Clinical Imaging 53 (2019) 191–194

Contents lists available at ScienceDirect

Clinical Imaging

journal homepage: www.elsevier.com/locate/clinimag

Breast Imaging

Recurrent desmoid tumor arising from latissimus dorsi flap: A case report

Arzu Canan[a], Xiaoqin Wang[b,*]

[a] Department of Radiology, UT Southwestern Medical Center, 5323 Harry Hines Boulevard, Dallas, TX 75390-9316, United States of America
[b] Department of Radiology, University of Kentucky, 800 Rose Street, HX-302, Lexington, KY 40536-0293, United States of America

ARTICLE INFO

Keywords:
Aggressive fibromatosis
Desmoid tumor
Latissimus dorsi flap

ABSTRACT

Fibromatosis or desmoid tumor in the breast is a very rare benign soft tissue tumor. We report a case of recurrent desmoid tumor arising from latissimus dorsi flap after lumpectomy for breast carcinoma. To our knowledge, this is the first case of desmoid tumor arising from the latissimus dorsi flap. Despite its benignity, desmoid tumor is often locally aggressive, therefore timely diagnosis and proper management are very important. Imaging and pathological diagnosis as well as treatment management are discussed. High clinical suspicion and multidisciplinary approach are essential for prompt diagnosis and management. Wide surgical resection is required, but there is no consensus regarding treatment due to limited data.

1. Introduction

Desmoid tumor in the breast, also known as aggressive fibromatosis, is a rare benign connective tissue disease with a prevalence of 0.2% of all breast tumors and 3% of all soft tissue tumors [1,2]. While most desmoid tumors have been reported in the trunk and extremities, there are several isolated case reports in the literature describing desmoid tumor in the breast. The etiology of desmoid tumor in the breast is unknown, but association with previous breast trauma, surgery (saline or silicone implants) [3–5], or hereditary disease such as familial adenomatous polyposis [6] and Gardner's syndrome [7] have been described.

Desmoid tumors affect patients between 13 and 83 years old. These are mostly found in women, but a few cases have been reported in men [8,9]. A desmoid tumor is a benign entity without metastatic potential. However, it can grow aggressively and infiltrate local structures. It has a local recurrence rate of 29% and recurrence mostly occurs within 3 years after surgery [1]. Therefore, timely diagnosis and wide surgical resection with clear margins are very important in successful management of this aggressive tumor. A desmoid tumor often presents clinically as a painless palpable mass on physical exam. The tumor is characterized as an irregular mass on various modalities of breast imaging, including mammogram, breast ultrasound, and MRI, which can mimic breast cancer or surgical changes [10–15]. Because it is very rare, there is no standard treatment for desmoid tumors [1,6].

Since the knowledge of its radiological features combined with patient's clinical history is important to prevent misdiagnosis and delay in treatment, we report how we diagnose and manage a very unusual case of a recurrent desmoid tumor in the breast through a multidisciplinary approach. To our knowledge, this is the first case of recurrent desmoid tumor arising from the latissimus dorsi flap after breast surgery for recurrent breast carcinoma.

2. Case report

The patient is a 59-year-old woman who had a history of recurrent breast cancer. She had initially undergone lumpectomy for left upper inner breast cancer (invasive breast cancer - not otherwise specified IBC-NOS, estrogen and progesterone receptor negative, HER2/neu positive) in 2000 at 40 years of age and had adjuvant chemotherapy (4 cycles of Adriamycin and Cytoxan) and radiotherapy after surgery. In 2014, the patient was found to have a recurrent breast cancer (IBC-NOS, estrogen receptor positive, progesterone receptor negative, and HER2/neu positive) in the same location of the left breast. The treatment plan including mastectomy was discussed extensively at the multidisciplinary tumor board and with the patient. Because of the patient's choice of lumpectomy, she received neoadjuvant chemotherapy (6 cycles of trastuzumab combined with docetaxel and carboplatin) before the surgery and had a great treatment response. The patient then underwent breast lumpectomy again with a latissimus dorsi rotational flap reconstruction. Two years after the second operation (2016), a follow-up mammogram detected a 3 cm non-calcified deeply located mass lesion with irregular borders in the upper outer quadrant of the left breast (Fig. 1A, B), which was confirmed on ultrasound (Fig. 1C–E). Ultrasound guided biopsy showed a spindle cell lesion with a fascicular arrangement and entrapped skeletal muscle.

* Corresponding author.
E-mail addresses: Arzu.Canan@utsouthwestern.edu (A. Canan), Xiaoqin.Wang@uky.edu (X. Wang).

https://doi.org/10.1016/j.clinimag.2018.10.025
Received 31 August 2018; Received in revised form 30 October 2018; Accepted 31 October 2018

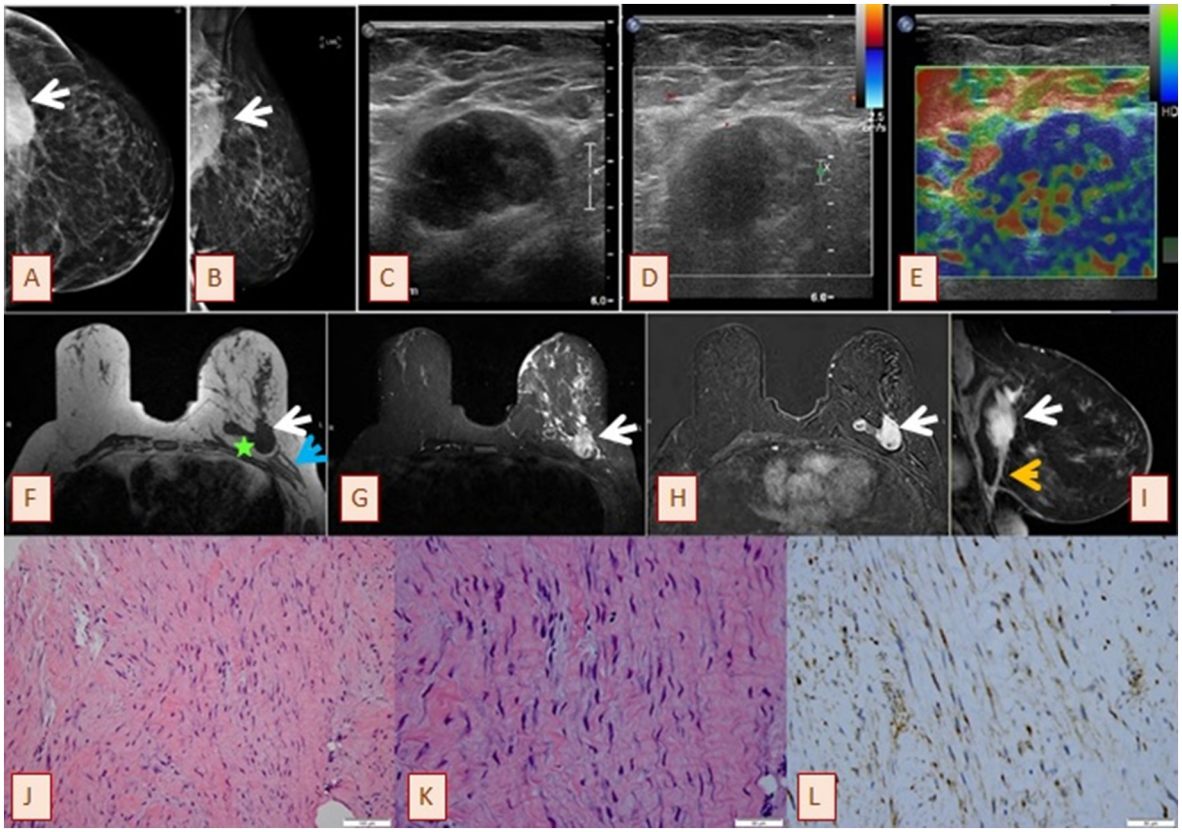

Fig. 1. Desmoid tumor in the left breast with latissimus dorsi flap found in 2016. Craniocaudal (A) and medio-lateral oblique (B) views of mammography showed non-calcified mass lesion (white arrow) with irregular borders in the upper outer quadrant of the left breast; ultrasound images demonstrated a hypoechoic oval mass (C) with circumscribed margins, measuring 3.5 cm without internal vascularity (D), intermediate stiffness on elastography (E); breast MRI images showed a lobulated mass lesion which was isointense on T1 weighted image (F) (green star—preserved fat plane between lesion and pectoralis muscle; blue arrow—latissimus dorsi flap), hyper-intense on T2 weighted image (G), with intense contrast enhancement (H), and tail like extension of the lesion (yellow arrow) probably to the flap on sagittal multiplanar image (I); H&E stained images at 20× (J) and 40× magnification (K); IHC (beta-catenin) stained image at 40× magnification (L) demonstrated histologic and immunophenotypic features of fibromatosis of the breast. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

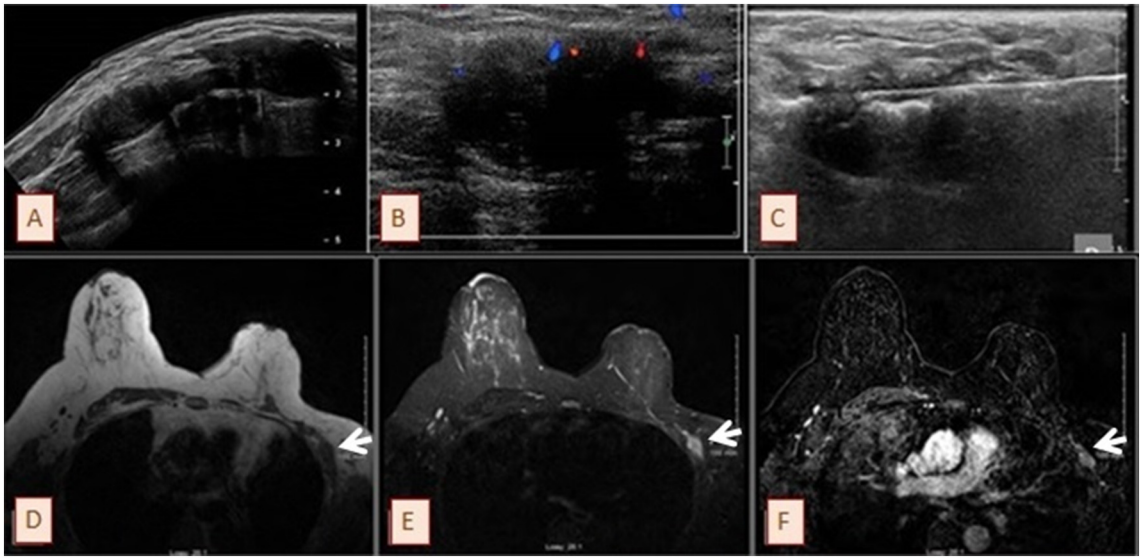

Fig. 2. Recurrent desmoid tumor in the left lateral chest wall found in 2018. Ultrasound images of the cord like lesion in left chest wall showed an irregular hypoechoic mass and marked posterior acoustic shadowing (A) without internal vascularity (B). Ultrasound guided biopsy (C) revealed recurrent desmoid tumor. Breast MRI demonstrated a mass only partially visualized in the left lateral chest wall, which is isointense on T1 weighted image (D), hyper-intense on T2 weighted image (E) with heterogeneous contrast enhancement (F).

Immunohistochemical stains were performed and the spindle cells were positive for beta-catenin (nuclear) and negative for S100 and cytokeratin AE1/AE3 (Fig. 1J–L). The morphology and immunoprofile were consistent with an aggressive fibromatosis (desmoid tumor). On the subsequent pre-operative breast MRI (Fig. 1F–I), the lesion appeared as a multilobulated, well defined and deeply located mass with persistent enhancement, measuring 7 × 4 × 7.5 cm in the lower outer left breast and in close proximity to the known latissimus flap (Fig. 1F-blue arrow). At the anterior and inferior borders of the lesion, a tail like extension probably to the aponeurosis of the latissimus dorsi flap was found (Fig. 1I-yellow arrow). A fat plane was maintained between the mass and the left pectoralis major muscle (Fig. 1F-green star). These imaging findings were considered in concordance with a desmoid tumor and suggested that the tumor likely developed from the latissimus dorsi flap.

Although a mastectomy was offered for the treatment of desmoid tumor and to prevent the high recurrence risk, the patient again chose local excision of the desmoid tumor. The lesion was removed with a wide surgical resection and all margins were free of tumor. The surgeon confirmed that the tumor arose from the aponeurosis of the latissimus muscle flap and was located in the reconstructed left breast during the operation and the latissimus dorsi flap was also removed. The patient recovered from this surgery without any complication. Two years later, the patient returned for a routine follow-up in clinic and a vague area of non-painful, cord like mass was palpated at the left chest wall near the left breast on physical examination. This palpable area was deemed as a post-surgical scar initially, but given her prior history of desmoid tumor, both breast ultrasound and MRI were performed. A focused ultrasound of this area showed an oval hypoechoic mass with indistinct margins, measuring 60 mm, located at 3 o'clock position, 15 cm from the nipple (Fig. 2A–C). The subsequent breast MRI demonstrated an ill-defined, enhancing mass, partially visualized in the lateral aspect of the left breast and mostly in the left chest wall (Fig. 2D–F). An ultrasound guided biopsy showed a spindle cell neoplasm composed of bland spindle cells with elongated nuclei and dense fibrous stroma, again consistent with aggressive fibromatosis. The treatment plan for the recurrent desmoid tumors was discussed at the multidisciplinary tumor board and with the patient. After exclusion of additional tumors within the body on CT chest, abdomen and pelvis, the decision was made to treat the recurrent desmoid tumor with a combination of surgery and hormonal therapy (tamoxifen). Radiation therapy was not performed given the patient's history of prior radiation treatment for her previous cancer (IBC-NOS) in the same breast. The patient will also have careful surveillance, including self-palpation, annual physical, mammogram and MRI examinations. The patient received a detailed explanation of her condition and the exclusion of malignancy in her case. She understands that her condition is a special kind of disease that tends to recur and requires close follow up. The patient agreed to share her information in this case report for educational purpose and to aid other patients.

3. Discussion

Desmoid tumor is a benign but locally aggressive fibrous tumor which often originates from the fascia or aponeurosis of the muscle in multiple body parts with predominance in the abdomen [16,17]. A desmoid tumor in the breast is rare. Even though its etiology is not well known, breast trauma, surgery and implants (either saline or silicone) have been reported in association with mammary fibromatosis. Recently it has been reported to originate from the prior pedicled transverse rectus abdominus myocutaneous (TRAM) flap [18]. In this case, the origin of the tumor was the aponeurosis of the latissimus dorsi flap and it re-occurred two years after the wide excision with clear margins, which was not previously reported in the literature. Besides its high local recurrent rate, the desmoid tumors arising from the musculoaponeurotic systems have an even higher incidence of recurrence [15].

Prompt diagnosis of a desmoid tumor in the breast can be challenging especially when the patient has had prior surgery and breast cancer. The most common clinical presentation of desmoid tumor in the breast is a painless palpable mass. It may cause skin or nipple retraction in some cases which may lead to the suspicion of malignancy [19,20]. Radiological imaging studies including mammography and ultrasound can facilitate early diagnosis of this entity but is not warranted since the desmoid tumor has a wide spectrum of imaging findings on mammography and ultrasound. The tumor usually presents as an irregular mass on imaging which can mimic post-surgical changes or breast carcinoma [10–13]. In this case, the first desmoid tumor was first detected as an irregular focal asymmetry on routine mammography without a palpable abnormality. The recurrent desmoid tumor was initially deemed as post-surgical change but was diagnosed with imaging and biopsy. High clinical suspicion by the radiologists and surgical team are very important in the diagnosis.

Breast MRI is the best radiological modality to characterize the soft tissue. It is recommended in patients with a history of desmoid tumors to evaluate for possible recurrence. It can also delineate the morphology of the desmoid tumor and define the extension and relationship of the tumor to the adjacent structures, therefore, MRI is almost mandatory for presurgical planning [15,21]. In our case MRI detected the actual size of the lesion which was larger than what was seen on mammography. Additionally, our patient's MRI of the first desmoid tumor not only excluded the pectoralis muscle involvement but also suggested that this desmoid tumor likely arose from the latissimus dorsi flap, which was subsequently confirmed by surgery. Of note, not all mammary fibromatosis tumors can be visualized by mammography and ultrasonography. Neuman et al. have reported that the lesion can be identified on mammography in only about one third of cases [22]. In our case, MRI evaluated the whole breast and showed more extensive tumor (7.5 cm) than on US (3.5 cm), which provides a better landscape for surgical planning, especially for this aggressive tumor with a high recurrence rate.

Although imaging modalities are very promising in the diagnosis of desmoid tumor, histopathological diagnosis is still the gold standard for final diagnosis. This is especially true for the patients with a history of breast carcinoma or surgery as the differentiation between breast carcinoma, post-operative fibrosis and soft tissue tumors such as fibrosarcoma and low grade fibromyxoid fibrosarcoma on imaging or by physical examination can be very challenging. The positive beta catenin expression in the lesion is very specific for the desmoid tumors but is not seen in other fusiform cell lesions [23].

The routine treatment for desmoid tumors is wide local resection of the lesion with clear margins. Positive margins are found to be associated with a high risk of recurrence. Hormonal therapy (most commonly tamoxifen) can be used as either a primary or adjuvant therapy especially in selected cases as some tumors were reported as estrogen and progesterone positive [16]. It was also suggested in the treatment of desmoids tumors with negative estrogen or progesterone receptors. Tamoxifen was found to be helpful for both stabilization and regression of the lesions [2]. Nonsteroidal anti-inflammatory drugs are another option to treat recurrent or nonresectable desmoid tumors with an overall response rate of 50% [24]. Radiotherapy has also been indicated to control the neoplasia and reduce the rate of recurrence [16,25]. However, due to the rarity of this tumor, there is no adequate evidence to support standardized management of desmoid tumors. Given the prior history of radiation treatment to the left breast for her previous breast cancer, no additional radiation therapy for the desmoid tumor was offered to the patient.

4. Conclusion

Desmoid tumor in the breast is an extremely rare entity and should be considered in the differential diagnosis of recurrent breast carcinoma or post-operative changes. Despite its benignity, early diagnosis is very

important for treatment because the tumor can grow rapidly and is often locally aggressive. High clinical suspicion, imaging and pathology are key for timely diagnosis. Breast MRI can define the anatomic relationship of the tumor and adjacent structures and aid in obtaining complete resection of the tumor. Wide surgical resection and clear margins can decrease the risk of local recurrence, but there is no consensus of desmoid tumor management due to limited evidence. Our experience of this unreported, complicated case will enhance our understanding and management of this rare disease.

Disclosures

Author Xiaoqin Wang is currently sponsored by grant #IRG 16-182-28 from the American Cancer Society, which has no impact on this report. Otherwise, she has no conflict of interest, grants, disclosures, or other assistance.

Author Arzu Canan declares that she has no conflict of interest, grants, disclosures, or other assistance.

References

[1] Schwarz GS, Drotman M, Rosenblatt R, Milner L, Shamonki J, Osborne MP. Fibromatosis of the breast: case report and current concepts in the management of an uncommon lesion. Breast J 2006;12:66–71.
[2] Janinis J, Patriki M, Vini L, Aravantinos G, Whelan JS. The pharmacological treatment of aggressive fibromatosis: a systematic review. Ann Oncol 2003;14:181–90.
[3] Dale PS, Wardlaw JC, Wootton DG, Resnick JI, Giuliano AE. Desmoid tumor occurring after reconstruction mammaplasty for breast carcinoma. Ann Plast Surg 1995;35:515–8.
[4] Vandeweyer E, Deraemaecker R. Desmoid tumor of the breast after reconstruction with implant. Plast Reconstr Surg 2000;105:2627–8. (19).
[5] Aaron AD, O'Mara JW, Legendre KE, Evans SR, Attinger CE, Montgomery EA. Chest wall fibromatosis associated with silicone breast implants. Surg Oncol 1996;5:93–9.
[6] Simpson RD, Harrison Jr. EG, Mayo CW. Mesenteric fibromatosis in familial polyposis: a variant of Gardner's syndrome. Cancer 1964;17:526–34.
[7] McMenamin ME, DeSchryver K, Fletcher CD. Fibrous lesions of the breast: a review. Int J Surg Pathol 2000;8:99–108.
[8] Burrell HC, Sibbering DM, Wilson AR. Case report: fibromatosis of the breast in a male patient. Br J Radiol 1995;68:1128–9. (12).
[9] Ormandi K, Lazar G, Toszegi A, Palko A. Extra-abdominal desmoid mimicking malignant male breast tumor. Eur Radiol 1999;9:1120–2.
[10] Lopez-Ruiz J, Ruiz M, Echevarria JJ, Lopez S, Imaz I. Mammary fibromatosis mimicking recurrent breast cancer: radiological findings. Eur Radiol 2005;15(9):2034–6.
[11] Shim HS, Kim SJ, Kim OH, Jung HK, Kim SJ, Kim W, et al. Fibromatosis associated with silicone breast implant: ultrasonography and MR imaging findings. Breast J 2014;20(6):645–9.
[12] Alanis L, Roth R, Lerman N, Barroeta JE, Germaine P. Radiologic images of an aggressive implant-associated fibromatosis of the breast and chest wall: case report and review of the literature. Radiol Case Rep 2017;12(3):431–8.
[13] Leibman AJ, Kossoff MB. Sonographic features of fibromatosis of the breast. J Ultrasound Med 1991;10(1):43–5.
[14] Yeo W, Teo PML, Kwan WH, Suen M. Desmoid tumor mimicking local recurrence of carcinoma of the breast. Int J Clin Oncol 1998;3(1):49–51.
[15] Brodt JK, Rhodes DJ, Glazebrook KN, Hruska C, O'Connor M, Boughey JC. Radiologic and pathologic images of mammary fibromatosis. Breast J 2011;17(2):207–9.
[16] Chummun S, McLean NR, Abraham S, Youseff M. Desmoid tumour of the breast. J Plast Reconstr Aesthet Surg 2010;63(2):339–45.
[17] Casillas J, Sais GJ, Greve JL, Iparraguirre MC, Morillo G. Imaging of intra- and extraabdominal desmoid tumors. Radiographics 1991;11:959–68.
[18] Zavlin D, Ruff ES, Albright SB. Breast desmoid tumor after ductal carcinoma treatment: salvaging a DIEP flap reconstruction. Plast Reconstr Surg Glob Open 2016;4:11.
[19] Yiangou C, Fadl H, Sinnett HD, Shousha S. Fibromatosis of the breast or carcinoma? J R Soc Med 1996;89(11):638–40.
[20] Matherne TH, Green Jr. A, Tucker JA, Dyess DL. Fibromatosis: the breast cancer imitator. South Med J 2004;97:1100–3.
[21] Milos RI, Moritz T, Bernathova M, Amann G, Panotopoulos J, Noebauer-Huhmann IM, et al. Superficial desmoid tumors: MRI and ultrasound imaging characteristics. Eur Radiol 2015;84(11):2194–201.
[22] Neuman HB, Brogi E, Ebrahim A, Brennan MF, Van Zee KJ. Desmoid tumours (fibromatoses) of the breast: a 25-year experience. Ann Surg Oncol 2008;15:274–80.
[23] Dj Lips, Barker N, Clevers H, Hennipman A. The role of APC and beta-catenin in the aetiology of aggressive fibromatosis (desmoid tumors). EJSO 2009;35:3–10.
[24] Lewis JJ, Boland PJ, Leung DH, Woodruff JM, Brennan MF. The enigma of desmoid tumors. Ann Surg 1999;229(6):866–73.
[25] Erguvan-Dogan B, Dempsey PJ, Ayyar G, Gilcrease MZ. Primary desmoid tumor (extraabdominal fibromatosis) of the breast. AJR Am J Roentgenol 2005;185(August (2)):488–9.