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Case report

Initial experience using ultrasound, automated breast volume scanner, and elastography for primary leiomyosarcoma of the breast: A rare case report

Fei-Yue Yang [a], Xiu Wang [b], Shao-Yi Lu [c], Yu-Jing Zhao [a,*], Wei-Wei Ren [d,e,f,**]

[a] Department of Medical Ultrasound, Shanghai Skin Disease Hospital, School of Medicine, Tongji University, Shanghai, 200443, China
[b] Department of Pathology, Shanghai Tenth People's Hospital, School of Medicine, Tongji University, Shanghai, 200443, China
[c] Department of Breast and Thyroid Surgery, Shanghai Tenth People's Hospital, School of Medicine, Tongji University, Shanghai, 200443, China
[d] Department of Medical Ultrasound, Shanghai Tenth People's Hospital, Shanghai, 200443, China
[e] Ultrasound Research and Education Institute, School of Medicine, Tongji University, Shanghai, 200443, China
[f] Shanghai Engineering Research Center of Ultrasound Diagnosis and Treatment, Shanghai, 200443, China

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ABSTRACT

Background: Primary leiomyosarcoma of the breast was a rare malignant tumor. Due to the extremely low morbidity and insufficient understanding of its imageological characteristics, there was a risk of misdiagnosis. In this case report, we presented the features of conventional US, elastography, automated breast volume scanner (ABVS), computed tomography (CT), and pathological findings of a case of primary leiomyosarcoma of the breast.

Case presentation: A 74-year-old woman detected a mass of the left breast by palpation. Both ultrasound and CT revealed a solid mass in the outer quadrant of the left breast. After admission, she underwent a modified radical unilateral mastectomy under general anesthesia (resection of the lesion with left breast reserved). Furthermore, the intraoperative frozen section revealed malignant spindle cells, and the postoperative histopathology revealed primary leiomyosarcoma of the breast. After discharge, the patient was generally in good condition after the procedure and was asked to seek medical treatment in the oncology department. Findings on various imaging examinations and clinical data were carefully evaluated. Additionally, we also reviewed the associated kinds of literature.

Conclusion: We reported the features of conventional US, elastography, ABVS, CT, and pathological findings of a rare case of primary leiomyosarcoma of the breast. Overall, our findings indicated that the above-mentioned features generally predict malignancy. However, compared to other malignant breast lesions, the features of this case were not specific enough.

* Corresponding author. Department of Medical Ultrasound, Shanghai Skin Disease Hospital, Tongji University School of Medicine, No. 1278 Baode Road, Shanghai, 200443, China.

** Corresponding author. Department of Medical Ultrasound, Shanghai Tenth People's Hospital, Tongji University School of Medicine, No. 301, Middle Yanchang Road, Shanghai, 200072, China.

E-mail addresses: zhaoyujing6551@163.com (Y.-J. Zhao), rww9456@163.com (W.-W. Ren).

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1. Introduction

Leiomyosarcoma displaying the features of smooth-muscle differentiation had occurred in numerous anatomical sites such as the uterus and gastrointestinal system. Primary leiomyosarcoma of the breast was a rare tumor, accounting for less than 1 % of all breast cancers. Therefore, it was challenging to diagnose the primary leiomyosarcoma of the breast [1–3].

Although the radiological evaluation was performed in a few cases, no case reports focused on some new modalities like elastography and ABVS features of the primary leiomyosarcoma of the breast. In this case report, we presented a patient with primary leiomyosarcoma of the breast in a 73-year-old woman. The clinical and radiological features, pathological diagnosis, and treatment of primary leiomyosarcoma of the breast were reported. The associated kinds of literature were also reviewed.

2. Case report

A 73-year-old female patient was referred to the university hospital for further evaluation due to a lump in her left breast detected by palpation on December 15, 2020. She was otherwise a healthy woman. Physical examination revealed a 2.5cm elastic-hard, well-circumscribed, mobile mass in the outer quadrant of the left breast. However, there were no signs of skin retraction, nipple discharge, or palpable regional lymph node. The right breast was negative. There was no relevant medical history of breast cancer. Afterward, she accepted conventional US, elastography, ABVS, and CT examinations in our hospital on December 16, 2020. Due to the patient's preference, preoperative pathological biopsy and magnetic resonance imaging (MRI) were not performed. Subsequently, she received a modified radical unilateral mastectomy under general anesthesia (resection of malignant breast tumor with left breast reserved) after a thorough evaluation by the Thyroid and Breast Surgeon on December 17, 2020. Besides, the patient underwent intraoperative and postoperative pathological examinations, including intraoperative freeze examination on December 17, 2020, postoperative histopathology on December 21, 2020, and immunohistochemical examination on December 24, 2020. The relevant examinations were reviewed as follows.

2.1. Imaging findings

2.1.1. Conventional US

Conventional US was performed with a LOGIQ E9 scanner (GE Healthcare, Milwaukee, WI, USA) equipped with a 9–14MHz linear array transducer. The ultrasonographic showed a 2.5 cm, oval, hypoechoic mass in the fat layer at the 3 o'clock position in the fat layer (Fig. 1 a-b). Color Doppler imaging showed that the vessels were relatively large inside and around the mass (Fig. 1 c-d). The lesion was classified as Breast Imaging Reporting and Data System (BI-RADS) category 4a [4]. A tissue diagnosis was recommended.

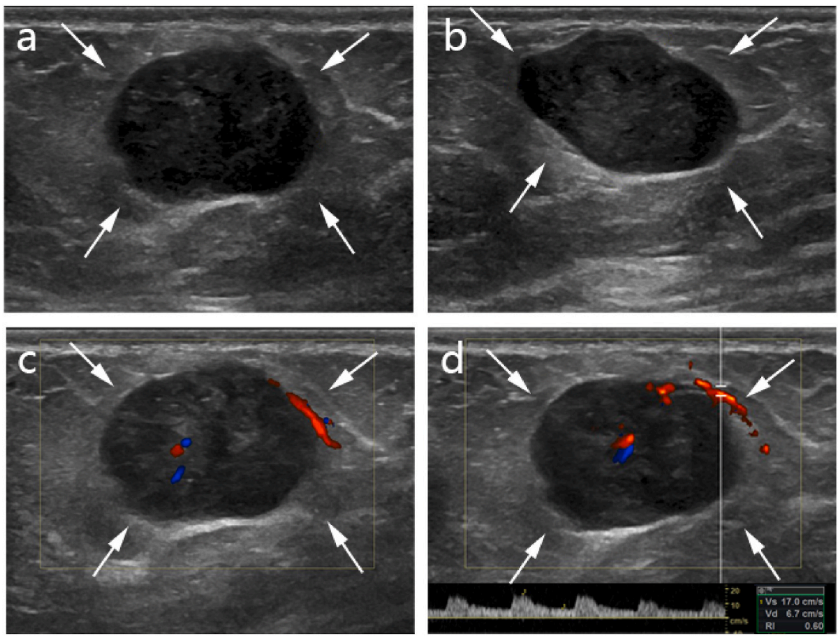

Fig. 1. (a, b) Gray-scale US showed a hypoechoic and well-defined lesion (white arrow) measuring 2.5cm of the breast, showing posterior acoustic enhancement. (c) Color Doppler imaging showed the vessels were relatively large inside and around the lesion (white arrow). (d) Pulsed wave Doppler imaging indicated the Vmax is 17cm/s, and RI is 0.60. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

2.1.2. Elastography

Elastography was performed with the Aixplorer US system (Super-Sonic Imagine, Aix-en-Provence, France), equipped with a linear array transducer of 4–15 MHz. The shear wave velocity map was presented as a color map (higher, red; intermediate, yellow, or green; lower, blue), respectively. The built-in region-of-interest (ROI) (Q-box; Super-Sonic Imagine) of the system was set to include the lesion and the surrounding normal tissue, which demonstrated a semitransparent color map of a tissue stiffness overlaid B-mode image [5]. We fixed a ROI with a diameter of 16mm over the stiffest part of the lesion, including the immediate adjacent stiff tissue or halo. The system calculated the minimum (E_{min}), maximum (E_{max}), mean (E_{mean}), and standard deviation (ESD) elasticity values in kPa for the mass. Firstly, ESD is a parameter that can show the heterogeneity of stiffness distribution [6], while E_{max} shows the stiffness of the stiffest part of the lesion, and E_{mean} indicates the average stiffness. The higher the ESD is, the more suspicious the lesion is. Shear-wave elastography (SWE) showed the lesion and surrounding tissue as heterogeneous, ranging from blue to yellow, with a halo around it. The ROI's E_{max}, E_{min}, E_{mean}, and ESD on SWE were 130.2 kPa, 0.1 kPa, 35.9 kPa, and 30.1 kPa, respectively. The measurable stiffness values of this lesion and its surrounding tissue were heterogeneous. Besides, there was a large area of color defect on the elastography (Fig. 2).

2.1.3. ABVS

ABVS was performed with an ACUSON Oxana 2 automated breast volume scanner (Siemens Medical Solutions, Mountain View, CA, USA) equipped with a 5–14MHz BV 3D transducer. Three-dimensional visualization of the lesion was shown on a US image from the sagittal, axial, and coronal views (Fig. 3 a-c). The sagittal view was achieved by screening directly, and the integrated software calculated the coronal and axial views. From the axial and coronal views, the position and distance of the lesion and papilla were intuitive. The coronal plane was unique to ABVS and unavailable for conventional ultrasound. It provided additional information for breast lesion detection and diagnosis [7]. The lesion had a circumscribed margin with no disruption of the surrounding tissue. There was no retraction phenomenon in the coronal plane in this case.

2.1.4. CT

A chest CT scan was conducted on a dual-source CT scanner (Somatom Definition Flash, Siemens Healthcare. Forchheim, Germany). The staging CT showed a middle density of space-occupying lesion found in the outer quadrant of the left breast, with a defined boundary (Fig. 4). The CT value was about 27 HU.

2.2. Pathological findings

On gross pathology, the lesion was border-clear and reddish, and its maximal dimension was 3 cm. Intraoperative frozen section examination of the lesion revealed malignant spindle cells. Therefore, she received a modified radical unilateral mastectomy under general anesthesia (resection of malignant breast tumor with left breast reserved).

On microscopic pathology, the tumor comprised spindle cells with very pleomorphic nuclei. It showed homogeneity without

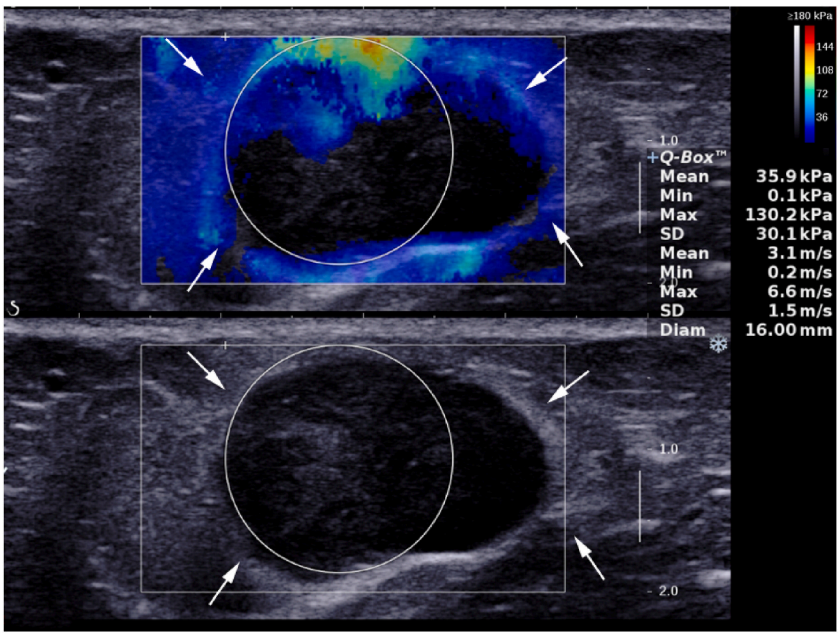

Fig. 2. In the lesion, the stiffness values (white arrow) of the region of interest (ROI) heterogeneously ranged from 0.1 kPa (0.2 m/s) to 130.2 kPa (6.6 m/s).

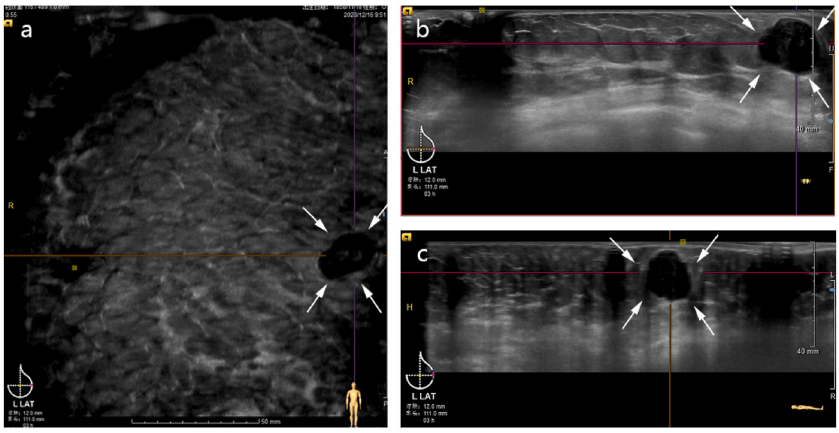

Fig. 3. Three-dimensional visualization of the lesion (white arrow). (a) The coronal view was calculated by the integrated software. (b) The sagittal view was achieved by screening directly. (c)The axial view was calculated by the integrated software.

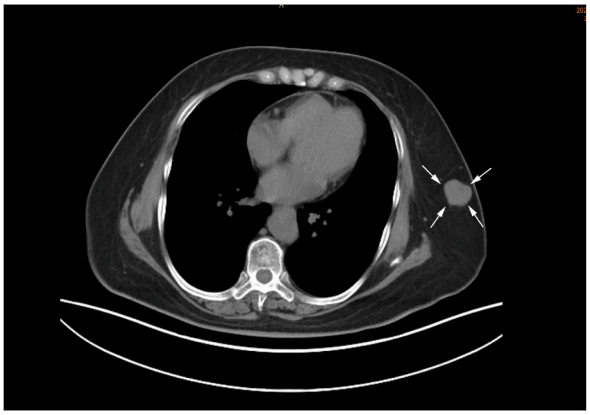

Fig. 4. A staging CT demonstrated a space-occupying lesion (white arrow) with a defined boundary in the outer quadrant of the left breast.

necrosis or calcification. There was no epithelial or ductal component. Mitotic activities were observed as more than 5 per 10 high power fields (Fig. 5 a-d).

Immunohistochemical staining showed that neoplastic cells were positive for smooth muscle actin (SMA), vimentin, desmin, calponin, and CD34, while they were negative for C-kit, S-100, hmb45, melan-a, myod1, myogenin, CD10, and CD68. Ki-67 was positive in up to 15 % of the cells. These immunohistochemical analyses helped confirm the diagnosis of leiomyosarcoma.

The tumor was treated with wide local excision. There was no residual disease in the incisal edge specimen. The metastatic examination was negative, including chest radiographs, tumor markers, and so on. Accordingly, the tumor was confirmed as a primary leiomyosarcoma of the breast.

According to the doctor's advice, the patient went to the oncology department and received chemotherapy. Four courses of chemotherapy were administered, starting on January 6, 2021, January 30, 2021, February 21, 2021, and March 16, 2021, respectively. Each course lasted three days, with 50 mg epirubicin and 2 g ifosfamide on day one, 40 mg of epirubicin and 2 g ifosfamide on day two, and only 2 g ifosfamide on day three.

Nevertheless, the patient did not complain of discomfort, and there were no adverse and unanticipated events during the treatment. The patient had been followed up for three years, and no recurrence or metastasis had been detected.

3. Discussion

Leiomyosarcoma occurred most frequently in the uterus, retroperitoneum, subcutaneous tissues, and gastrointestinal tract but rarely in the breast [5,6]. To our knowledge, a total of 22 primary leiomyosarcomas (mean size, 4.95 cm ± 3.68; range, 0.5–15 cm) of the breast in 22 patients (mean age, 49.13 years ± 15.47; age range 18–73 years) referred to ultrasound had been reported in world literature (Table 1). It had been reported that most primary leiomyosarcoma of the breast occurs in female patients with a palpable mass. Some tumors might be misdiagnosed as fibroadenoma because imaging features tended to be benign. Therefore, the final diagnosis was often dependent on postoperative pathology.

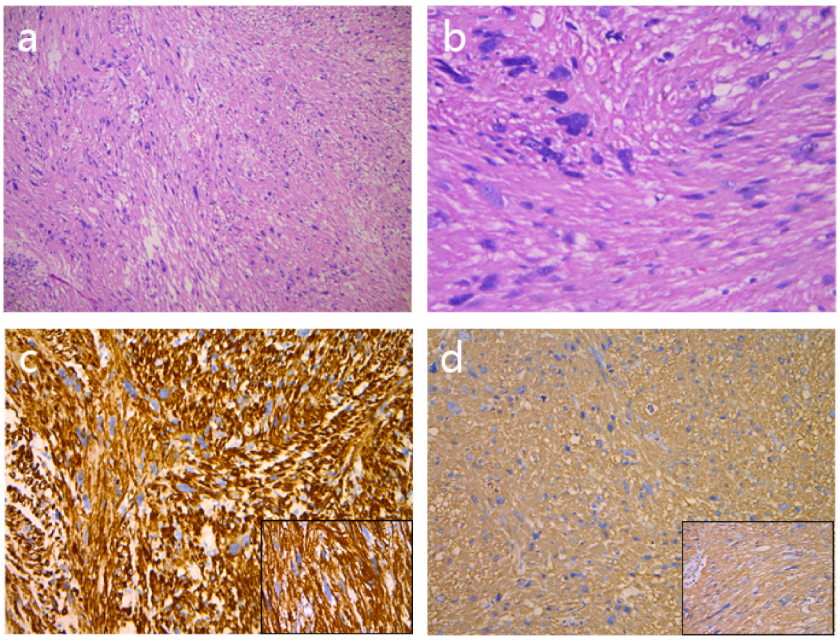

Fig. 5. (a) Pathological examination confirmed the diagnosis of leiomyosarcoma of the breast (hematoxylin-eosin stain; original magnification, × 100). (b) Pathological examination confirmed the diagnosis of leiomyosarcoma of the breast (hematoxylin-eosin stain; original magnification, × 400). (c) Immunohistochemical staining confirmed smooth muscle actin (SMA) was positive. (d) Immunohistochemical staining confirmed desmin was positive.

The diagnosis of breast leiomyosarcoma was mainly based on imaging and needle biopsy. In imaging, leiomyosarcoma was frequently presented as a significant and phyllode-shaped mass.

They were conventionally treated by wide local excision or simple mastectomy, modified radical mastectomy, or radical mastectomy, whereas radiotherapy and chemotherapy had proven debatable [10,24,27,30]. No positive lymph node in the primary leiomyosarcoma of the breast was reported before. However, local recurrence and hematogenous metastasis existed [1,3].

Overall, breast leiomyosarcoma was considered to have a poor prognosis. However, only three metastasis cases had been reported, including two metastases to the lung and one metastasis to the femur. This might be associated with the limited number of cases and short follow-up time, while the longest follow-up time of previous studies was only up to 60 months.

In summary, a typical case of breast leiomyosarcoma was reported. The patient only detected a non-specific palpable mass. The tumor was treated with wide local excision, and the diagnosis was confirmed by postoperative pathology. To our knowledge, this was the first case where elastography and ABVS were performed on primary leiomyosarcomas of the breast. It should be noted that previous studies showed that benign lesions tended to be homogeneous and soft (80kPa or less) on SWE, whereas malignancies appeared heterogeneous and stiff [31,32]. Accordingly, the present breast leiomyosarcoma was heterogeneous in stiffness and had a large-area of color defect on the elastography, suggesting an extremely high or low stiffness within the lesion. Qualitatively, the lesion was consistent with malignancy.

However, based on our case, the features of breast leiomyosarcoma were nonspecific in conventional US and ABVS. Besides, the elastography results might not be potential features of the primary leiomyosarcoma of the breast, affected by variable factors such as excessive pressure applied during the elastography procedure or improper adjustment of the elastography range. It was challenging to distinguish breast leiomyosarcoma from other malignant breast lesions by elastography alone. Thus, we believed their value was limited for diagnosing. In addition, this was a single case report, so the result tended to be only descriptive. In the future, we need more clinical cases to confirm our results. Therefore, elastography and other imaging examinations were still auxiliary diagnostic means for primary breast leiomyosarcoma, and pathological biopsy was still an irreplaceable gold standard in the diagnosis of primary breast leiomyosarcoma.

4. Conclusion

We reported the features of conventional US, elastography, ABVS, CT, and Pathological findings of a rare case of primary leiomyosarcoma of the breast. Overall, our findings indicated that the above-mentioned features generally predict malignancy. However, compared to other malignant breast lesions, the features of this case were not specific enough.

Table 1
22 primary leiomyosarcomas reported in world literature.

Case	Author/year	Location	Age/ Sex	Size (cm)	Ultrasound features	Mitosis (/10Hpf)	Treatment	Follow up
1	Szekely et al., /2001 [8]	Right/lower inner quadrant	73/F	4.8	Lobulated	20–22	Simple mastectomy	Alive, 12 months
2	Hussien et al., /2001 [9]	Right/lower inner quadrant	49/F	2	Solid, non-homogeneous, irregular aspect ratio > 1, acoustic shadowing	12	Wide local excision	Alive, 18 months
3	Kusama et al., /2002 [10]	Left/upper outer quadrant	55/F	1.8	Hypoechoic, heterogeneous, irregular, rough border, irregular boundary echo	10	Modified radical mastectomy + Chemotherapy	Metastasis to the lung, 12 months
4	Shinto et al., /2002 [11]	Left/lower outer quadrant	59/F	12	Phyllode-shaped, heterogeneous, hypoechoic, clear margins	19	Simple mastectomy	Alive, 8 months
5	Liang et al., /2003 [12]	Left/upper outer quadrant	25/F	4	Lobulated, solid	5	Wide local excision	Alive, 32 months
6	Munitiz et al., /2004 [13]	Right/upper outer quadrant	58/F	4	hypo-echogenic gap, poorly defined contours, posterior reinforcement	14	Modified radical mastectomy	Alive, 12 months
7	De la Pena et al., /2008 [14]	Left/upper outer quadrant	50/F	3.2	Hypoechoic	–	Simple mastectomy	Alive, 11 months
8	Wong et al., /2008 [15]	Left/nipple area	52/F	1.5	Lobulated	7	Simple mastectomy	Alive, 4 days
9	Cobanoglu et al., /2009 [16]	Left/upper outer quadrant	64/F	3.5	Hypoechoic	12	Modified radical mastectomy	Alive, 22 months
10	Boehm et al., /2010 [17]	Right/central localization	62/ M	4.4	Hypoechoic solid mass	40	Modified radical mastectomy	Alive, 24 months
11	Masannat et al., /2010 [18]	Right/ underneath the nipple	59/ M	2	Benign looking mass	–	Simple mastectomy	Alive, 26 months
12	Fujita et al., /2011 [19]	Right	18/F	7.2	Lobulated	10	Simple mastectomy + radiotherapy	Alive, 60 months
13	Nagao et al., /2011 [20]	Right/upper outer quadrant	61/F	2.2	Well-circumscribed, hypoechoic mass, heterogeneous internal echo, clear margins, acoustic shadowing	9	Wide local excision	Alive, 18 months
14	Ulhas Rane et al.,/2012 [21]	Left	19/F	7	Well-circumscribed, oval	20–25	Wide local excision	Alive, 36 months
15	Karabulut et al., /2012 [22]	Right/upper and lower outer quadrants	48/F	10	Well-circumscribed	–	Radical mastectomy + axillary lymph node dissection	Alive, 1 month
16	Yener et al., /2013 [23]	Left/lower quadrant	44/F	3.5	Lobulated, hypoechoic, heterogeneous, an irregular shape, rough border, an irregular boundary echo	Few	Excision + radiotherapy	Alive, 12 months
17	Amaadour et al., /2013 [24]	Right	44/F	9.2	hypoechoic and heterogeneous	6	Chemotherapy	Died, 1 month
18	Sokolovskaya et al., /2014 [25]	Right	58/F	15	Heterogeneous, primarily hypoechoic, lobulated	–	Modified radical mastectomy	Metastasis to the femur, 24 months
19	Tajima et al., /2015 [26]	Left	50/F	4.8	Cystic appearance	6	Simple Mastectomy	Alive, 6 months
20	Kim et al., /2015 [27]	Right/upper outer quadrant	51/F	0.5	Oval, hypoechoic mass with no vascularity in the subcutaneous fat layer	15	Excision + chemotherapy	Alive, 60months
21	Testori et al., /2017 [28]	Left	62/F	3.5	Solid, highly vascularized, oval shape, well-defined margins, slightly heterogeneous echostructure. Extended into the overlying subcutaneous fatty tissue	5	Excision + radiotherapy	–
22	Amberger et al., /2018 [29]	Left/lower quadrant	20/F	3	Likely hematoma	–	Simple mastectomy + axillary lymph node dissection	Metastasis to the lung, 32 months

5. Limitation

This was a single case report, so the result tended to be only descriptive. In the future, we need more clinical cases to confirm our results. In addition, the patient refused to receive an MRI examination and preoperative biopsy. Thus this study lacked MRI imaging data and preoperative pathological diagnosis results. Owing to potential resemblances in imaging characteristics, the convergence of elastography properties, and the diverse morphological presentations, it was challenging to distinguish primary breast leiomyosarcoma, breast fibroadenoma, and breast cancer.

Informed consent

Informed consent was acquired from the patient, and the patient consented to the publishing of all images, clinical data, and other data included in the manuscript.

Data availability statement

Data associated with this study has not been deposited into a publicly available repository. Due to the nature of this case report, participants of this article do not agree for their data to be shared publicly. Therefore, supporting data is not available.

CRediT authorship contribution statement

Fei-Yue Yang: Writing – review & editing, Writing – original draft. **Xiu Wang:** Investigation. **Shao-Yi Lu:** Methodology. **Yu-Jing Zhao:** Resources. **Wei-Wei Ren:** Resources.

Declaration of competing interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: Yujing Zhao reports writing assistance was provided by Shanghai Anticancer Association. Yujing Zhao reports a relationship with Shanghai Anticancer Association that includes: employment and non-financial support. None has patent none pending to none. none If there are other authors, they declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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