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症例報告

A case of inflammatory myofibroblastic tumor (low-grade malignant spindle cell tumor) of the breast
低悪性度乳腺紡錘形細胞腫の一例

Kimito YAMADA[1)3)], Masakazu KOJIKA[2)3)], Jun MATSUBAYASHI[2)], Akihiko OGATA[1)3)], Fuyou UENO[1)3)], Daisuke OHTA[1)], Hiroshi KAISE[1)], Keiichi IWAYA[2)], Harubumi KATO[3)], Norio KOHNO[1)]

山田公人[1)3)]　小鹿雅和[2)3)]　松林純[2)]
緒方昭彦[1)3)]　植野芙英[1)3)]　太田大介[1)]
海瀬博史[1)]　岩屋啓一[2)]　加藤治文[3)]
河野範男[1)]

[1)]Departments of Breast Oncology, Tokyo Medical University Hospital
[2)]Diagnostic Pathology, Tokyo Medical University Hospital
[3)]First Department of Surgery, Tokyo Medical University
[1)]東京医科大学乳腺科
[2)]東京医科大学病理診断学講座
[3)]東京医科大学外科学第一講座

Abstract

Inflammatory myofibroblastic tumor (IMT) is a distinctive pseudosarcomatous inflammatory lesion that may occur in any soft tissue or visceral location. IMT is composed of myofibroblastic spindle cells accompanied by an inflammatory infiltrate of plasma cells, lymphocytes, and eosinophils. IMT of the breast is an extremely rare condition : only 9 cases have been reported in the international literature so far.

We encountered a case of IMT in a woman's breast. The tumor enlarged rapidly within the previous year, and it was surgically excised. The tumor was pathologically examined and diagnosed as low-grade malignant spindle cell tumor.

Introduction

Inflammatory myofibroblastic tumor (IMT) is a unique form of inflammatory pseudotumor (IP), a distinctive pseudosarcomatous inflammatory lesion that may occur in any soft tissue or visceral location. Its nature and outcome differ from one organ to another. (1) According to the site of presentation, there are wide

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Corresponding author : Kimito YAMADA, Tokyo Medical University Hospital, 6-7-1 Nishi-shinjuku, Shinjuku-ku, Tokyo 160-0023, Japan
TEL +81-3-3342-6111 Ext. 5071, FAX +81-3-3349-0326
E-mail : kimito@tc5.so-net.ne.jp

clinical and histological variations. (2) IMT is composed of myofibroblastic spindle cells accompanied by an inflammatory infiltrate of plasma cells, lymphocytes, and eosinophils. IMT of the breast is an extremely rare condition: only 9 cases have been reported in the English literature so far. (3)

We report a case of IMT in a woman's breast. The tumor enlarged very rapidly, and it was surgically excised. The tumor was pathologically examined.

Case report

A 59-year-old Japanese woman noticed a mass in her left breast 3 years previously. The size of the tumor was approximately 2 cm in diameter, but she left the mass alone. It gradually enlarged, so she was referred to our breast oncology department from her home doctor for more detailed examinations and therapy. There was no family history of breast cancer. On physical examination there was a tender mass, approximately 8 cm in diameter, located in the outer lower quadrant of the left breast. The overlying skin was extended with

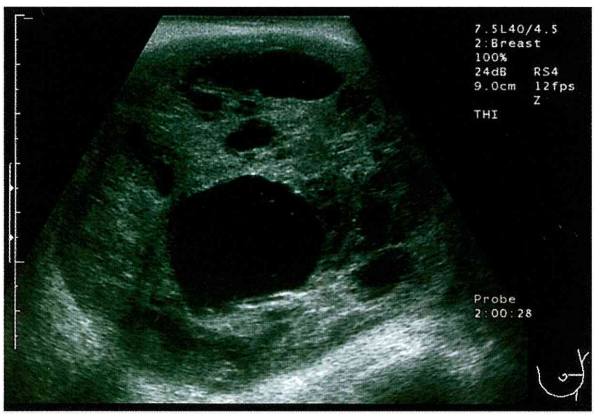

Fig. 1 Ultrasonography showed a well-defined heterogeneous hypoechoic lesion. The tumor, approximately 10 cm in diameter, consisted of groups of multiple nodules with cystic changes. Vascular signals were scanty.

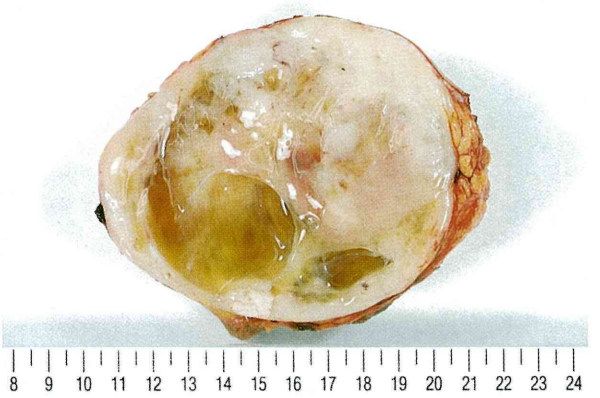

Fig. 2 A gross section of the surgical specimen showed a white-to-tan solid lesion for the most part, but the cystic lesion included transparent mucous fluid.

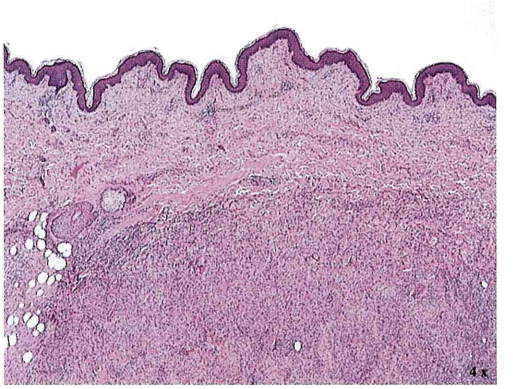

Fig. 3 Histopathological findings of the tumor. (H&E; x5) The tumor consisted of diffuse proliferation of spindle cells without specific arrangement mixed with inflammatory cells. Spindle cells infiltrated extensively, into breast grand ducts, fat tissue, and partially into the dermis.

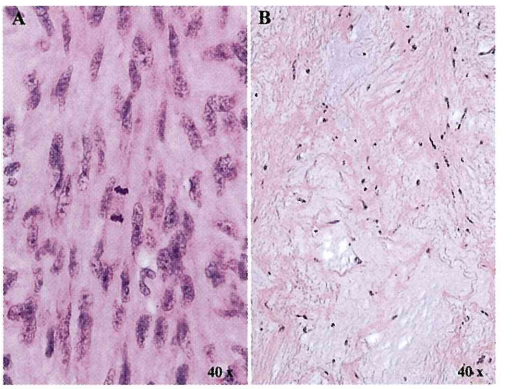

Fig. 4 Histopathological findings of the tumor. (H&E, x20) (A) Spindle cells have comparatively small nuclei. Nuclear pleomorphism, remarkable fission, and necrosis were absent. (B) The myxoid stroma lesion.

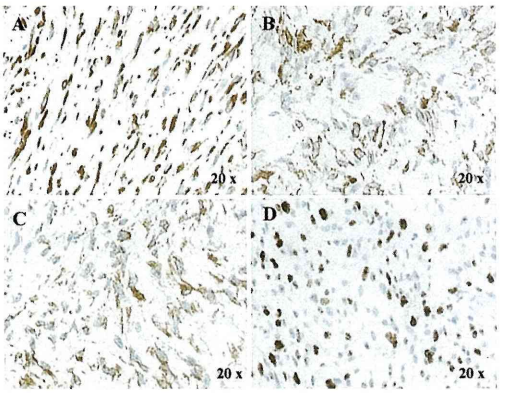

Fig. 5 Immunohistochemical staining findings of the tumor. It is positive reaction for vimentin (A) (x20) in diffuse, predominantly positive for α-smooth muscle actin (SMA) (B) (x20) and HHF-35 (C) (x20). These findings suggest differentiation of myofibroblasts. Furthermore, a large number of MIB-1 (D) (x20) -positive cells suggestive high proliferative activity.

inflammation. The mass had an irregular contour and was movable over the pectoral muscle. Axillary lymph nodes were not palpable.

Ultrasonography of the left breast showed a well-defined heterogeneous hypoechoic lesion. The tumor, approximately 10 cm in diameter, consisted of groups of nodules containing cystic changes. Some processes appeared in the tumor border. Vascular signals were scanty. (Fig. 1)

Magnetic Resonance Mammography (MRM) showed a well-defined round tumor without lobulation or spiculation. Membranes were enhanced strongly, but heterogeneous fluid appeared inside the tumor, suggesting a cystic malignant tumor.

Core needle biopsy was performed. Histopathological examination revealed the proliferation of spindle cells with no epithelial component. Those findings are suggestive of spindle cell tumor, for example phyllodes tumor. Surgical excision was considered advisable because of the possibility of malignancy. The tumor was excised and the postoperative course was uneventful.

Gross examination showed an oval solid mass, 10.5×8×6.5 cm in diameter. It was a well circumscribed firm gray-whitish elastic tumor completely encapsulated by a fibrous capsule. A gross section showed a white-to-tan solid lesion for the most part, but a cystic lesion including some transparent mucous fluid. (Fig. 2)

Histopathologically the lesion consisted of diffuse proliferation of spindle cells without specific arrangement in a myxoid stroma background. Spindle cells infiltrated extensively, into breast grand ducts, fat tissue, and partially into the dermis. (Fig. 3) Spindle cells have comparatively small nuclei. Nuclear pleomorphism, remarkable fission, and necrosis were absent. (Fig. 4)

Immunohistochemistry showed diffuse positive reaction for vimentin, predominantly positive reaction for α-smooth muscle actin (SMA) and HHF-35, and positive for desmin in part. On the other hand it was negative for pan-cytokeratin (AE1/AE3), S-100 protein, and CD34. These findings are suggested differentiation of myofibroblasts. Further more, a large number of MIB-1-positive cells suggest a high proliferative activity. (Fig. 5)

The final diagnosis was inflammatory myofibroblastic tumor (WHO classification), and careful pathological follow-up was advised.

Discussion

An IMT in a woman's breast enlarged rapidly, growing more than 5 cm in diameter within the previous year, and it was surgically excised. The tumor was pathologically examined and diagnosed as low-grade malignant spindle cell tumor.

A limited number of cases of IMT have been reported in the breast. The myofibroblast is the principal cell found in granulation tissue and in the desmoplastic stroma. (4) Inflammatory myofibroblastic tumor is defined as a distinctive lesion composed of myofibroblastic spindle cells accompanied by an inflammatory infiltrate of plasma cells, lymphocytes, and eosinophils. IMT is a heterogeneous clinicopathological entity that may occur at any anatomical location. IMT is classified as one of the mesenchymal tumors. (5) There is uncertainty as to whether IMT is reactive or neoplastic in nature. Some authors regard IMT as a low grade sarcoma.

Histopathologically the lesion consists of a proliferation of spindle cells with the morphological and immunohistochemical features of myofibroblasts, arranged in interlacing fascicles or in a haphazard fashion, and variably admixed with an inflammatory component of lymphocytes, plasma cells and histiocytes. (5)

The majority of myofibroblastomas are immunoreactive for vimentin, actin, and CD34. Many are also desmin positive. The tumors are not immunoreactive for cytokeratin or factor VIII and, only rarely, weakly reactive for S-100 protein. (6) In this case, the results of histopathological findings and immunohistochemical staining coincided well with the above-mentioned feature.

A few cases of plasma cell granuloma (inflammatory pseudotumor : IP) of the breast, aged 16 to 66, were reported. (7, 8, 9) All patients underwent tumor excision. In an analysis of extra-pulmonary IP, Coffin and colleagues found the most common mode of presentation to be a mass approximately 6 cm in diameter, but cases up to 17 cm have also been encountered. Good results have generally been obtained with surgical excision of the lesion, regardless of the site. There have been no cases of recurrence. (10) In the present case, the patient was a 59-year old woman, and the tumor size was 10.5×8×6.5 cm in diameter. This is compatible with the above reports.

The final diagnosis was inflammatory myofibroblastic tumor (WHO classification), and we are following her up at least once a year.

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