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CASE REPORT

Can Epithelial-Myoepithelial Carcinoma of the Breast Benefit from TROP2 Antibody-Drug Conjugate?

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Abstract: Epithelial-myoepithelial carcinoma (EMC) of the breast is a rare biphasic tumor composed of intermixed malignant epithelial and myoepithelial components. Breast epithelial myoepithelial carcinoma lacks therapeutic strategies due to its rarity, and currently local treatment is still the main treatment. Herein we report an epithelial-myoepithelial carcinoma of the breast in a 33-year-old woman undergoing breast conserving surgery, sentinel lymph node biopsy, adjuvant chemotherapy and radiotherapy, with rapid liver and lung metastasis. After radiofrequency ablation therapy for metastatic lesions, vinorelbine plus cisplatin and TROP2 antibody-drug conjugate (ADC) treatment were given successively. After a brief improvement, drug resistance developed and the disease progressed. The patient died with the PFS 5.6 months.

Keywords: epithelial-myoepithelial carcinoma, breast cancer, TROP2 antibody-drug conjugate, malignant epithelial myoepithelial carcinoma, treatment

Case Report

A 33-year-old female patient came to our department in July 2021 for finding her right breast mass one month ago. One month ago, she inadvertently palpated a right breast mass, which was 1.0 cm in size located next to the sternum, with no pain and no nipple discharge. Recently, she found the mass had increased to 2.0 cm, so she came to hospital. Past history: In 2020, the patient underwent a right breast tumor resection surgery in other hospital, and the pathology was intraductal papilloma with localized atypical hyperplasia. A right quadrant lumpectomy was performed in January 2021 for a right breast mass (Figure 1A), and pathology revealed an intraductal papilloma with localized atypical hyperplasia. Physical examination showed: a 5.0 cm long arc-shaped scar was visible in the right breast, and a 2.0 cm × 1.8 cm, tough, border clear mass was in the right breast margin next to the sternum. Ultrasound (Figure 1B) mammography (Figure 1C and D) showed the right breast mass with still clear borders. A right breast mass excision was performed, and pathology (Figure 2) indicated: malignant epithelial-myoepithelial tumor of the breast. Immunohistochemistry (IHC) shows negative expression of ER, PR, Calponin, while positive expression of CK5/6, P63, Ki67 index was about 50%, P53 index was about 5%. A further breast MRI (Figure 3) showed residual breast tissue had suspicious lesion, and PET/CT showed no distant metastasis. After the multi-disciplinary treatment (MDT), right breast-conserving surgery and sentinel lymph node biopsy were performed. Intraoperative frozen pathology showed intraductal papilloma visible at the sternum margin, so the medial excision was extended to the skin. No mutation was detected by *BRCA1/2* gene test, and goserelin was given for ovarian protection. AC-T regimen, doxorubicin liposomal (35 mg/m^2 on day 1, triweekly), cyclophosphamide (600 mg/m^2 on day 1, triweekly) for 4 cycles, and nab-paclitaxel (260 mg/m^2 on day 1, triweekly) for 4 cycles and radiotherapy (whole breast irradiation and tumor bed boosts) were carried. A follow up in June 2022 revealed lung

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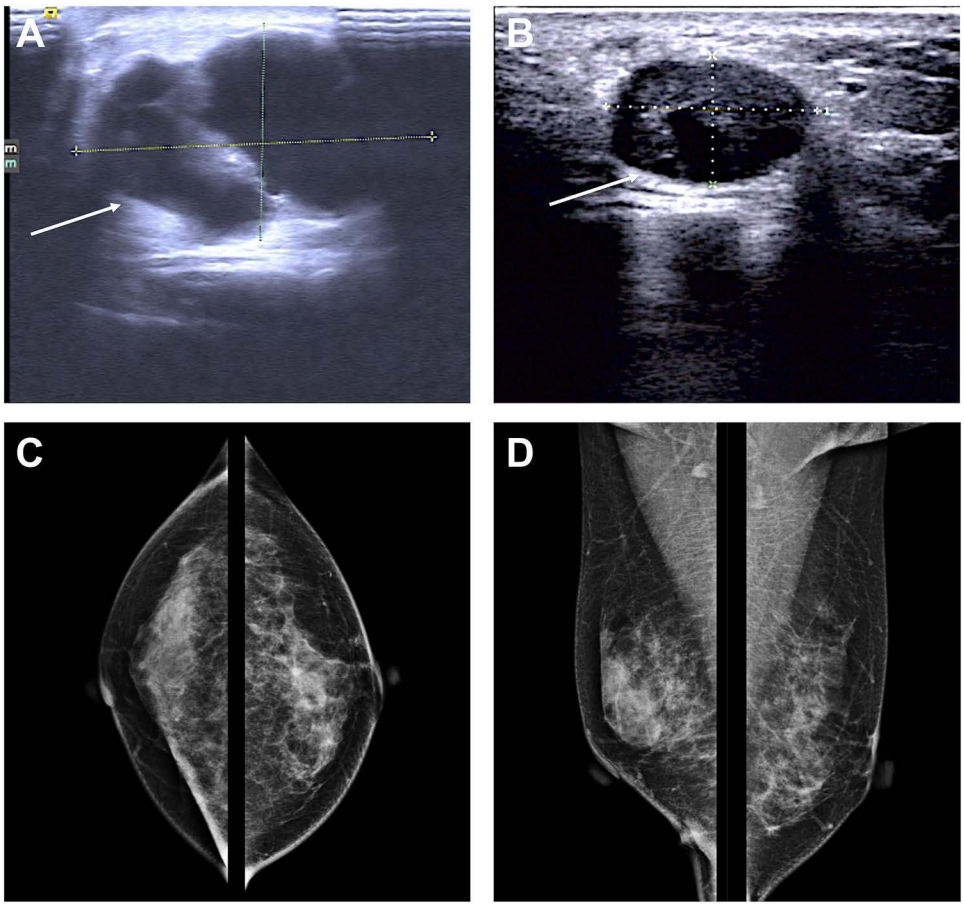

Figure 1 Breast ultrasound and mammography images.
Notes: (**A**) Ultrasound image of the right breast lesion initially detected in January 2021: the right breast lesion showed mixed cystic and solid echoes with clear boundaries, regular morphology, size 4.02 × 2.45 cm, consistent with BI-RADS 4A. (**B**) Ultrasound image of the right breast lesion in July 2021: the right breast lesion had clear boundaries, regular morphology, and internal echoes homogeneous, size 2.26 × 1.69 cm, conforming to BI-RADS 4A. (**C**) CC position of the mammography images in June 2021. (**D**) MLO position of the mammography images in June 2021. The white arrows are the right breast lesion.

metastases (Figures 4A, B and 5B) and liver metastases (Figures 4C, D and 6A) and a review of the lung CT in December 2021 showed the lesion in the upper lobe of the right lung already existence (Figure 5A). Ultrasound-guided core needle biopsy of the liver lesion and radiofrequency ablation of the liver lesion under ultrasonography (power of 35 W, 16 min) (Figure 4F), CT-guided core needle biopsy of the lung lesion and microwave radiofrequency ablation (power of 50 W, 3 min) were carried (Figure 4C). The pathology (Figure 7) showed that lung tissue: metastatic epithelial myoepithelial carcinoma, liver tissue: metastatic epithelial myoepithelial carcinoma. IHC shows negative expression of CK20, ER, PR, Villin, TTF1, Pax8, CD34, while strongly positive expression of CK7, CK, CK5/6, P53 index was about 10%, Ki67 index was about 50%. The image that one month after radiofrequency ablation was shown in the Figure 5C. NP regimen, cisplatin (75 mg/m^2 on day 1, triweekly) and vinorelbine (35 mg/m^2 on day 1 and 8, triweekly) chemotherapy was performed, RESICT 1.1 assessment of SD (Figures 5D and 6B). Progression of lung and liver lesions and enlargement of breast lesions were occurred after 5 cycle chemotherapy (Figures 5E and 6C), with PFS 4 months. Core needle biopsy of breast upper outer quadrant mass was performed and the pathology also showed malignant adenomyoepithelioma, infiltration of adipose tissue. IHC shows negative expression of ER, PR, Her-2, Calponin, S-100, SOX10, WT-1, Pax-8, while positive expression of CK5/6, p63, P40, Vimentin, CK8, Ki67 index was about 40%. The patient joined OptiTROP-Breast01 (NCT05347134) clinical trial on 14 November 2022 with the drug TROP2 ADC (SKB264, 340 mg on day 1 and 14, every 28 days), with RECIST 1.1 assessment SD after 2 cycles chemotherapy (Figures 5F and 6D), SD after 4 cycles (Figure 6E) and PD after 6 cycles (Figure 6F), with PFS 5.6 months. The patient died on 27 June 2023, the summary listed in the Figure 8.

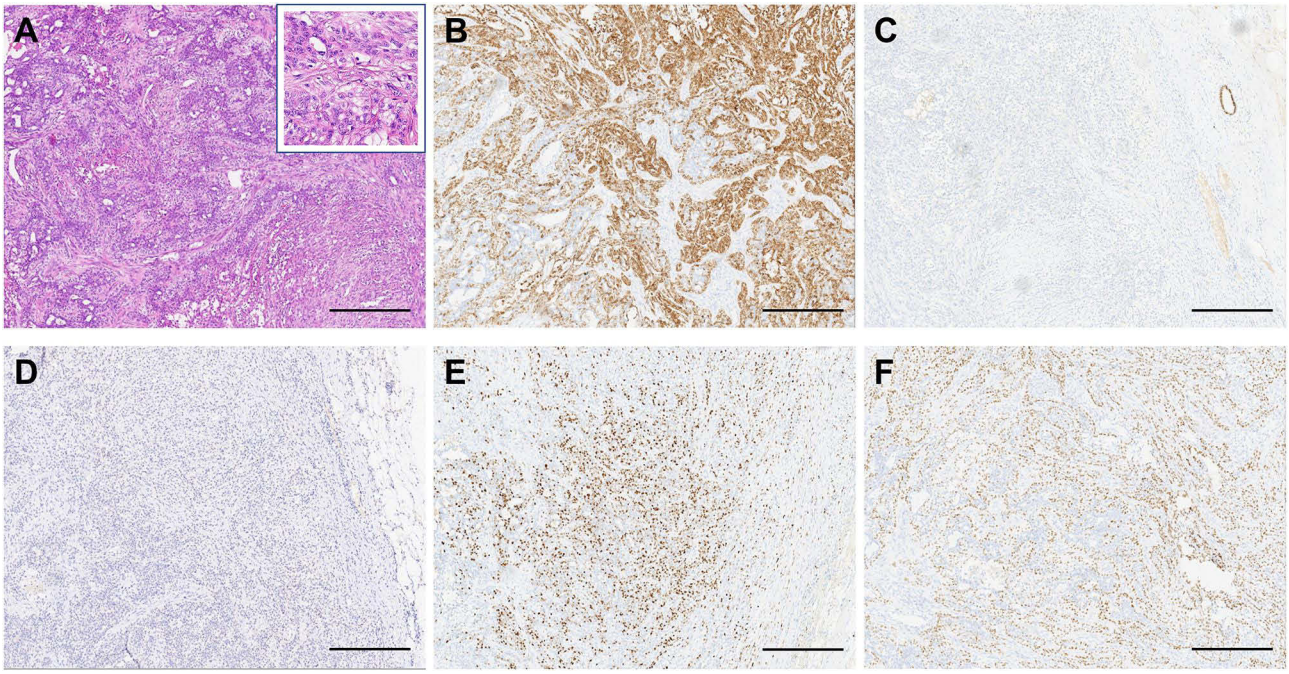

Figure 2 Pathology of MEMC (×40).
Notes: (**A**) H&E section shows obvious hyperplasia of glandular epithelium and myoepithelium, atypical hyperplasia of some cells, visible mitotic figures, occasional focal necrosis. The small figure in the upper right corner is a local zoom-in (×200). (**B**) IHC of CK5/6 indicates positive. (**C**) IHC of ER indicates negative. (**D**) IHC of PR indicates negative. (**E**) IHC of Ki67 indicates nearly 50% positive. (**F**) IHC of P63 indicates positive. (Scale bar: 400 μm).

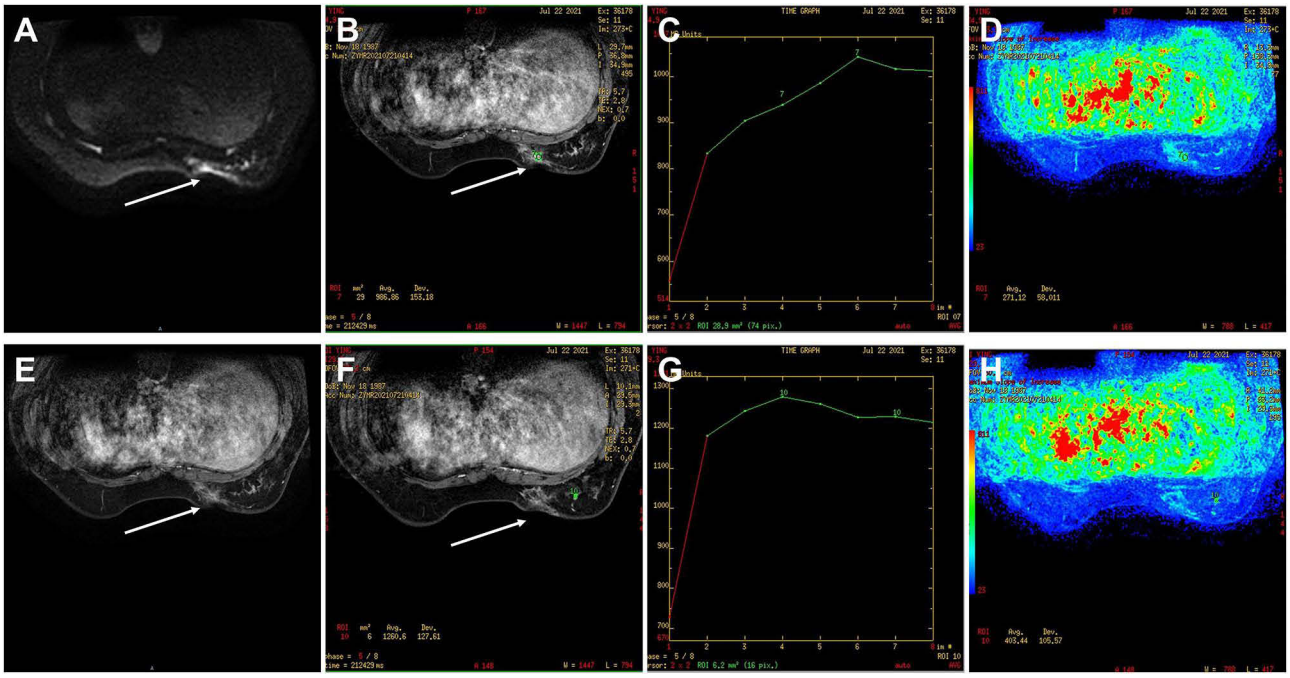

Figure 3 Breast MRI after lesion excision in July 2021.
Notes: (**A**) DWI image shows visible high signal in the operated and posterior nipple areas. (**B**) MR enhancement scanning image of target area 7. (**C**) Enhancement curve image of target area 7. (**D**) MR enhancement pseudo-color image of target area 7. (**E**) Arterial phase visualization of breast MR enhancement scan. (**F**) MR enhancement scanning image of target area 10. (**G**) Enhancement curve image of target area 10. (**H**) MR enhancement pseudo-color image of target area 10. The white arrows the residual breast suspicious lesion.

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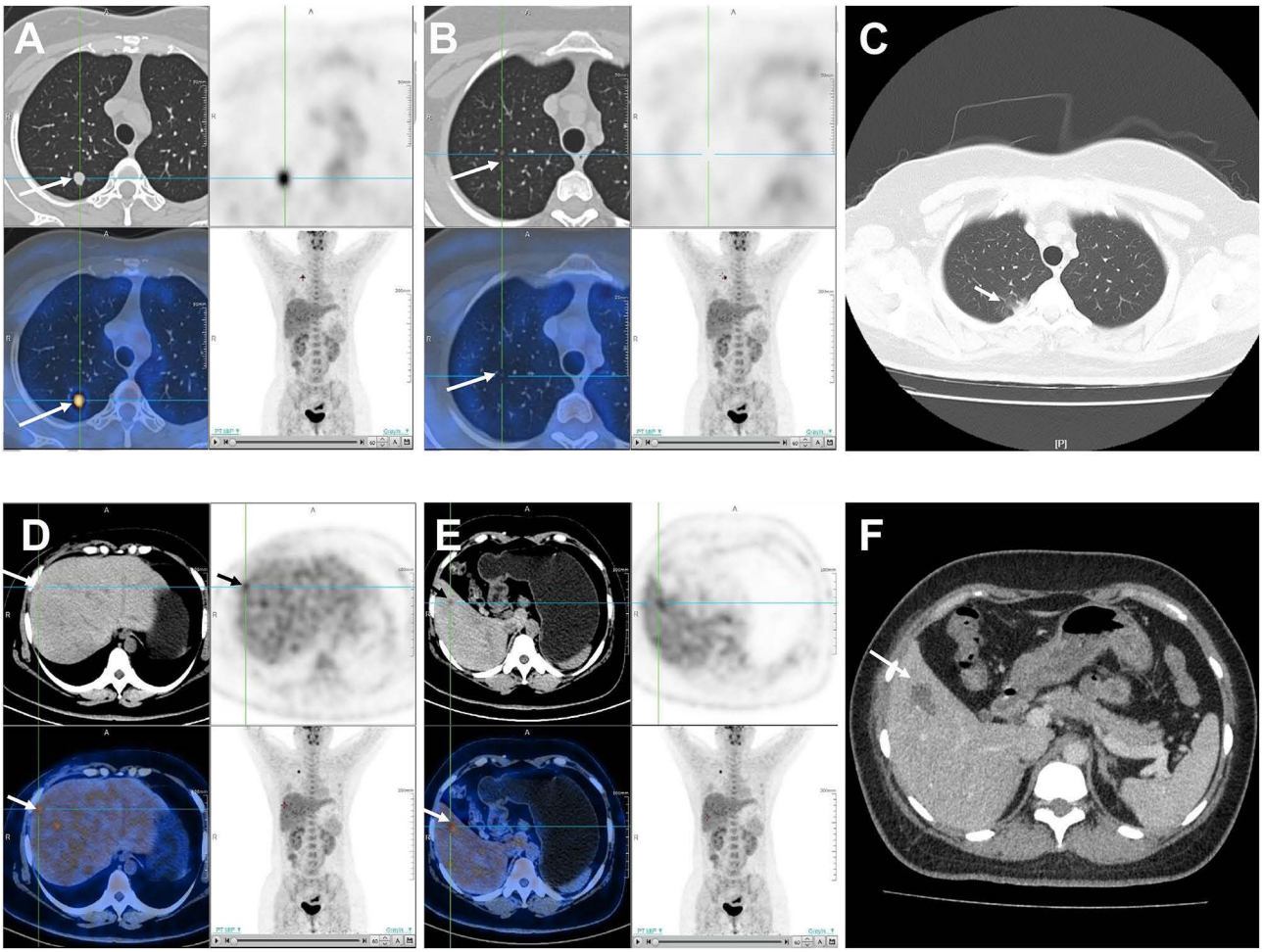

Figure 4 Images of lung and liver metastases.
Notes: (**A**) Lung PET/CT: a solid nodule with increased glucose metabolism is seen in the posterior segment of the upper lobe of the right lung, with a maximum SUV value of 8.62 and an approximate size of 11.8 × 9.7 mm. (**B**) Lung PET/CT: nodule without glucose metabolism is seen in the dorsal segment of the right lower lobe of the right lung, with a diameter of 3.6 mm. (**C**) Lung CT image after radiofrequency ablation. (**D**) Liver PET/CT: nodule with increased glucose metabolism in the right anterior lobe of the liver, with a diameter of 8 mm. (**E**) Liver PET/CT: nodule with glucose metabolism a maximal SUV value of 4.50 in the right anterior lobe of the liver, with a diameter of about 10.5 mm. (**F**) Liver CT after radiofrequency ablation: patchy hypodense area in the right lobe of the liver. The arrows show the lung and liver metastases.

This study was approved by the ethics committee of China-Japan Union Hospital of Jilin University. Written, informed consent was obtained from the patients' husband for publication of this case report and accompanying images.

Discussion

Malignant epithelial myoepithelial carcinoma (MEMC) is a rare breast carcinoma in women. In 5th World Health Organization (WHO) Classification of Breast Tumors 2019, EMC 8562/3, malignant epithelial myoepithelial tumors include adenoid cystic carcinoma (ACC) and cancers that occur on the basis of adenomyoepithelioma (AME) 8983/3. AME includes luminal epithelial carcinoma, myoepithelial carcinoma, and epithelial myoepithelial carcinoma, while EMC refers to tumors with malignant transformation of both epithelial and myoepithelial components, but not necessarily the presence of AME components.[1,2] Epithelial cells and myoepithelial cells which share a common progenitor cell have different functions. The epithelial cells consist of an inner layer of cells and have the function of milk production and conduction, while the myoepithelial cells consist of a semicontinuous outer layer, supporting the basal membrane, have the function of contraction. Epithelial cells stain for CK 7, 8, 18 and 19, while myoepithelial cells stain for CK 5/6, 14, 7, SMA, calponin, SMM-HC and p63, which help differentiate these the cell population of lesions. According to a case analysis from the National Cancer Database,[3] EMC is more common in postmenopausal women, with a median

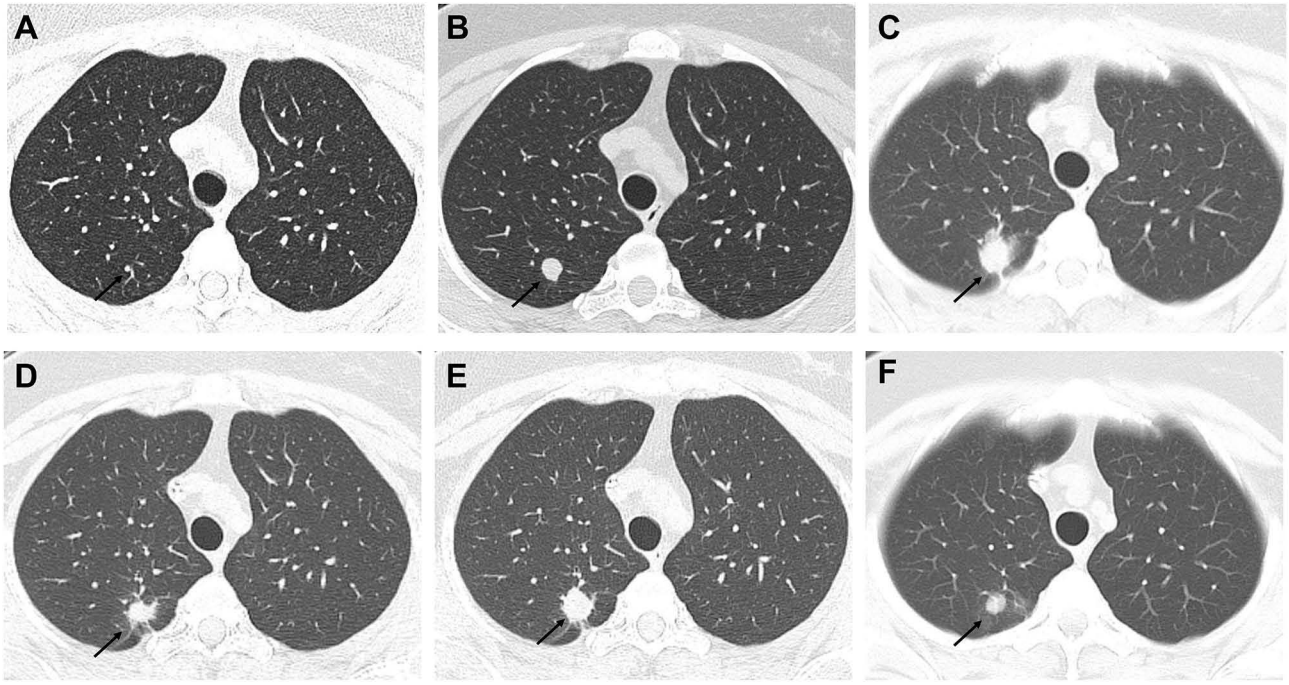

Figure 5 Lung CT images.
Notes: (**A**) Lung CT of December 2021 showed a right lung upper lobe nodule size 2 mm. (**B**) Lung CT of June 2022 showed the right lung upper lobe nodule size increasing to 11.8×9.7 mm. (**C**) Lung CT of July 2022, the right lung nodule size increased to 18×11 mm, CT value 23HU. (**D**) Lung CT of September 2022 showed the nodule size is 16×8 mm, CT value 18HU. (**E**) Lung CT of November 2022 the nodule size is 16×12 mm, CT value 23HU. (**F**) Lung CT of January 2023, the size decreased to 14×13 mm, CT value 19HU. The black arrows show the lung metastases.

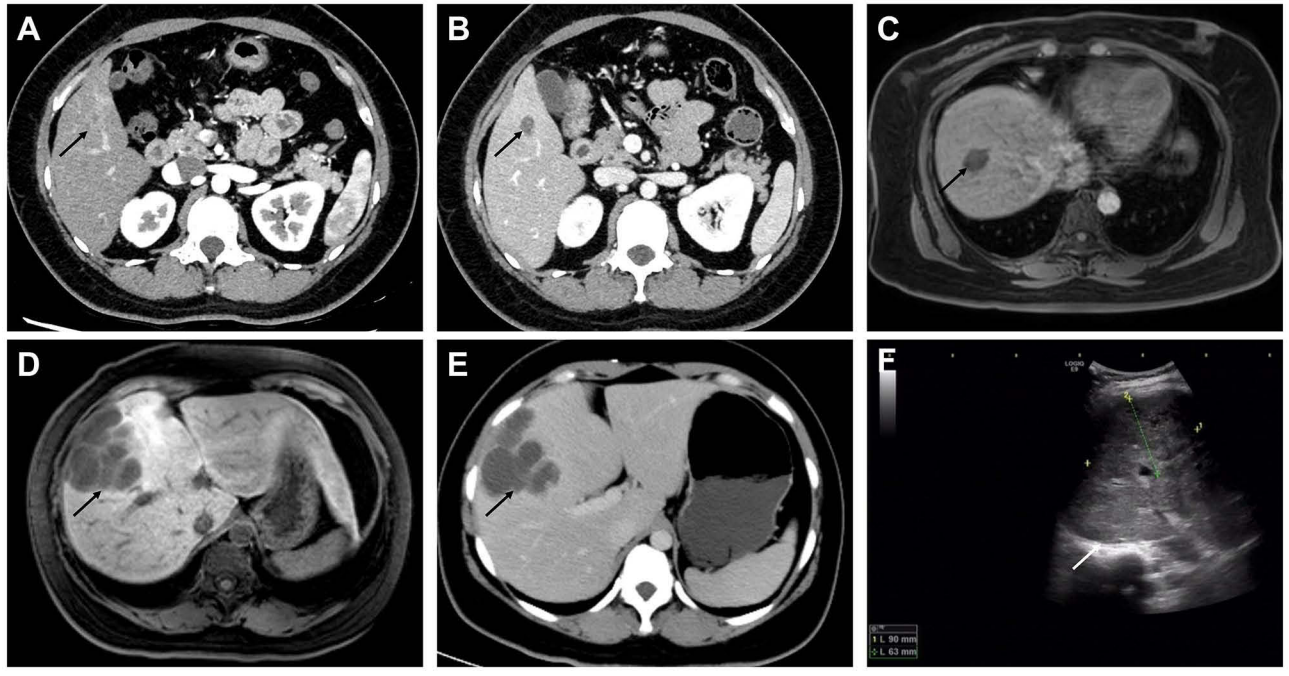

Figure 6 Liver Imaging.
Notes: (**A**) Liver CT enhancement of June 2022 showed a circular slightly low-density mass with a size of 0.9 cm is observed in the anterior lower segment of the right lobe of the liver. (**B**) Liver CT enhancement of September 2022 showed the mass size 2.8×1.0 cm. (**C**) Liver MR enhancement of November 2022 showed multiple nodular abnormal signals in the right lobe of the liver, with a size of 0.8–4.6 cm, and enhanced scanning with circular enhancement. (**D**) Liver MRI of January 2023 showed the masses ranging in size from 0.8 to 5.3 cm. (**E**) Liver MRI enhanced of March 2023 showed the masses size from 0.8 to 5.6 cm. (**F**) Liver ultrasound of June 2023 showed the largest mass measuring 13 cm in diameter. The arrows show the liver metastases.

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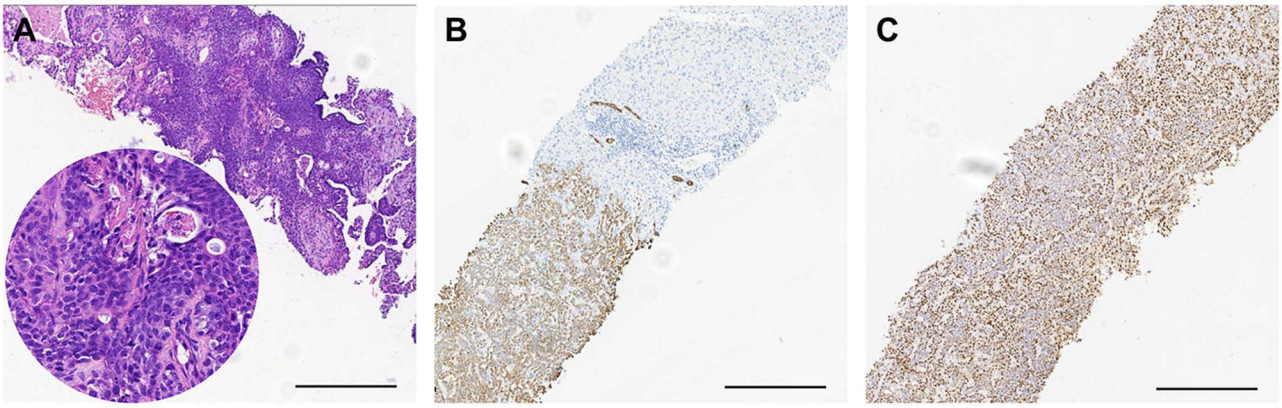

Figure 7 Pathology of Core needle biopsy of the lung and liver masses (×40).
Notes: (**A**) H&E section of lung tissue. The small figure in the lower left corner is a local zoom-in (×200). (**B**) IHC of CK7 of liver tissue is positive. (**C**) IHC of CK63 of liver tissue is positive. (Scale bar: 400 μm).

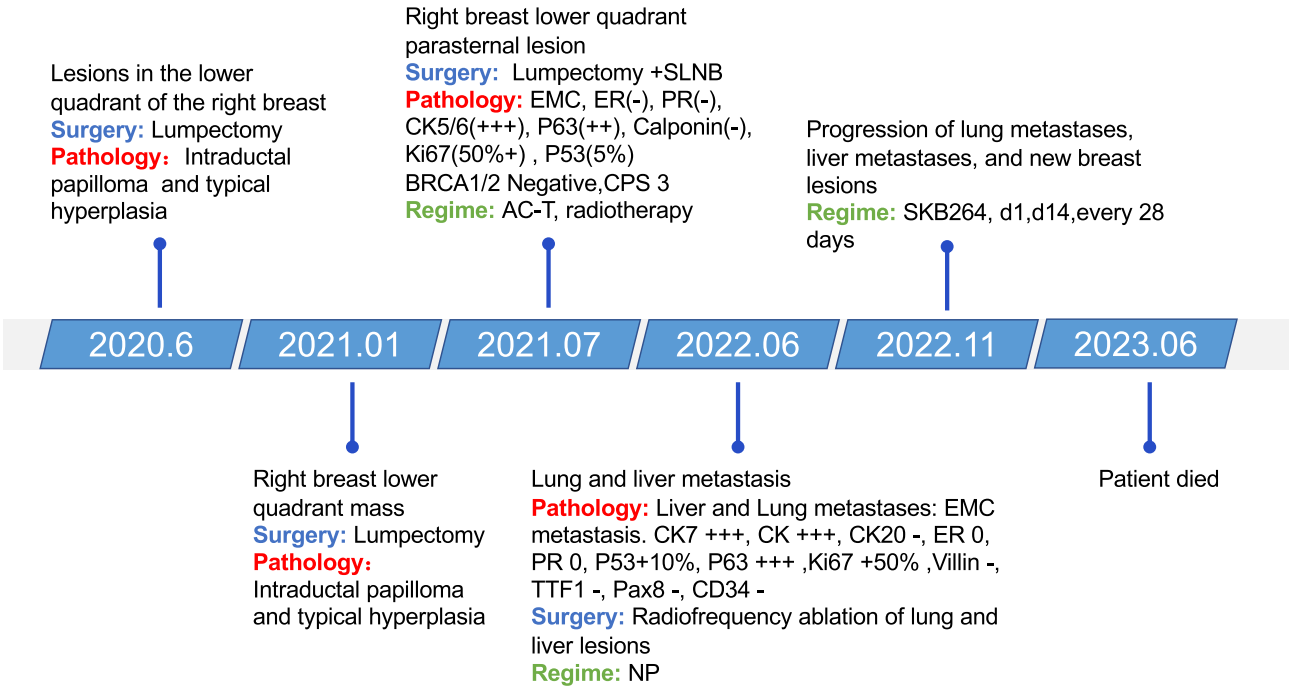

Figure 8 Timeline of the case.
Notes: The patient was diagnosed with EMC in July 2021. Before the diagnosis, she underwent breast surgeries twice, and the pathology showed intraductal papilloma and typical hyperplasia. After the EMC diagnosis, breast conserving surgery, chemotherapy and radiotherapy were performed. Metastasis occurred during chemotherapy, followed by radiofrequency ablation of metastatic lesions. Then NP regimen chemotherapy, Trop2 ADC treatment were given for a brief remission. The patient died 2 years after EMC diagnosis.

age at diagnosis of 67 years, triple-negative phenotype, and milder biological behavior than that of other triple-negative breast cancers. Lymph node involvement and distant metastasis are very rare, though there are individual case reports of distant metastasis. The median follow-up time was 67.6 months, and the 5-year overall survival rate (OS) was 74.3%, which was significantly correlated with tumor size (82.7% for tumors ≤ 2 cm, 76.5% for 2–5 cm, and 50% for > 5 cm).

There are few reports on MAME, and even rarer reports on MEMC. Therefore, our subsequent diagnosis and treatment review will mainly on MAME reports. At present, there is limited research data on the molecular phenotype of malignant AME, with only a few reports of *HRAS* gene Q61R/K, G12D, G13R, and G12S mutations in malignant AME.

ER and PR negative malignant adenomyosis can have both *PIK3CA* gene H1047R and *HRAS* gene G12/G13 hotspot mutations.[4–6] Geyer et al reported the telomerase reverse transcriptase (*TERT*) gene hotspot mutations and *CDKN2A* (p16INK4a) homozygous deletion associated with carcinomas arising in AME.[7] A study has found that malignant AME where both glandular epithelium and myoepithelial cells undergo malignant transformation exhibit amplification and overexpression of the *C-myc* gene, but not in the tumor tissue of adenomyoepithelial tumors. Literature reports suggest that the *C-myc* gene plays an important promoting role in the malignant transformation of adenomyoepithelial tumors.[8] Jones et al reported a case of malignant AME with losses at chromosomes 11q and 16q in the breast mass and an additional loss at 12q in the metastasis.[9]

Malignant AME sometimes are misdiagnosed as some benign diseases such as intraductal papilloma, sclerosing adenosis and tubular adenoma. Malignant adenomyoepithelial tumors with papillary growth patterns need to be differentiated from intraductal papillary tumors. AME is mainly characterized by myoepithelial hyperplasia with a special double layered sheath structure, while intraductal papilloma only shows myoepithelial cells on the outer side of the papillary region, such as epithelial hyperplasia, mainly glandular epithelium, and no significant proliferation of nest like or nodular myoepithelium. It is worth noting that focal intraductal papillomatous areas are visible in mature adenomyosis,[10] but should not alter the overall diagnosis of adenomyosis. However, in intraductal papillary carcinoma, the myoepithelium of the nipple axis is missing, rather than the bidirectional differentiation of glandular epithelium and myoepithelium in malignant adenomyoepithelioma. The detection of *HRAS* mutations in malignant adenomyosarcoma can help with diagnosis.[5] In this case report, the patient had a history of two surgeries before being diagnosed with MEMC, both of which were diagnosed with intraductal papilloma. During the surgical process of diagnosing MEMC, intraductal papilloma still existed. Therefore, we also believe that MEMC is a special variant or morphological progression of intraductal papilloma, and further in-depth research is needed.

Trophoblast cell surface antigen 2 (TROP2) is a type I cell surface glycoprotein, also known as tumor-associated calcium signal transducer 2 (TAC-STD2), which is a transmembrane glycoprotein commonly expressed on the surface of various human cell types and stem cells and is critical for embryonic development. It also exhibits low-level expression in normal tissues but high-level expression in epithelial tumors such as breast cancer, which is closely related to tumor proliferation, invasion and metastasis.[11] The United States Food and Drug Administration (FDA) approved the application of sacituzumab Govitecan (SG) for triple-negative breast cancer (TNBC) patients who have received two or more prior therapies (ASCENT, NCT02574455), which improved median progression-free survival (PFS: 4.8 v 1.7 months; hazard ratio (HR), 0.41 [95% CI, 0.33 to 0.52]) and median overall survival (OS; 11.8 v 6.9 months; HR, 0.51 [95% CI, 0.42 to 0.63]) over treatment of physician's choice (TPC).[12] Datopotamab deruxtecan (Dato-DXd) is another TROP2 ADC with a potent DNA topoisomerase I inhibitor (DXd), and also has a positive result in Previously Treated Inoperable/Metastatic Hormone Receptor-Positive Human Epidermal Growth Factor Receptor 2-Negative Breast Cancer (TROPION-Breast 01, NCT05104866), which significantly reduced the risk of progression or death versus investigator's choice of chemotherapy (ICC) (PFS by BICR HR, 0.63 [95% CI, 0.52 to 0.76]; $P < 0.0001$).[13] As a newly developed TROP2 ADC drug, SKB264, has a longer half-life, stronger targeting effect and better anti-tumor activity than IMMU-132, that is, it has stronger anti-tumor activity.[14] In our case report, the patient used the drug TROP2 ADC (SKB264, 340 mg, D1, D14), shrinkage of lung, liver and breast metastases can be noticed for 3 months. We may consider that TROP2 ADC drug has an effect on the patient.

In our review of the latest 5 years (searching time 14/02/2025), we searched the PubMed, Web of Science and Google Scholar as the topic of "malignant adenomyoepithelial", "malignant epithelial myoepithelial carcinoma" and "the breast", and there are 78 articles related. Excluding benign cases and no full text, a total of 26 articles were left, involving 34 patients with malignant AME (Table 1). In Table 1, the mean age of patients is 61.7 year (range 27-84 years), with only one male. It is worth noting that two patients malignant AME founded after silicone implant surgery,[15,16] and further research is needed to confirm whether implant implantation is related to the occurrence of MAME. We also searched the PubMed, Web of Science and Google Scholar to find metastatic malignant AME (Table 2). Moreover, malignant AME has a strong potential for local recurrence and distant metastasis (lung,[17] liver,[9] brain,[18] kidney,[18] heart,[18] adrenal,[19] ovarian,[19] thyroid gland[20] and bone[21]). Bult reviewed such cases and recognized the ratio of local recurrence and distant metastasis was 1/3,[20] while we considered the rate of local recurrence and distant metastasis was nearly 1/6. At present,

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Table 1 Malignant AME Literature in the Latest 5 years

Case	Author	Publish Year	Age(y)/ Sex	T Category (Size cm)	ER/PR Status	Ki67 Index	Surgical Methods	Recurrence (Time to Surgery)	Lymph Node Metastasis	Metastasis	Radiotherapy	Chemotherapy Regimen
1	Grenier[24]	2020	76/F	T2 (3–4)	NM	NM	Mastectomy +SLNB	NM	1/2	NM	NM	NM
2	Jameel[25]	2022	62/F	T1 (1.5)	Negative	NM	Lumpectomy	No	/	NM	No	No
3	Wang[26]	2022	34/F	T2 (3.2)	Negative	60%	Lumpectomy +SLNB	NM	0/4	NM	Yes	AC-T
4			45/F	T2 (3.0)	Negative	75%	Lumpectomy +SLNB	NM	0/5	NM	Yes	AC-T
5	Zhao[27]	2024	27/F	T2 (2.5)	Positive	80%	Lumpectomy	No	/	NM	Yes	No
6	Wu[28]	2024	67/F	T2 (3.8)	NM	NM	Mastectomy +SLNB	No	0/6	NM	NM	NM
7	Oda[18]	2021	53/F	T1 (2.0)	NM	57.1%	Lumpectomy +SLNB	Yes (24 and 32 month)	0	No	No	No
8	Tabbaa[29]	2023	84/F	T3 (5.5)	Positive	10-20%	Mastectomy +SLNB	NM	NM	NM	Yes	No
9	AlQurashi[30]	2022	64/F	NM	NM	NM	Lumpectomy	No	/	NM	NM	NM
10			74/F	T1 (2)	NM	NM	Lumpectomy	No	/	NM	NM	NM
11			49/F	T2 (4)	NM	NM	Mastectomy +SLNB	NM	NM	/	NM	NM
12	Ma[31]	2025	65/M	T1 (1.5)	Negative	5%	Mastectomy +SLNB	No	0/2	No	No	No
13	Zhai[32]	2021	48/F	T1 (2)	Negative	30%	Mastectomy +SLNB	No	0/3	No	No	AC-T
14			56/F	T2 (4)	Negative	25%	Mastectomy +SLNB	No	0/5	No	No	EC*4
15	Bojja[33]	2022	70/F	T2 (5)	NM	NM	Lumpectomy	No	/	NM	NM	NM
16	Chen[34]	2023	59/F	NM	Negative	20-30%	NM	NM	NM	NM	NM	NM
17	Lari[35]	2020	39/F	T2 (3)	Negative	30%	Mastectomy +SLNB	No	0	NM	NM	NM
18	Moro[19]	2020	64/F	T4	NM	44%	Lumpectomy, Mastectomy +ALND	Yes (6 month)	0/26	Brain, Lung, Heart, Kidney, Ovary, Adrenal gland	No	FEC-T
19	Zhang[36]	2021	64/F	T2 (4.5)	Negative	NM	Mastectomy +SLNB	NM	0/3	NM	NM	NM
20	Hu[15]	2022	55/F	T2 (2.8)	Negative	30%	Mastectomy +SLNB+ reconstruction	No	0/4	NM	NM	AC*3
21	Ma[37]	2025	67/F	T1 (1.7)	Negative	10%	Mastectomy +SLNB	No	0/1	NM	NM	NM
22	Hu[16]	2022	80/F	T3 (6)	Negative	30%	Mastectomy +ALND	No	0	No	No	No
23			56/F	NM	Negative	15-30%	Mastectomy +SLNB	No	0	No	No	AC*1
24	Amano[38]	2020	64/F	T2 (3)	Negative	NM	Lumpectomy	No	/	NM	NM	NM
25	Ginter[39]	2020	42/F	T2 (4.8)	Negative	NM	Mastectomy	No	0	NM	NM	NM
26			66/F	T1 (1.2)	Positive	NM	Mastectomy	No	N1a	NM	NM	NM
27			78/F	Tis (1.4)	Negative	NM	Lumpectomy	No	0	NM	NM	NM
28			56/F	T4 (2.2)	Negative	NM	Mastectomy	No	N1a	Lung	NM	NM
29	Ha[40]	2020	50/F	T2 (3.5)	Negative	30%	Lumpectomy; Mastectomy +ALND	Yes (1 month)	NM	No	NM	NM

30	Khan[22]	2024	61/F	T2 (5)	Negative	30%	Mastectomy +ALND	No	NM	Lung, Brain	Yes	Yes (NM)
31	Uchida[41]	2022	64/F	NM	NM	NM	Mastectomy +ALND	Yes (NM)	NM	NM	No	NM
32	Zhang[42]	2022	84/F	T2 (2.2)	NM	NM	Lumpectomy +SLNB	NM	NM	NM	NM	No
33	Zha[43]	2023	59/F	NM	NM	NM	Lumpectomy	No	/	NM	NM	
34	Joyon[44]	2023	46/F	T1 (1.8)	Negative	60%	Mastectomy	NM	/	NM	Yes	Yes (NM)
35	Hui[45]	2024	75/F	T2 (3.2)	Nearly negative	NM	Mastectomy	NM	/	NM	No	Yes (TC*1)

Abbreviations: F, Female; M, Male; NM, Not mention; ALND, Axillary lymph node dissection; SLNB, Sentinel lymph node biopsy.

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there is no unified guideline for the treatment of malignant AME. Studies suggest that complete breast tumor resection with negative margins can reduce the local recurrence rate of the tumor, as axillary lymph node metastasis is rare in malignant adenomyosis. In Table 1, 3 in 34 patients lymph node metastasis, 13 lumpectomy (4 recurrence near the scar), 20 mastectomy, 3 distance metastasis, the patient we reported in this case report also experienced recurrence, but the site of recurrence was in another quadrant. Therefore, we believe that mastectomy is better for malignant AME. There is still controversy over whether axillary lymph node dissection is necessary. Regarding the management of metastatic tumors, Khan[22] underwent pneumonectomy and multiple lung metastases occurred in a short period of time. Takahashi[23] underwent osteotomy, and in a short time another organ had metastasized. The other patients who played lobectomy in the Table 2 had a long disease survival. The patient reported in our article emerged lung and liver metastases, and died 12 months after metastasis. In Table 2, the average survival time of patients with organ metastasis is 6 months, and we performed interventional radiofrequency ablation treatment, which may play an important role to prolonging her overall survival. Local treatment (radiofrequency ablation) may be recommended to perform in metastatic tumors, which has a positive significance.

Previous studies have shown that malignant AME is insensitive to chemotherapy, but in Table 1, 8 patients still carried chemotherapy, all containing anthracycline-based regimens. In our case report, disease progression during the AC-T adjuvant chemotherapy indicated malignant AME was ineffective against anthracyclines and paclitaxel. Among the distant metastasis patients, one had a brief clinical remission during FEC treatment, while the disease progressed during eribulin treatment.[19] Another showed a positive effect on eribulin,[50] and gemcitabine, capecitabine all invalid. During the 4-month treatment with TROP2 ADC, the patient in this article showed a brief clinical remission in lung and breast metastases. Malignant AME usually has a good prognosis, while sometimes it can progress very aggressively as it did in our case.

Table 2 Malignant AME Literature of Distant Metastases

Case	Author	Publish Year	Age (Year)	T Category	Metastasis	Radiotherapy	Chemotherapy Regimen	Surgery	Time from Metastasis to Death
1	Bult[20]	2000	64	/	Thyroid	Yes	CMF, A	/	6m
2	Samant[46]	2009	50	T3	Lung	No	FEC	/	NG
3	Maffini[47]	2013	44	T1	Lung	Yes	ViFup	Lobectomy	NG
4	Yuan[21]	2017	58	T2	Bone	Yes	TEC	/	11m
5	Korolczuk[48]	2016	56	NG	Lung	NG	/	Lobectomy	NG
6	Loose[49]	1992	42	T2	Lung, bone, brain	Yes	/	/	10m
7	Moro[19]	2020	64	T4	Brain, Lung, Heart, Kidney, Ovary, Adrenal gland	No	FEC, eribulin	Lobectomy	17m
8	Ginter[39]	2020	56	T4	Lung	NG	NG	/	8m
9	Khan[22]	2024	61	T2	Lung, brain	Yes	NG	Lobectomy	NG
10	Lee[50]	2015	51	NG	Liver, pleura, abdominal wall	/	TP, NP, AC, T, gemcitabine, capecitabine, eribulin	/	NG
11	Simpson[51]	1998	50	T2	Lung	Yes	/	/	NG
12	Michal[52]	1994	77	T3	Lung	Yes	NG	/	5m
13	Kihara[53]	2001	86	T2	Lung	No	/	/	2w
14	Trojani[54]	1992	51	T2	Lung	Yes	NG	Lobectomy	NG
15	Takahashi[23]	1999	60	T4	Bone, lung, mediastinal lymph node	Yes	FEC	osteotomy	6m
16	Chen[55]	1994	54	T3	Bone	/	NG	/	7m
17	Foschini[56]	1995	60	T2	Lung	/	/	/	NG
18	Rasbridge[57]	1998	76	T3	Brain	/	/	/	6m

Abbreviations: NG, not given; C, cyclophosphamide; M, methotrexate; F, 5-fluorouracil; E, adriamycin; T, docetaxel; ViFup, vincristine and fluorouracil; N, vinorelbine; P, cisplatin.

Conclusion

Intraductal papilloma may be associated with the occurrence and development of malignant AME, and multi-focal and multi-centered may occur in the malignant AME. It is very important to assess the extent of the primary lesion by MRI and mastectomy may have a favorable outcome after recurrence. AC-T chemotherapy regimen was ineffective to malignant AME, while TROP2 ADC may be effective on it. After distant metastasis, radiofrequency ablation may be a better option.

Data Sharing Statement

Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

Ethics and Consent Statements

The case details and accompanying images published were approved by the ethics committee of China-Japan Union Hospital of Jilin University. Written, informed consent was obtained from the patients' husband for publication of this case report and accompanying images.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors report no conflicts of interest in this work.

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