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CASE REPORT

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Solitary metastases of lower extremity myxoid liposarcoma to breast: A case report and review of literature

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Abstract

Solitary breast metastases from myxoid liposarcoma are extremely rare. Resection with negative margins seems as an effective treatment leading to improved survival.

KEYWORDS

adjuvant treatment, breast metastasis, Myxoid liposarcoma, surgical resection

1 | INTRODUCTION

Solitary metastases of myxoid liposarcoma (ML) to the breast are extremely rare. We report a case of 53-year-old woman with left popliteal ML presented with solitary metastatic involvement of right breast five years later. She underwent breast-conserving surgery and sentinel lymph node biopsy followed by adjuvant radiation.

Metastatic involvement of breast with extramammary malignancies is very rare and constitutes less than 0.5-2 percent of breast malignancies[1,2]; lung cancers, lymphoma, leukemia, malignant melanoma, sarcoma (MFH, rhabdomyosacroma), ovarian cancers, renal tumors, thyroid tumors, colon cancers, gastric cancers, hepatocellular carcinoma, esophageal SCC, cervical cancer, and prostate cancers are all have been reported as primary tumor in breast metastases.[1-4] Breast involvement in this setting is usually accompanied with disseminated disease and has poor prognosis.[1,3,4] Among these malignancies, metastases from lung cancers are more common and from sarcoma is extremely rare.[1,2]

Metastatic involvement of breast with myxoid liposarcoma (ML), a common subtype of liposarcoma, is an extremely rare entity which is mostly result of disseminated systemic metastases outside the breast. Solitary metastasis to the breast is extremely rare, and we found only four cases in the English literature.[1,5] Herein, we report a case of a 53-year-old woman with history of left popliteal ML, presented with solitary metastatic involvement of right breast 5 years later.

2 | CASE REPORT

A 53-year-old lady presented with a large mass in lateral area of her right breast. In physical examination, a 5cm firm mass in 9 o'clock of right breast was palpable; no axillary lymph nodes were detected. She had history of left popliteal myxoid/round cell type liposarcoma (10 × 8 × 0.5.5 cm, round cell component more than 25%; IHC staining negative for CK and EMA, and positive for CD99, vimentin, S100, CD34; ki67 expressed in 50% of tumoral cells) 5 years ago, which had been treated by wide local excision and adjuvant radiation. Since then, she had regular follow-up with no sign and symptom of local recurrence.

Breast's imaging, including mammography and ultrasonography, showed a suspicious lesion (B4b) in right breast with no pathologic axillary lymph nodes (Figure 1).

Core needle biopsy of the mass was done, and pathologic examination reported malignant undifferentiated neoplasm; IHC staining was positive for Bcl2, CK5/6, and CK14 and negative for PanCK, CAM5.2, LCA, CD20, CD3, CD4, Melan A, CD31, CD34, and P63; regarding histology review and IHC results the pathologist claimed that metaplastic carcinoma could not be excluded, and the findings were against the diagnosis of lymphoma, melanoma, and vascular tumors.

According to patient's past history, metastatic workup including thoracic, abdominopelvic CT scan, bone scan, and PET scan was done which revealed no distant metastases. The patient scheduled for surgery and breast-conserving surgery with reduction type oncoplasty and right axillary sentinel lymph node biopsy was done.

In pathologic examination, a rather well-defined creamy-whitish mass with elastic consistency was noted in breast tissue (Figure 2). In microscopic examination, predominantly back-to-back round cells with high nucleocytoplasmic ratio and prominent nucleoli were seen. Capillary vessels and myxoid background were inconspicuous but interspersed adipocytes were noted. Mitotic figures were significant, and foci of necrosis (less than 50%) were identified. Regarding patient's history, review of pathology slides of left leg and immunohistochemistry (IHC) study on her breast lesion were done. Review of popliteal lesion pathology slides revealed pretty similar findings to breast lesion. To rule out metaplastic carcinoma of breast—which have a higher incidence among breast malignancies compared with sarcomas—IHC study for CKAE1/AE3, CK5/6, Cam 5.2, CK14, and P63 was done and to rule out lymphoma, melanoma, and angiosarcoma, LCA, CD31, CD34, and S100 were done. IHC study results were as follows: negative epithelial markers, LCA, CD31, CD34, and Melan A. They were positive for S100 (figure 3). Results were compatible with liposarcoma, myxoid variant, containing more than 90% round cell component. Regarding patient history and pathologic findings, metastatic nature of breast myxoid liposarcoma was confirmed.

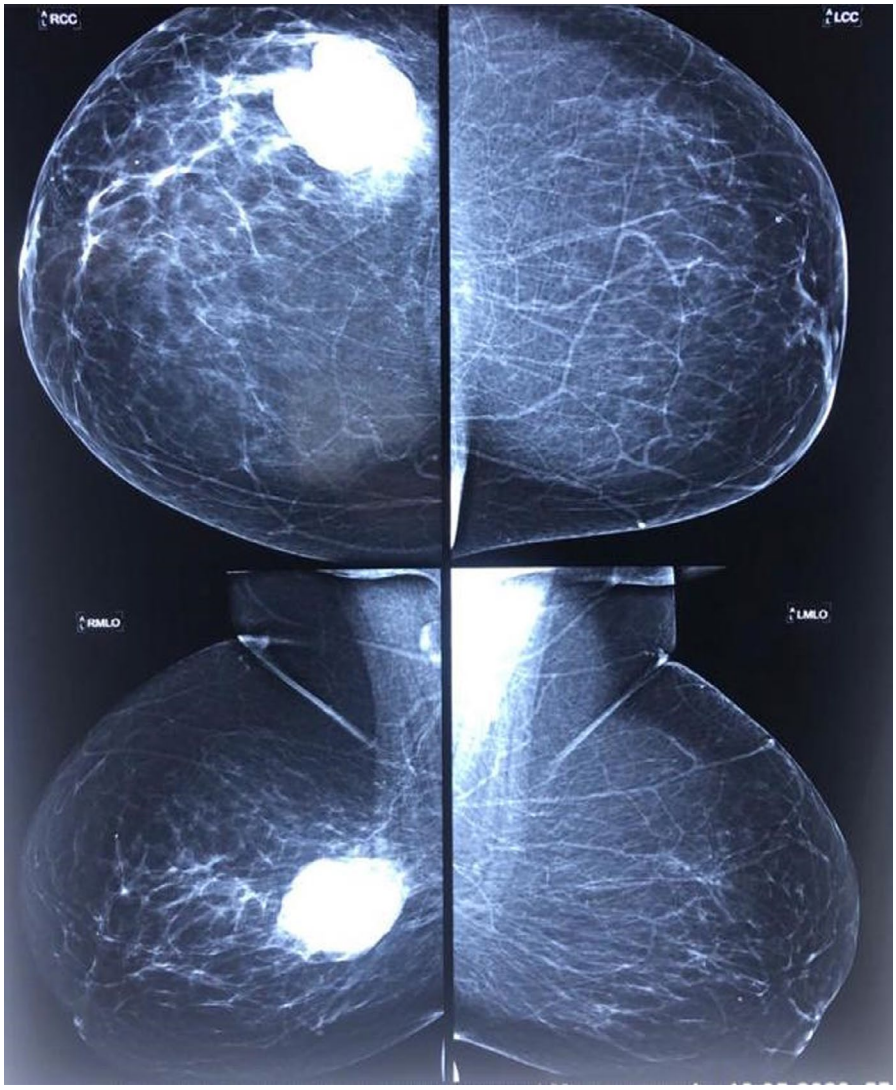

FIGURE 1 MLO and CC view of patient's mammography which shows the circumscribed mass in right breast

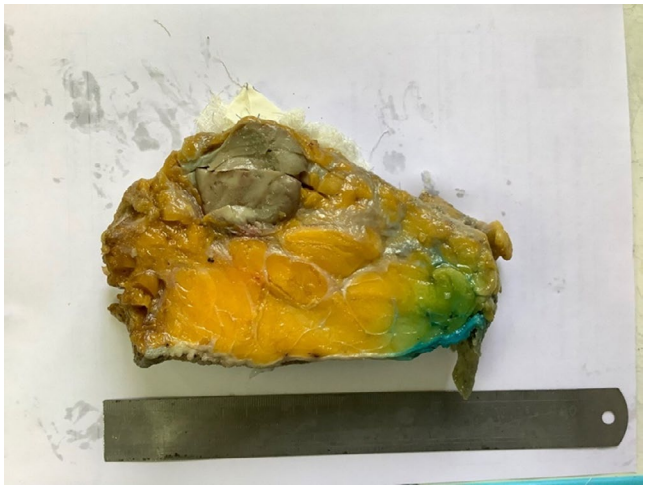

FIGURE 2 Breast tumor: a rather well-defined creamy-whitish mass with elastic consistency

The pathologic review of surgical specimen showed more than 2cm free margins and two reactive lymph nodes.

The patient had an uneventful postoperative course. She presented to MDT session to decide about her adjuvant treatments. MDT members claimed that there is no strong evidence of chemotherapy benefit in this case, but it can be recommended regarding tumor's myxoid subtype. The patient refused systemic therapy and had adjuvant radiotherapy. Now, she has been followed up for 6 months with no events.

3 | DISCUSSION

Metastatic involvement of breast with extramammary malignancies can occur in both males and females.[2,6] Median interval from diagnosis to metastasis is reported to be 12 months.[3]

These lesions are usually superficial, palpable, moveable, firm, and without adhesion to skin or pectoralis muscle; generally, no nipple discharge is seen.[2] In imaging, they present as multiple or single lesions, mostly circumscribed, usually with no microcalcification or spiculation; however, microcalcification can be seen in 10% of such lesions mostly in lesions with ovarian, lung, and thyroid origin[1,2,6.] Metastases of rhabdomyosarcoma or ovarian tumors can have microlobulated margins in ultrasound.[1]

In pathologic evaluation, these metastatic lesions usually present as high-grade adenocarcinoma that can mimic grade 3 ductal carcinoma.[2] Estrogen receptor(ER) and progesterone receptor (PR) status is not helpful in discriminating primary from metastatic breast lesions, as high-grade breast carcinomas can be hormone receptor negative; on the other hand, ER and PR can be positive in endometrial, ovarian, lung, and hepatic tumors.[2]

Treatment of metastases in these patients is usually done according to type of primary tumors. These conditions are usually accompanied with disseminated metastatic disease with poor prognosis and most patients die within one year of diagnosis (median 4 months).[1-4] Metastatic involvement

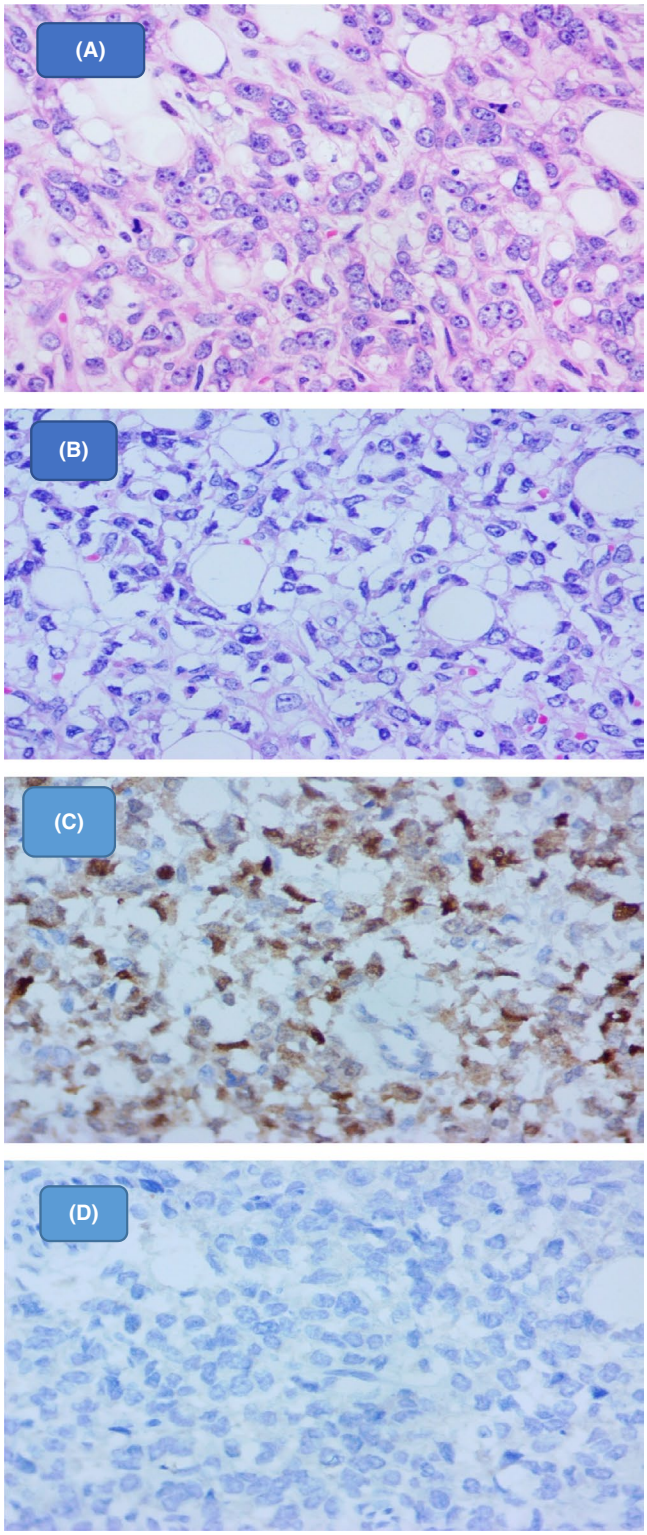

FIGURE 3 A) Popliteal tumor with predominance of round cell, prominent nucleoli, and mitotic figures. B) Breast tumor with similar findings to popliteal tumor. C) Breast tumor nuclear staining for S100.D) Breast tumor negative staining for CKAE1/AE3

of breast with soft tissue sarcoma and ML is too rare and is mostly result of disseminated systemic metastases outside the breast.

Liposarcomas- including well-differentiated, dedifferentiated, myxoid/round cell, and pleomorphic subtypes- constitute the second most common soft tissue sarcomas.[7,8] Myxoid liposarcoma(ML) is the second most prevalent liposarcoma.[1] One third of patients with ML develop metastases which generally has poor prognosis.[1] Some authors have proposed that increased tumoral round cell component –suggested by some as >5%-is associated with higher likelihood of metastatic disease.[9,10]

In contrast to other sarcomas which have a predilection for lung metastases, ML has a unique metastatic pattern with tendency for extrapulmonary sites, mostly without pulmonary involvement. Reported involved sites include soft tissue of opposite site, bone, retroperitoneum, abdominal wall, abdominal cavity, thoracic wall, pelvic wall, pleura, and pericardium.[1,7,9-12] Patients with ML and extrapulmonary metastases have better survival compared to patients with pulmonary metastasis.[1,9]

Solitary metastases of ML to the breast is extremely rare, with only four reported cases in the English literature so far.[1,5] Youkouchi et al reported a case of 66-year-old woman with history of right thigh ML who presented with left breast solitary metastases 21 months later. She underwent only local excision with negative margins without adjuvant treatment. She had been followed up for 5 years with no evidence of recurrence. The authors concluded that curative surgical resection was an effective treatment for the patient.[1] Biyi et al reported a case of 51-year-old woman with right thigh ML presented with synchronous left breast metastasis. The patient was treated with six chemotherapy cycles (Doxorubicine, Ifosfamide) with good control of disease on 3-year follow-up.[5] The two other cases provided insufficient information.[12,13] Regarding these reports, interval time from primary tumor treatment to breast metastasis diagnosis of 0-4 year have been represented.[1,5,13]

Patients with extrapulmonary involvement have better survival than those with pulmonary involvement.[1,8,9] In one study by Lin et al on 4181 patients with liposarcoma (19.9% ML subtype), the metastasis rate was 5.4% which more than two third of them were extrapulmonary. five-year survival in metastatic patients was 12.1% with mean survival of 21.4 months.[14] In another study by Spillane et al in Royal Marsden hospital on 50 patients with ML with median follow-up of 43 months, the five year soft tissue metastases rate was reported to be 31%. Median interval to metastases diagnosis was 23 month, and median survival was 35 months.[9] Pearlstone et al studied 102 patients with ML in MD Anderson Cancer Center in 13 years. 32.4% of patients had metastasis which most of them(94%) were extrapulmonary.[12] In another study by Durr et al on 43 patients, four patients showed metastases that all of them were extrapulmonary; all patients died in median 33 months.[7]

Regarding its rarity, there is no standard treatment for solitary metastatic ML, including breast metastasis. Some suggest that treatment options should be chosen according to origin and type of tumor.[2] Some authors, as Yokouchi et al have postulated that complete surgical resection with negative margins can be an effective treatment in such patients and can lengthen patient survival. According to these reports and as patients with extrapulmonary metastases have better outcome, it is suggested that these tumors should be managed aggressively.[1,9] Regarding adjuvant treatments, the role of chemotherapy in theses patient is not clear[1]; but ML is generally sensitive to radiotherapy and this modality is recommended by some as adjuvant treatment in these patients.[1,8]

4 | CONCLUSION

Solitary breast metastases from Myxoid liposarcoma is extremely rare, with very few cases reported so far. As these patients usually have better prognosis than ML patients with pulmonary metastases, surgical resection with negative margins seems reasonable as it has been shown that effective treatment can improve survival. Regarding adjuvant treatments, the role of chemotherapy is not clear, but radiotherapy is recommended by some as adjuvant treatment. As there are very few cases of this entity, more reports with longer follow-ups are necessary to better clarify prognosis and treatment options in these patients.

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CONFLICT OF INTEREST

None to declare.

AUTHOR CONTRIBUTIONS

Ramesh Omranipour: contributed to patient's treatment and data gathering. Negar Mashoori: contributed to data gathering and writing manuscript. Behnaz Jahanbin: contributed to pathologic evaluation of surgical specimens and preparation of photographs of pathology slides. Dorsa Ghasemi: contributed to pathologic evaluation of surgical specimens and preparation of photographs of pathology slides.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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REFERENCES

1. Yokouchi M, Nagano S, Kijima Y, et al. Solitary breast metastasis from myxoid liposarcoma. *BMC Cancer*. 2014;14(1):482.
2. Sauer T. Fine-needle aspiration cytology of extra mammary metastatic lesions in the breast: A retrospective study of 36 cases diagnosed during 18 years. *Cytojournal*. 2010;7:10.
3. Yeh CH, Lin CH, Chen M. Clinical and ultrasonographic characteristics of breast metastases from extramammary malignancies. *Am Surg*. 2004;70(4):287-290.
4. Lee SK, Kim WW, Kim SH, et al. Characteristics of metastasis in the breast from extramammary malignancies. *J Surg Oncol*. 2010;101(2):137-140.
5. Biyi A, Doudouh A. Soft tissue uptake of 99mTc-MDP in a case of myxoid liposarcoma with breast metastasis. *Pan Afr Med J*. 2016;25:184.
6. Surov A, Fiedler E, Holzhausen HJ, Ruschke K, Schmoll HJ, Spielmann RP. Metastases to the breast from non-mammary malignancies: primary tumors, prevalence, clinical signs, and radiological features. *Acad Radiol*. 2011;18(5):565-574.
7. Dürr HR, Rauh J, Baur-Melnyk A, et al. Myxoid liposarcoma: local relapse and metastatic pattern in 43 patients.BMC. *Cancer*. 2018;18:304.
8. Wang D, Wu J, Yu J, Zhang H, Liu H. Solitary pancreatic metastasis of extremity myxoid liposarcoma: a case report and literature review.BMC. *Cancer*. 2018;18:1121.
9. Spillane AJ, Fisher C, Thomas JM. Myxoid liposarcoma–the frequency and the natural history of nonpulmonary soft tissue metastases. *Ann Surg Oncol*. 1999;6(4):389-394.
10. Fuglo HM, Maretty-Nielsen K, Hovgaard D, Keller JO, Safwat AA, Petersen MM. Metastatic Pattern, Local Relapse, and Survival of Patients with Myxoid Liposarcoma: A Retrospective Study of 45 Patients. *Sarcoma*. 2013;2013:548628.
11. Estourgie SH, Nielsen GP, Ott MJ. Metastatic patterns of extremity myxoid liposarcoma and their outcome. *J Surg Oncol*. 2002;80(2):89-93.
12. Pearlstone DB, Pisters PW, Bold RJ, et al. Patterns of recurrence in extremity liposarcoma. *Cancer*. 1999;85(1):85-92.
13. Cheng EY, Springfield DS, Mankin HJ. Frequent incidence of extrapulmonary sites of initial metastasis in patients with liposarcoma. *Cancer*. 1995;75(5):1120-1127.
14. Lin F, Duan J, Lin Y, et al. Survival and risk factors in patients with liposarcoma with distant metastasis. *Am J Transl Res*. 2020;12(5):2071-2082.

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