palpable breast mass.

Conflict of Interest:

No conflict of interest has been disclosed by the authors.

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Author Contributions:

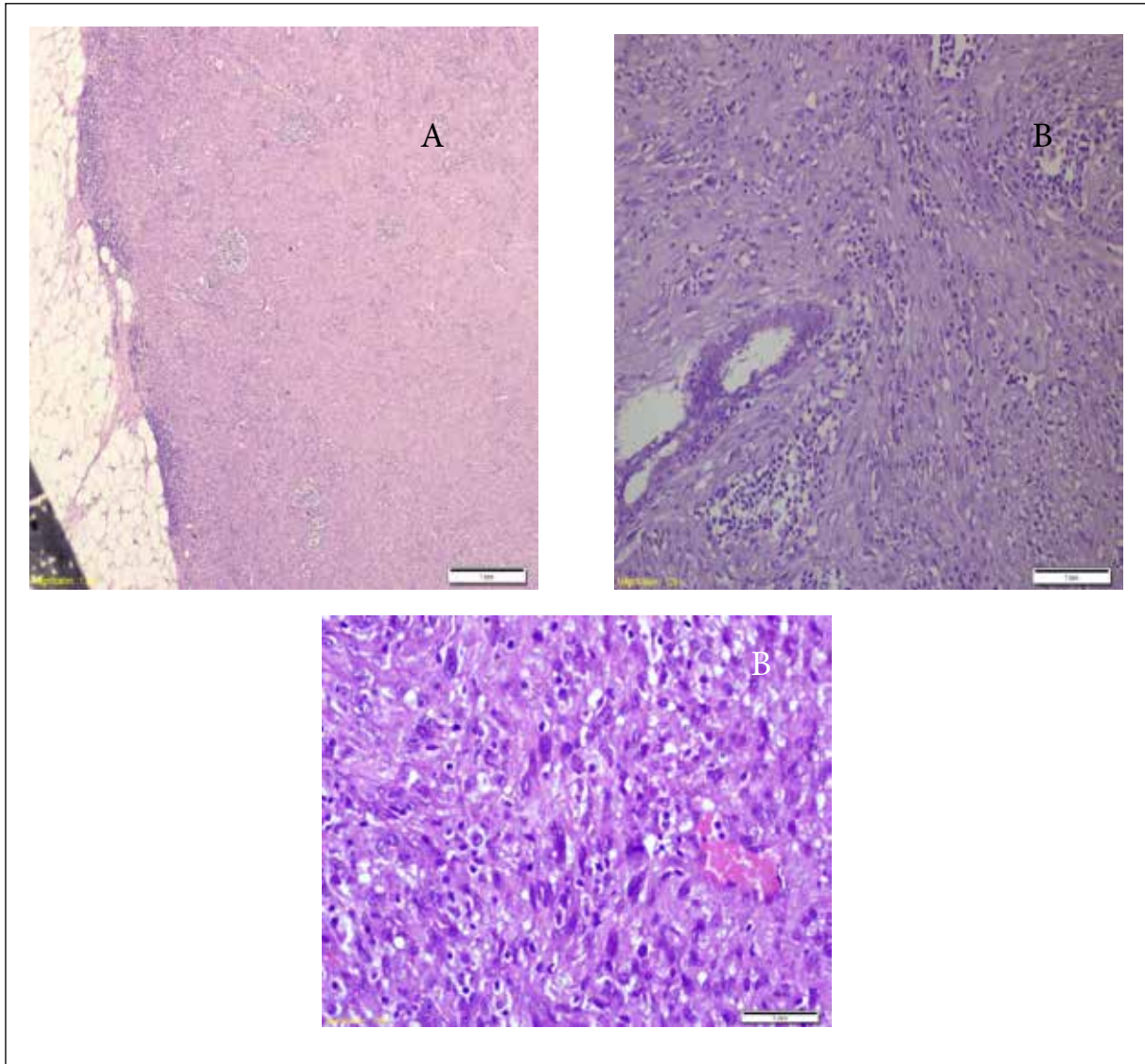


Figure 3 (a, b, c): Tumor consisting of spindle shaped neoplastic cells with accompanying inflammatory cells [Hematoxylin and eosin stain, 40× (a), 200× (b), and 400× (c)].

A.S. Karaoglu designed the research study and wrote the manuscript. M. H. Demir, A. Ayaz, and H. Uysal contributed to the designation of the study and performed the research. T. Söylemez and A. Aydın performed the research and contributed to the collection and assembly of the data. O.

Alimoğlu and T. Eren performed the research, contributed to the collection and interpretation of the data, and performed the critical revision of the article for important intellectual content.

Ethical Approval issue: N/A.

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