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Case Report

Radiologic images of an aggressive implant-associated fibromatosis of the breast and chest wall: case report and review of the literature

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ABSTRACT

Fibromatosis of the breast is a rare benign disease compromising <0.2% of all primary breast tumors. Although the chest wall is a common location, occurrences of implant-associated fibromatosis of the breast are extremely rare; only 33 cases have been reported. We present a case of a 42-year-old female who underwent breast augmentation with silicone breast implants, and 2 years later developed an aggressive implant-associated fibromatosis of the breast and chest wall. On imaging studies, the tumor mimicked breast carcinoma, and despite chemotherapy, the fibromatosis rapidly enlarged and was locally invasive requiring wide surgical excision. Unlike previously reported imaging findings, magnetic resonance imaging revealed an oval circumscribed mass with fringe-like internal architecture. We provide a review of the literature and discuss the imaging features of implant-associated fibromatosis of the breast.

Introduction

Extra-abdominal desmoid tumor of the breast, mammary fibromatosis, or fibromatosis of the breast is a type of extremely rare breast tumor [1]. Unlike fibromatosis in the abdominal wall which is frequently reported, fibromatosis of the breast constitutes <0.2% of all primary breast tumors [1–4]. Fibromatosis of the breast is a benign, nonmetastasizing stromal tumor that can grow aggressively and invade locally in an infiltrating pattern and can mimic malignant

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types of breast cancer on both clinical and imaging findings [1,3,5–8]. Fibromatosis of the breast is more common in young and fertile women, but occurrences in men have been reported [1,6]. Patients range in age from 13 to 83 years old, with a reported mean age at diagnosis between 37 and 50.3 years [6,9].

Case report

A 42-year-old woman with breast implants presented to our institution with the chief complaint of a palpable lump in her left breast for 5 months, with more dramatic growth 1 month before her visit. She denied any skin changes, nipple retraction, or discharge. On clinical examination, the left breast was noted to be asymmetrically enlarged with a palpable firm mass in the left upper inner breast adjacent to the left sternal border. The patient reported minimal tenderness, and no nipple or skin changes were noted. Two years before her presentation, she had undergone bilateral breast augmentation with silicone implants. A month before her presentation, mammography at an outside institution revealed the left breast to be extremely dense, limiting sensitivity of mammography, without any obvious mass, though, accompanying ultrasound of the left breast revealed a 6.6-cm hypoechoic heterogeneous solid vascular mass anterior to the implant capsule at the 11-o'clock position. Repeat ultrasound at our institution confirmed presence of a suspicious hypoechoic mass with posterior acoustic shadowing (Figs 1A and B). A left breast ultrasound-guided 14-gauge core biopsy was performed; pathology revealed spindle cell proliferation compatible with fibromatosis of the breast. Immunohistochemical stains revealed the spindle cells were positive for vimentin, beta-catenin, and with foci of positivity for smooth muscle actin. The lesion had predominantly negative results for CD34, CD54, S100, estrogen receptor antibody, CK 903, and pan-cytokeratin; the findings further supported the diagnosis of fibromatosis [10–13]. A week following the core biopsy, a contrast-enhanced computed tomography (CT) of the chest was performed demonstrating a hypoattenuating circumscribed mass in the left breast compatible with known fibromatosis causing anterolateral displacement of the breast implant (Figs 2A and B). Surgical resection was considered, though given the size and local extension of the tumor, chemotherapy was initiated in an attempt to decrease tumor size before resection as this treatment has been shown to have the highest response rate. The patient underwent weekly methotrexate and vinorelbine (Navelbine; Pierre Fabre Medicament Production, France) for 3 months; however, the mass significantly grew in size despite chemotherapy. A 3-month follow-up contrast-enhanced CT of the chest showed an interval increase in size of the fibromatosis of the left breast despite chemotherapy (Figs 3A and B). Five days following the CT of the chest, a bilateral contrast-enhanced breast magnetic resonance imaging (MRI) demonstrated an 11.2-cm mass in the left breast with marked distortion of the left breast implant (Figs 4A and B). Four weeks later, the patient underwent surgical excision of the left breast tumor and removal of the bilateral breast implants. Gross examination revealed a 12 × 9 × 4.5 cm pink-tan soft-tissue mass with a glistening smooth surface. On sectioning, scattered areas of hemorrhage were noted without gross evidence of necrosis or calcifications. Microscopic examination demonstrated a proliferation of bland-appearing spindle and stellate cells in intersecting fascicles morphologically compatible with desmoid-type fibromatosis (Fig. 5). The lesion had an infiltrative edge, involving the fascia and extending to the inked specimen margin (Fig. 6). Immunohistochemical stains performed on this resection specimen showed a similar immunoprofile to the previous core biopsy, with strong nuclear positivity for beta-catenin (Fig. 7), positivity for smooth muscle actin, focal weak positivity for desmin, and negative expression of panCK, CAM 5.2, CD34, S100, and CD117. The Ki-67 revealed a proliferation index of ~1%. The patient resumed chemotherapy postoperatively. A follow-up bilateral contrast-enhanced breast MRI revealed no residual tumor mass within the left chest wall or breast (Figs 8A and B). The patient was referred to radiation oncology to discuss the option of future radiation to the chest wall given the positive surgical margins at the time of surgery.

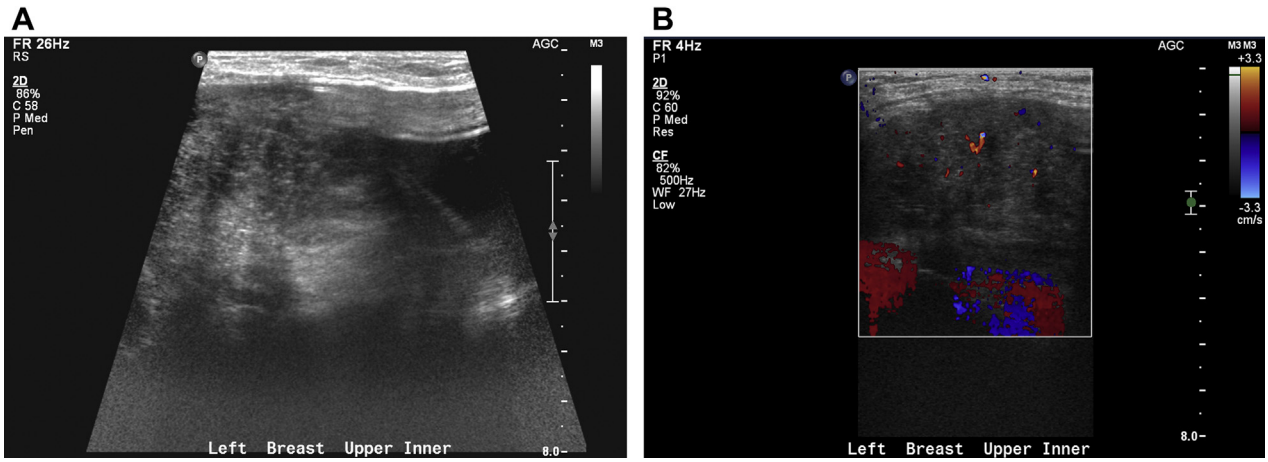

Fig. 1 – (A) Ultrasound images of the left upper inner quadrant shows a 7.6-cm hypoechoic mass with irregular margins and posterior acoustic shadowing. (B) Color Doppler ultrasound image shows increased vascularity within and around the lesion.

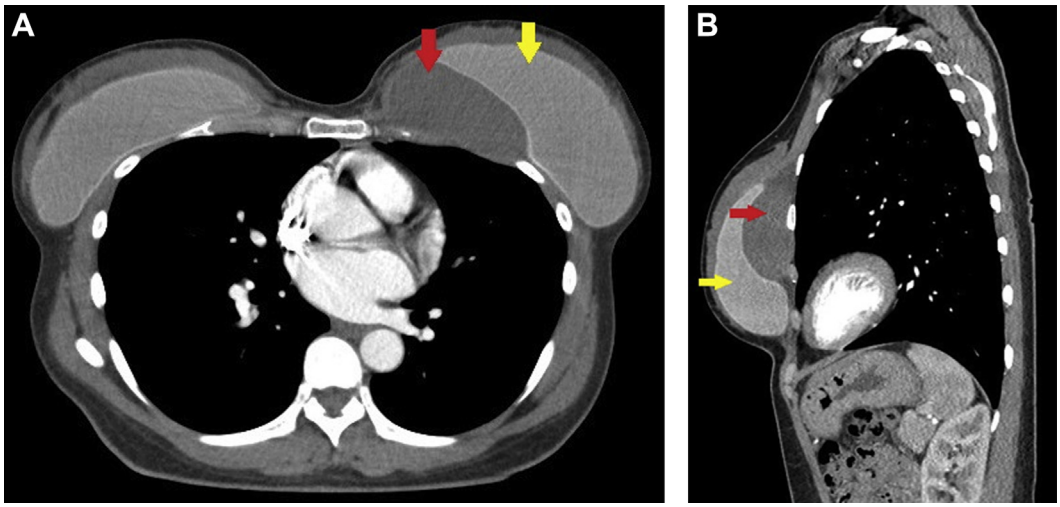

Fig. 2 – (A) Axial and (B) sagittal images of a contrast-enhanced CT of the chest reveals a 3.5 cm × 7.7 cm × 7.3 cm hypoattenuating lesion (red arrow) deep and medial to the left breast implant (yellow arrow).

Discussion

Clinically, fibromatosis unusually presents as a solitary, palpable, mobile, firm, and typically painless mass [6,9]. Some patients may report skin and nipple retraction [4,6,9]. Fibromatosis of the breast is often unilateral, but up to 4% of patients may present with bilateral fibromatosis [6].

On gross pathology, these tumors range in size from 0.3 to 15 cm and are firm, poorly circumscribed, and white or gray in color [6]. Pathologically, these lesions are characterized by spindle cell proliferation of fibroblastic cells resembling desmoid-type fibromatosis arising in other sites [2,3,6]. Fringe-like extensions may extend from the periphery of the lesion into adjacent breast parenchyma and adipose tissue [4]. Definitive diagnosis is based on histology, which shows lesions composed of bundles of long sweeping and intersecting spindle cells with varying amounts of collagen deposition and low mitotic rate [2–4,6]. The histologic differential diagnosis includes benign and malignant entities such as low-grade fibrosarcoma which usually shows increased cellularity with more pronounced cytologic pleomorphism and atypia and increased mitotic activity; metaplastic, or spindle cell carcinoma which usually has areas of cohesive atypical epithelium and immunohistochemical cytokeratin staining; nodular fasciitis which usually presents as a tender, rapidly growing subcutaneous mass and histologically contains abundant inflammatory cells dispersed in the loose myxoid stroma; phyllodes tumor which usually has epithelial-lined clefts and CD34 positivity; and scar and biopsy-site reaction [1,2,4,6,9]. Fibromatosis of the breast typically

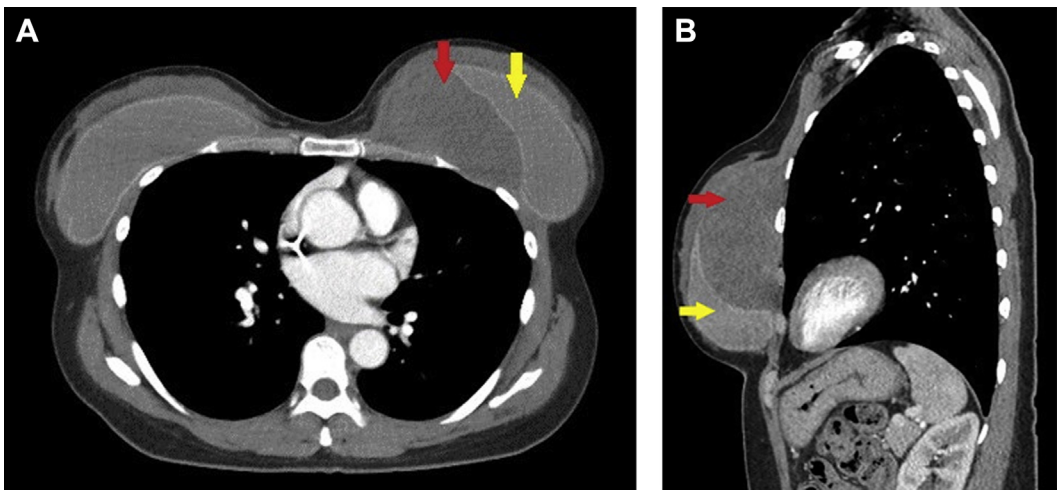

Fig. 3 – (A) Axial and (B) sagittal images of a contrast-enhanced CT of the chest reveals a 9.4 cm × 5.3 cm × 9.8 cm hypoattenuating lesion (red arrow) deep and medial to the left breast implant (yellow arrow) which increased in size over a 3-month period.

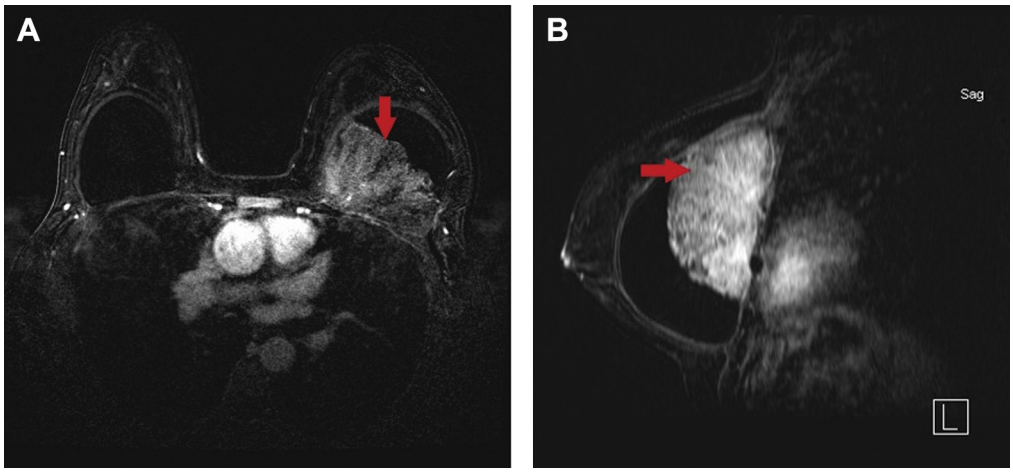

Fig. 4 – (A) Axial and (B) sagittal T2 fat-saturated postcontrast images from a contrast-enhanced bilateral breast MRI demonstrates a 5.8 cm × 8.5 cm × 11.2 cm hyperintense mass with fringe-like appearance (red arrow) posteromedial to the left breast implant.

exhibits nuclear positivity for beta-catenin and lacks estrogen receptor and progesterone receptor positivity [6].

On mammography, these lesions usually present as irregular, noncalcified, high-density masses with spiculated margins and can be mistaken for breast cancer [1,4,6,7]. On ultrasound, these tumors are usually solid, spiculated or microlobulated, irregular hypoechoic mass with posterior acoustic shadowing, and straightening and tethering of Cooper ligaments [1,4,7].

On breast MRI, fibromatosis of the breast has morphologic features similar to malignancy such as spiculated margins [3]. In 90% of cases, they present as ill-defined hypo- to isointense masses on T1-weighted images and heterogeneously hyperintense on T2-weighted images [3,4,6]. However, in contrast to invasive breast cancer which typically enhances and washes out rapidly, fibromatosis has a persistent enhancement pattern [3]. The mass may display variable signal intensity on T2-weighted imaging which can be explained by variations in the quantity of collagen or myxoid stroma across portions of the tumor [13]. MRI is the best imaging technique for evaluation of tumor extent, pectoralis muscle involvement, and chest wall involvement before surgical planning [1,3,4].

The etiology of these tumors is not well understood. Fibromatosis of the breast can occur anywhere in the breast and has been reported to arise primarily within the breast tissue or from the underlying pectoralis major muscle [4]. An association with Gardner syndrome, surgical trauma, or breast augmentation with saline or silicone breast implants has been reported [1,2,4,6,9]. Clonality of desmoid-type fibromatosis has been described [14]. Although most cases are sporadic, some may arise in patients with familial adenomatous polyposis. Activating mutations of the beta-catenin gene have been described in up to 45% of cases and mutations in the adenomatous polyposis coli gene or 5q loss in 33% [15].

Occurrences of implant-associated fibromatosis of the breast, as was the case with our patient, are extremely rare; only 33 cases have been reported (Table 1)

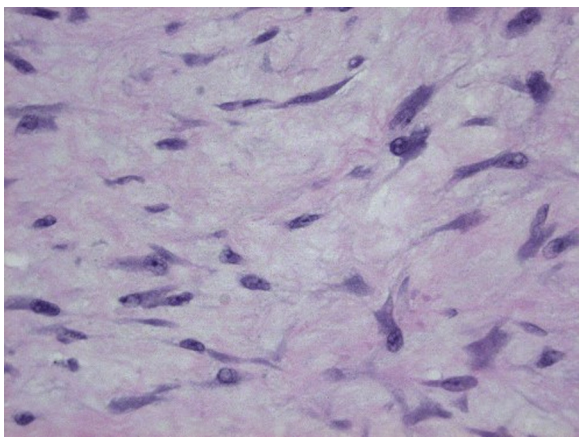

Fig. 5 – Fibromatosis showing bland-spindle and stellate cells in a loose background. Hematoxylin and eosin stain (H&E), 400×.

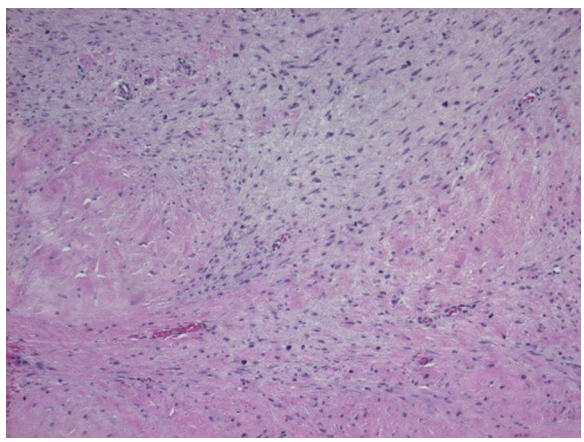

Fig. 6 – Fibromatosis with infiltration of adjacent fascia. H&E, 100×.

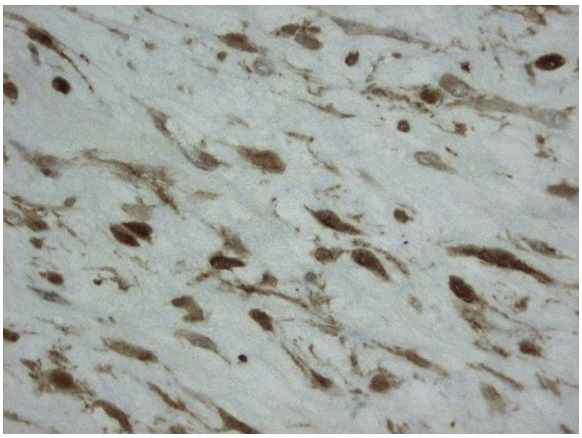

Fig. 7 – Characteristic nuclear staining with beta-catenin. Beta-catenin immunohistochemistry, 400×.

[10–13,18–20,22–24,29,30,34,35]. It is thought that fibromatosis arises from the fibrous capsule surrounding the breast implant, as reported cases show the tumor arising in conjunction with, or adjacent to, the fibrous capsule [1,6,19,22,24,29,30]. However, the exact etiology of implant-associated fibromatosis remains unclear [13]. Of the 33 cases listed, 16 involved silicone implants, 8 involved saline implants, 2 involved both silicone and saline implants; 7 cases did not include information on the implant type. However, a strong association between fibromatosis of the breast and silicone implants cannot be assumed. Instead, this likely stems from the fact that silicone implants are more prevalent than saline implants. The mean interval time between implant placement and tumor occurrence was 3 years (Table 1). The median tumor diameter was 7.6 cm. Three patients received adjuvant radiotherapy, 2 received chemotherapy, 1 received chemotherapy and hormone therapy, 1 received hormone therapy, and 1 received anti-inflammatory medication [11,22–25,29,30]. Local recurrence occurred in 5 cases; of those, 3 experienced 2 recurrences each [17,22,29].

On imaging, fibromatosis of the breast parenchyma presents as a noncalcified spiculated mass. In contrast, implant-associated fibromatosis has been reported to have a different appearance on CT and MRI imaging. Among the 33 reported cases, 10 cases had imaging data. In all 10 of these cases, the tumor had a well-defined border. Prior studies have reported implant-associated fibromatosis as having a smooth margin on both CT and MRI with a band-like hypointense portion on MRI [13,33]. However, our imaging findings are different from those previously reported. In our case, the implant-associated fibromatosis presented as an oval circumscribed tumor with anterior displacement of the implant with a fringe-like internal architecture on MRI but without a hypointense band-like lesion. The fringe-like internal architecture was also observed on ultrasound likely corresponding to spindle cell proliferation of fibroblastic cells on pathology.

Optimal management of fibromatosis of the breast remains controversial. However, the current treatment of choice is wide local excision with negative margins [2–4]. If tumor invasion involves surrounding structures such as the skin or chest wall, then resection of the involved structures to obtain clear margins should be performed [3]. Local recurrence rates can be high, particularly with positive margins, and have been reported in 21%-29% [1–3,5]. Fibromatosis of the breast usually recurs within 3 years of excision and requires radical excision [3,4,6,22]. The size, atypia, cellularity, and number of mitoses cannot predict likelihood of recurrence [9].

Studies have reported the use of interferons, tyrosine kinase inhibitors, cytotoxic agents, radiation, hormone therapy, and/or nonsteroidal anti-inflammatory drugs to help control or eradicate recurrent tumor with variable success [1,6,10,11,20,30,36]. Numerous cytotoxic agents have been found to be effective in the management of fibromatosis of the breast [10]. Some authors have reported tumor regression with conventional low-dose chemotherapy with a weekly schedule of vinblastine and methotrexate [1,6,24]. Others report achieving a 10-year progression-free interval of 67% on a similar combination of vinblastine and methotrexate [10]. However, this was not the case in our patient whose tumor grew while on chemotherapy over a 3-month period.

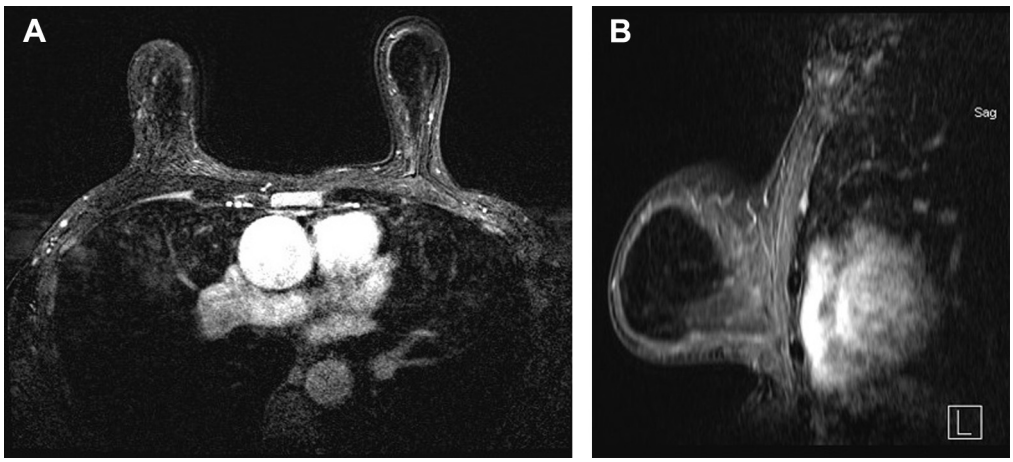

Fig. 8 – Contrast-enhanced bilateral breast MRI. (A) axial and (B) sagittal images following the removal of the bilateral breast implants and resection of the fibromatosis of the left breast reveals no residual abnormal areas of enhancement at the site of the tumor in the left chest wall or breast.

Table 1 – Characteristics of reported cases of implant-associated fibromatosis of the breast.

Reference	Age (y)	Biomaterial	Interval (y)	Tumor size (cm)	Treatment	Outcome
Jewett and Mead, 1979 [16]	54	Saline	2	3	WE	RF 8 mo
Rosen and Ernsberger, 1989 [17]	35	Saline	"Several years"	NR	Excision	First RE 7 mo, second RE 18 mo, RF 12 mo
Schuh and Radford, 1994 [18]	41	Silicone	2	6.5	WE	RF 36 mo
Schiller et al, 1995 [19]	66	Silicone	NR	13	WE	NR
Dale et al, 1995 [20]	65	Silicone	7	13	WE	NR
Crestinu, 1995 [21]	NR	Silicone	2	NR	WE	RF 90 mo
Aaron et al, 1996 [22]	43	Saline	2	NR	WE + RT	first RE 24 mo, second RE 6 mo, RF 12 mo
Vandeweyer and Deraemaecker, 2000 [23]	45	Saline/silicone	3	3	WE + RT	RF 24 mo
Abraham et al, 2002 [15]	55	Silicone	NR	6	Excision	NR
Khanfir et al, 2003 [24]	52	Saline	1.6	8	CHT + WE + HT	RE 8 mo
Jandali et al, 2004 [25]	24	Silicone	9	6	WE + RT	RE 36 mo, RF 21 mo
Gandolfo et al, 2006 [26]	22	Silicone	2	16	WE	NR
Jamshed et al, 2008 [27]	30	Saline	3	6.0	WE	RF 24 mo
Neuman et al, 2008 [28]	5 cases of implant-associated breast fibromatosis					
Balzer and Weiss, 2009 [29]	64	Silicone	1.8	6.7	WE	RF 40 mo
	NR	Silicone	2	4.5	WE	RF 48 mo
	37	Silicone	2.5	3.3	WE	RF 42 mo
	28	Saline	2	11	WE	RF 36 mo
	38	Silicone/Saline	2	12	Partial excision	first RE 24 mo, second RE 19 mo, AWD 46 mo
	29	Silicone	2	7.4	CHT	AWD 92 mo
Chummun et al, 2010 [10]	22	Silicone	2	5	Excision	RF 5 mo
Matrai et al, 2011 [30]	34	Silicone	2	9	Excision + HT	RF 55 mo
Gergele et al, 2012 [11]	43	NR	3	6.7	Anti-inflammatories	NR
Hammoudeh and Darian, 2012 [12]	38	Saline	4	3.5	Excision	RF 12 mo
Mazzocchi et al, 2009 [31]	52	Silicone	4	6	WE	RF 36 mo
	38	Silicone	7	1.5	WE	RF 12 mo
Jeong et al, 2013 [32]	34	Silicone	2	6.4	Excision	NR
Shim et al, 2014 [33]	29	NR	2	7	WE	RF 8 mo
Seo et al, 2015 [13]	27	Saline	2	7.5	Partial excision	NR
Present study, 2016	42	Silicone	2	12	WE + CHT	RF 2 mo

AWD, alive without disease; CHT, chemotherapy; HT, hormonal therapy; NR, not reported; RE, recurrence; RF, recurrence-free; RT, radiotherapy; WE, wide excision.

Postoperative radiation therapy has been shown to control residual tumor and significantly reduce the rate of recurrence in patients with unresectable disease, gross residual disease, or positive margins following resection [6,10,11,22,36,37]. Some studies have shown that postoperative radiation therapy can improve the 10-year recurrence-free survival rate in patients with positive margins (54% without radiation vs 24% with radiation) [4,37].

In conclusion, implant-associated fibromatosis of the breast is a rare benign disease that may develop adjacent to breast implants. Our case report describes a case of an aggressive implant-associated fibromatosis in a patient with bilateral breast implants mimicking carcinoma, and whose fibromatosis rapidly enlarged despite being on chemotherapy. The diagnosis should be considered when a smoothly marginated mass with fringe-like appearance is seen on MRI adjacent to a breast implant in a patient with history of breast augmentation. Tissue sampling is necessary to exclude malignancy. Although there is no definite association between fibromatosis of the breast and breast augmentation, close observation with physical examination and imaging is recommended to ensure early diagnosis and proper management in women with history of breast augmentation.

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