Case Report

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Myxoid Liposarcoma of the Breast Mimicking Phyllodes Tumor: A Case Report

엽상종양과 유사한 유방의 점액성 지방육종: 증례 보고

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Myxoid liposarcoma is an extremely rare malignant breast tumor. We report the case of a 44-year-old woman who had myxoid liposarcoma of the breast with a history of phyllodes tumor and describe the imaging findings on US, mammography, and MRI. Before surgery, the mass was considered to be a recurrent phyllodes tumor. However, using US, we retrospectively identified some differences between myxoid liposarcomas and phyllodes tumors.

Index terms Breast; Myxoid Liposarcoma; Phyllodes Tumor; Elastography

INTRODUCTION

Myxoid liposarcoma is a common soft tissue sarcoma that predominantly occurs in the extremities. However, it is a rare type of malignant breast tumor (1). Myxoid liposarcoma of the breast can develop as a primary liposarcoma or as a malignant stromal transformation from preexisting tumors such as phyllodes tumors (2). To the best of our knowledge, there have been a few reports of myxoid liposarcoma in the breast, which are mostly related to its pathologic findings (2, 3). Herein, we report the rare case of a 44-year-old woman who had myxoid liposarcoma of the breast with a history of phyllodes tumor and describe its imaging findings.

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CASE REPORT

A 44-year-old woman presented with a rapidly growing mass in her left breast. She had a wide excision of the left breast for borderline phyllodes tumors seven years earlier. An iU22 scanner (Philips Healthcare, Andover, MA, USA) was used for US which revealed a 3.6-cm oval, well-circumscribed, isoechoic mass in the left breast at the 11-o'clock position. Doppler US imaging revealed increased internal vascularity as well as vessels in the rim (Fig. 1A).

Upon this visit, a 6.2 cm × 5.8 cm oval and an equal-to-high density mass in the left upper central to upper outer quadrant of the breast, unobscured by overlying breast parenchyma and free of microcalcifications, were observed in standard mammography (Fig. 1B). An ML6-15 linear transducer (LOGIQE10 scanner; GE Healthcare, Waukesha, WI, USA) was used to perform a breast US, in which a 5.0-cm oval, well-circumscribed solid mass was observed in the left breast at the 12-o'clock position, adjacent to a previous excision scar. The mass was heterogeneous, with multiple faint echogenic lines. It was visible as a partially hazy and slightly hyperechoic lesion that blended in with the echogenic lines. There were no clefts, which are common radiologic features of cystic spaces with slit-like patterns. Color Doppler US imaging revealed mild peripheral vascularity. The mass was compressible and soft. Shear-wave elastography revealed homogeneous dark blue in color, with elasticity values ranging between 0 and 36 kPa (Fig. 1C). No suspicious lymph nodes were observed in the left axilla. Considering the high relapse rate, the recurrence of phyllodes tumors was suspected. As a result, we classified it as category 4B according to the Breast Imaging Reporting and Data System. Pathological examination of a US-guided 14-gauge core biopsy specimen demonstrated atypical spindle cell proliferation, which suggested the recurrence of a phyllodes tumor. Dynamic contrast-enhanced MRI revealed an 8.0 cm × 6.5 cm × 7.0 cm oval, well-circumscribed, and predominantly hypointense mass on T1-weighted imaging. The lesion showed hyperintensity on T2-weighted images. The first post-contrast acquisition and four repeated post-contrast images were acquired at 90- and 60-second intervals, respectively. In the early phase (90 seconds after the enhancement), they found heterogeneous peripheral enhancement and a type I persistent enhancement pattern on the time–signal intensity curve (Fig. 1D).

Fig. 1. A myxoid liposarcoma of the breast in a 44-year-old woman with a history of phyllodes tumor.
A. A phyllodes tumor was confirmed 7 years ago. Gray-scale US shows a 3.6-cm oval, well-circumscribed, isoechoic mass in the left breast at the 11 o'clock position. Doppler imaging shows increased internal and rim vascularity in the mass.

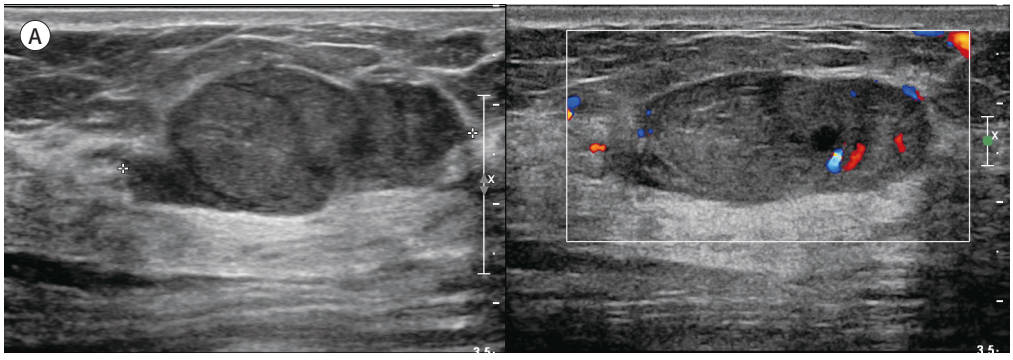

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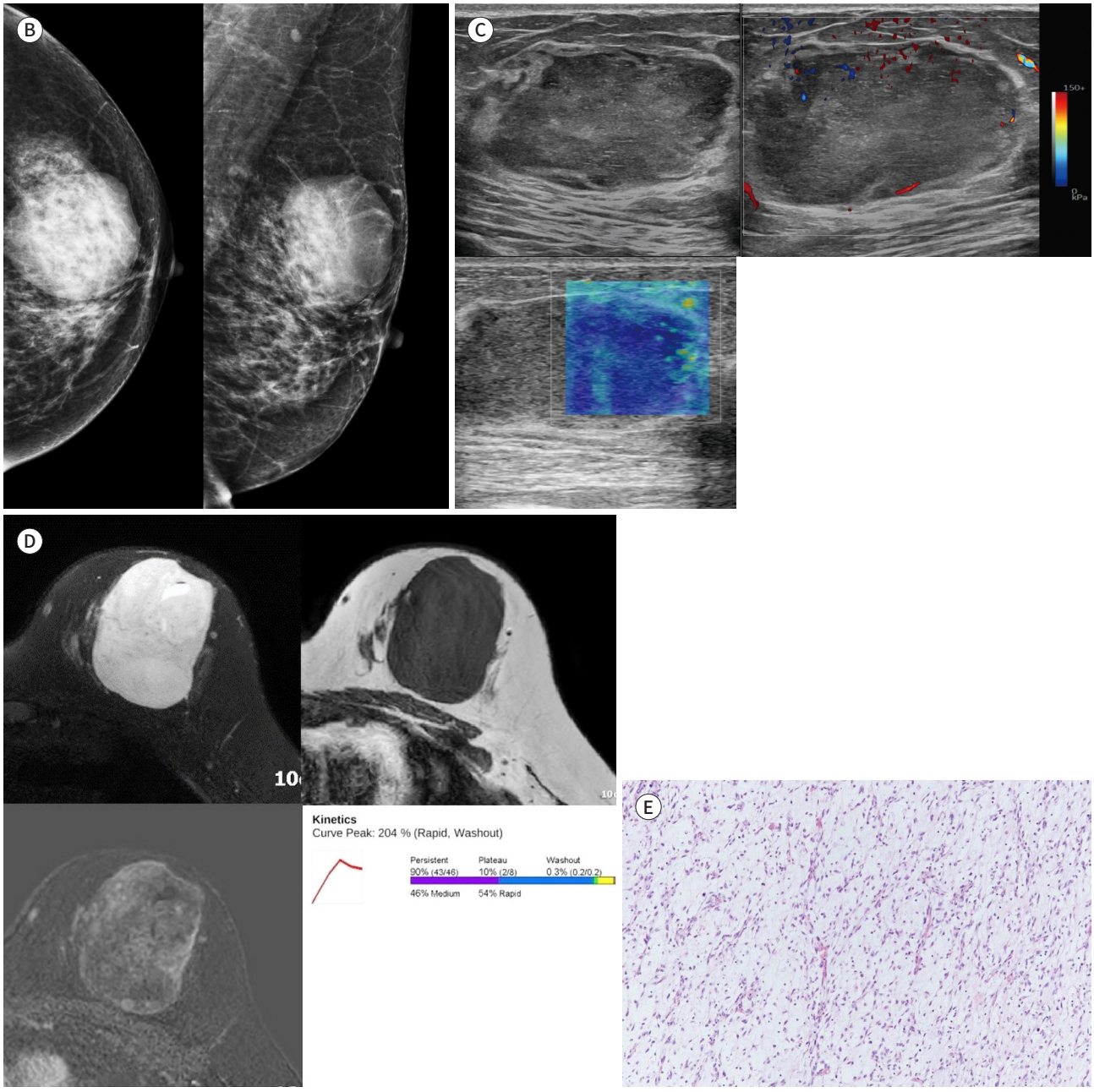
Fig. 1. A myxoid liposarcoma of the breast in a 44-year-old woman with a history of phyllodes tumor.
B. Upon this visit, craniocaudal and mediolateral oblique mammogram shows a 6.2 cm × 5.8 cm, oval, and equal-to-high density circumscribed mass on the left upper central to upper outer breast.
C. Gray-scale US shows a 5-cm oval and well-circumscribed heterogeneous solid mass at the 12 o'clock position of the left breast, close to a previous excision site. Color Doppler US shows mild peripheral vascularity. Shear wave elastography shows the soft elasticity of the mass.
D. T2-weighted spectral attenuated inversion recovery (left upper) shows the high-signal intensity of the lesion, and T1-weighted imaging (right upper) shows the low-signal intensity of the lesion on an MRI. In dynamic studies, the first post-contrast subtracted image (lower) demonstrates heterogeneous, gradual peripheral enhancement with a type-I persistent kinetic curve.
E. Microscopic examination shows that the tumor contains abundant myxoid stroma with a striking plexiform, delicate, and arborizing capillary network. The tumor shows patternless arrays of uniformly small and ovoid cells without morphological adipocyte differentiation (hematoxylin & eosin stain, × 100).



Subsequently, a partial mastectomy was performed to resect the lesion entirely. The resected mass was well encapsulated (7.5 cm × 7.0 cm × 5.0 cm in size). The cut surface showed a grayish, gelatinous mass. Microscopic examination revealed abundant myxoid stroma with a striking plexiform, delicate, and arborizing capillary network. The tumor showed patternless arrays of uniformly small and ovoid cells, without morphological adipocyte differentiation (Fig. 1E). The final pathological diagnosis was a low-grade myxoid liposarcoma of the breast. Therefore, re-excision was performed to ensure safe resection margins. The patient was referred to an oncologist and subsequently treated with adjuvant radiation therapy.

DISCUSSION

Liposarcoma rarely occurs in the breast, accounting for < 0.3% of all breast cancers (4, 5). There are five histological subtypes of liposarcomas, among which well-differentiated liposarcoma is the most common and myxoid liposarcoma is the second most common type. Histologically, myxoid liposarcoma comprises proliferating small, round to oval, monotonous mesenchymal cells and scattered lipoblasts in a prominent myxoid matrix with characteristic arborizing vessels. Unlike well-differentiated liposarcomas, myxoid liposarcomas have only a small amount of microscopic fat (< 10%) (1).

Myxoid liposarcoma can develop de novo or by malignant stromal transformation from preexisting tumors like phyllodes tumors (3, 5). Typical lipoblasts that are positive for the S-100 protein are key features for the diagnosis of liposarcoma, in which epithelial elements should not be seen (2). Contrastingly, phyllodes tumors consist of hypercellular stroma (spindle cells) underneath cleft-like spaces lined by benign glandular epithelium. In our case, a US-guided core biopsy showed atypical spindle cell proliferation, and the result was reported as a phyllodes tumor. The reason for this result was that only part of the mass was analyzed without including lipoblasts in the specimen.

No known risk factors were associated with primary breast sarcomas. However, secondary breast sarcomas can occur after radiation or in patients with chronic lymphedema (6). Our patient had no history of radiation or lymphedema, which are risk factors for secondary breast sarcomas. Additionally, although a myxoid liposarcoma occurred near the previous excision site of the phyllodes tumor, no typical leaf-like growth pattern was observed that indicates a phyllodes tumor. So, our case was considered to be a primary sarcoma rather than a malignant transformation from a preexisting phyllodes tumor.

Myxoid liposarcomas most frequently affect the lower extremities, where patients typically present with large, painless, and palpable masses (1). The radiologic findings of myxoid liposarcomas in other body parts were nonspecific. On US, a myxoid liposarcoma is a complex and well-defined hypoechoic solid mass. On an MRI, it is typically a well-defined, multilobulated mass with a hypointense signal on T1- and a hyperintense signal on T2-weighted images due to its high water content. A further variable enhancement pattern was observed on an MRI. Identifying adipose tissue is helpful for diagnosis, as 42%–78% of cases show the appearance of fatty content in myxoid liposarcoma (1).

However, myxoid liposarcoma of the breast has rarely been reported (4, 5). Even though several previous studies on myxoid liposarcomas of the breast have focused only on their

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pathological findings, very little is known about their imaging findings (2, 4). To the best of our knowledge, only one report has described the radiological findings of myxoid liposarcomas of the breast on US and MRI. Saito et al. (4) showed a lobulated mass with cystic changes and a hyperintense mass on US and T2- and diffusion-weighted images (DWI). These radiological features of myxoid liposarcomas in the breast are similar to those observed in other parts of the body. In our case, we observed an oval, well-circumscribed solid mass on US and T1-hypointense and T2-hyperintense masses on an MRI. We further discovered a persistent enhancement pattern on the dynamic MRI. Because its imaging features are nonspecific, distinguishing myxoid liposarcoma from other well-circumscribed solid masses, such as fibroadenomas or phyllodes tumors, is difficult.

Since the possibility of a phyllodes tumor was considered high based on the biopsy results and history of the patient, we retrospectively compared the imaging findings of our cases with those of cases with phyllodes tumors. We identified some distinctions in US findings between myxoid liposarcomas and phyllodes tumors. On US, phyllodes tumors generally manifest as complex cystic echogenicity, with or without a cleft, and are firm on elastography. Kim et al. (7) reported that median elasticity (E_{mean}) and maximum elasticity (E_{max}) were significantly higher for phyllodes tumors than for fibroadenomas (E_{mean}, 66.7 vs. 15.7 kPa; E_{max}, 76.7 vs. 21.0 kPa; $p < 0.01$). The exact quantitative values of elasticity for myxoid liposarcoma have not been reported, but Ohshika et al. (8) reported that mucinous malignant tumors, which include myxoid liposarcoma, had low elasticity. In our case, the mass was dark blue in color, with elasticity values ranging between 0 and 36 kPa. Therefore, myxoid liposarcoma of the breast revealed a very soft tumor on elastography and a relatively higher echogenicity than a phyllodes tumor on US. Myxoid liposarcomas do not contain clefts, which are characteristic features of phyllodes tumors, which is another distinguishing feature.

Although the detection of fatty components could suggest the diagnosis, in our case, no fat was apparent on an MRI. To date, there has been no research analyzing the features of myxoid liposarcoma of the breast compared to phyllodes tumors on an MRI. Yabuuchi et al. (9) showed that the apparent diffusion coefficient (ADC) decreased more in malignant phyllodes tumors than in benign phyllodes. Myxoid liposarcoma in the extremities also shows a lower ADC value than myxoma (10). Thus, both myxoid liposarcomas and phyllodes tumors are likely to have low ADC values. As we did not acquire DWI data, we could not compare the ADC values between myxoid liposarcoma and phyllodes tumors.

In summary, we report the relatively rare imaging findings of myxoid liposarcoma of the breast. We found that US can help differentiate myxoid liposarcomas from other benign breast tumors, especially phyllodes tumors. Even though it is an extremely rare tumor, the possibility of myxoid liposarcoma can be considered in patients with a history of phyllodes tumor if the lesion shows soft elasticity. This is because liposarcomatous differentiation can occur in phyllode tumors. Furthermore, an accurate histopathological examination should be performed to avoid misdiagnosis.

Author Contributions

Conceptualization, R.J.K.; data curation, L.S.J., R.J.K.; formal analysis, L.S.J., W.K.Y., H.S.; investigation, L.S.J.; supervision, R.J.K.; writing—original draft, L.S.J.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

1. Murphey MD, Arcara LK, Fanburg-Smith J. From the archives of the AFIP: imaging of musculoskeletal liposarcoma with radiologic-pathologic correlation. *Radiographics* 2005;25:1371-1395
2. Abdullah MA, Sain AHM, Ghani FA. Myxoid liposarcoma: a rare soft tissue tumour in the breast. *Malaysian J Med Health Sci* 2020;16:131-133
3. Kim JH, Lee JY. Malignant phyllodes tumor of the breast with liposarcomatous differentiation: a case report with imaging findings. *Radiol Case Rep* 2019;14:531-534
4. Saito T, Ryu M, Fukumura Y, Asahina M, Arakawa A, Nakai K, et al. A case of myxoid liposarcoma of the breast. *Int J Clin Exp Pathol* 2013;6:1432-1436
5. Kim JM, Jung EJ, Kim JY, Lee HS, An HJ, Park T, et al. A rare case of mixed type liposarcoma of breast arising in malignant phyllodes tumor. *Breast J* 2020;26:271-273
6. Li GZ, Raut CP, Hunt KK, Feng M, Chugh R. Breast sarcomas, phyllodes tumors, and desmoid tumors: epidemiology, diagnosis, staging, and histology-specific management considerations. *Am Soc Clin Oncol Educ Book* 2021;41:390-404
7. Kim GR, Choi JS, Han BK, Ko EY, Ko ES, Hahn SY. Combination of shear-wave elastography and color Doppler: feasible method to avoid unnecessary breast excision of fibroepithelial lesions diagnosed by core needle biopsy. *PLoS One* 2017;12:e0175380
8. Ohshika S, Saruga T, Ogawa T, Ono H, Ishibashi Y. Distinction between benign and malignant soft tissue tumors based on an ultrasonographic evaluation of vascularity and elasticity. *Oncol Lett* 2021;21:281
9. Yabuuchi H, Soeda H, Matsuo Y, Okafuji T, Eguchi T, Sakai S, et al. Phyllodes tumor of the breast: correlation between MR findings and histologic grade. *Radiology* 2006;241:702-709
10. Morán LM, Vega J, Gómez-León N, Royuela A. Myxomas and myxoid liposarcomas of the extremities: our preliminary findings in conventional, perfusion, and diffusion magnetic resonance. *Acta Radiol Open* 2022;11:20584601221131481

엽상종양과 유사한 유방의 점액성 지방육종: 증례 보고

이세진[1] · 류정규[1*] · 원규연[2] · 한상아[3]

유방에 발생하는 점액성 지방육종은 극히 드문 유방의 악성종양이다. 저자들은 엽상종양의 과거력이 있는 44세 여자 환자에서 발생한 유방 점액성 지방육종의 1예를 보고하고 유방촬영술, 초음파, 자기공명영상의 영상 소견을 보고하고자 한다. 종양은 수술 전 영상검사에서 엽상종양의 재발로 오인되었으나 후향적으로 비교했을 때 점액성 지방육종과 엽상종양의 감별에 도움이 될 수 있는 초음파 소견을 고찰하였다.

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