Case Report

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Bilateral Inflammatory Pseudotumour of the Breast: A Case Report and Review of the Literature

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ABSTRACT

Inflammatory tumour (IPT) consists of spindle cells, mature plasma cells, histiocytes, lymphocytes and eosinophils. Most frequently presenting in the respiratory tract it can also affect other sites such as breast. This case was a 73-year old woman presenting with a left breast lump, clinically indeterminate (P3), proven on biopsy to be IPT. Seven years later she returned with bilateral breast lumps and underwent triple assessment followed by wide excisions which confirmed the diagnosis of IPTs. Because it can be difficult to differentiate IPT from a low-grade spindle cell metaplastic breast carcinoma (SpCMBC) wide excision to achieve clear margins should be achieved to exclude malignancy.

Keywords: Breast, inflammatory myofibroblastic tumour, inflammatory pseudotumour, breast cancer

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Introduction

Inflammatory pseudotumour (IPT) is a rare benign condition usually seen in the lungs and airways of young individuals. There is a multiplicity of nomenclature for this lesion including pseudomalignant spindle cell proliferation, plasma cell granuloma, post-inflammatory tumour, pseudosarcomatous fibromyxoid tumor, nodular fasciitis, inflammatory pseudotumour, post-operative spindle cell nodule, or pseudosarcomatous myofibroblastic proliferation (1).

It comprises spindle cells admixed with mature plasma cells, histiocytes, lymphocytes and eosinophils. Extrapulmonary sites include the gastrointestinal tract, urinary tract, retroperitoneum, peritoneum, mesentery, pancreas, spinal cord meninges, intracranial spaces, liver, thyroid, spleen and lymph nodes (1). It occurs very rarely in the breast and was first described by Pettinato et al. (2) in 1988. Bilateral inflammatory pseudotumour of the breast has been reported in only two previous cases (1, 2). Treatment is usually excision biopsy because many are suspected pre-operatively to be malignant.

Case Presentation

A 73-year-old woman was initially seen in 2004 in the Breast Clinic at Guy's Hospital with a right breast lump. This measured 15 mm clinically and mammograms and ultrasound revealed a suspicious abnormality (E3, M3, U4). She underwent ultrasound-guided core biopsies and these showed acute and chronic inflammation so the patient was reassured. The lump persisted and was re-imaged in 2005. Imaging was reported as showing signs of unequivocal malignancy (M5, U5), (Table 1). Repeat core biopsy showed chronic inflammation and fibrosis. Imprint cytology however showed atypia (C3) with few small cohesive groups of epithelial cells with superimposed myoepithelial cells, mild nuclear atypia, and fragments of degenerate, vascularised connective tissue stroma.

The patient was advised to have an excision biopsy of the lump to which she agreed. Histology showed an ill-defined lesion composed of an irregular proliferation of fibrous connective tissue associated with severe inflammatory reaction. The spindle cells were without atypia or significant mitotic activity. The inflammatory component consisted of plasma cells, lymphocytes and eosinophils. Additionally, there was inflammatory cell infiltration of blood vessel walls. Immunohistochemistry showed the lymphocytic infiltrate to be polyclonal and light chain restriction was not demonstrated. Final histology was in keeping with an inflammatory pseudotumour (IPT).

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Table 1. Sequential triple assessment findings in patient with IPT

Year	Clinical	Mammo	USS	Cytology	Core
2004	R E3 L E1	R M3 L M2	R U4 L U1	R C3	B2
2005	R E4 L E1	R M5 L M1	R U5 L U1	R C3	
2007	R E2 L E1	R M2 L M1	R U2 L U1	-	
2011	R E3 L E1	R M3 L M4	R U5 L U5		R B2 IPT L B2 IPT

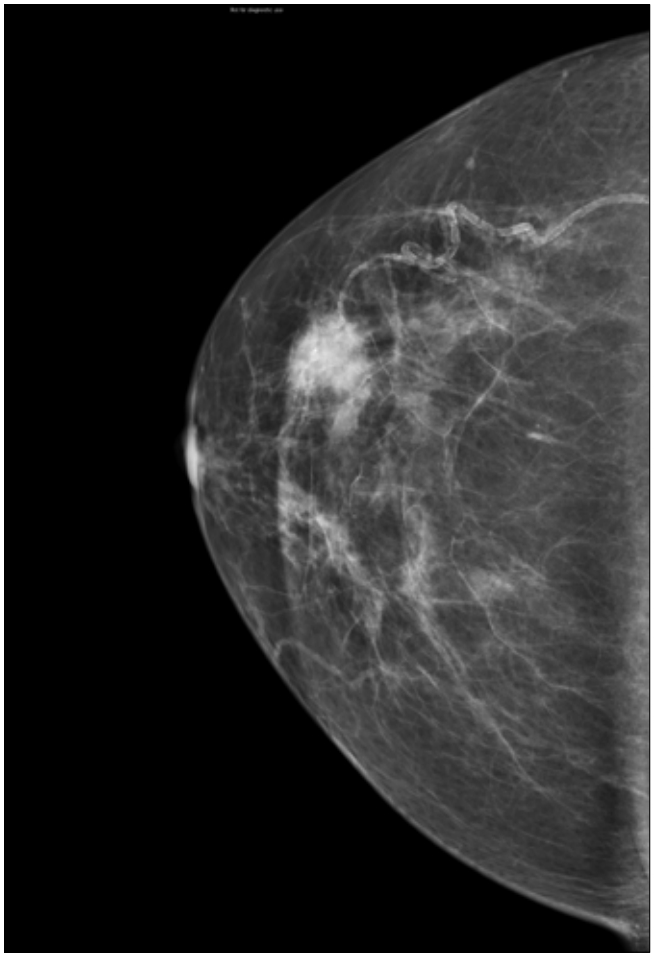

Figure 1. Right mammogram CC view

During follow-up in 2007 she was asymptomatic and there was no abnormality on clinical examination. Bilateral mammograms and breast USS showed benign changes only (M2, U2). She was reassured and discharged.

She returned to the Breast Clinic in 2011 giving a 3-month history of right breast lump. On clinical examination there was a 15-mm mass lateral to the biopsy scar in the upper outer quadrant of right breast. The mass was hard with an irregular surface, but not fixed to skin or chest wall. Right mammogram (MMG) (Figure 1, 2) showed an indeterminate (M3) area of increased density in the upper outer aspect. Left MMG (Figure 3, 4) showed a new area of suspicious asymmetrical

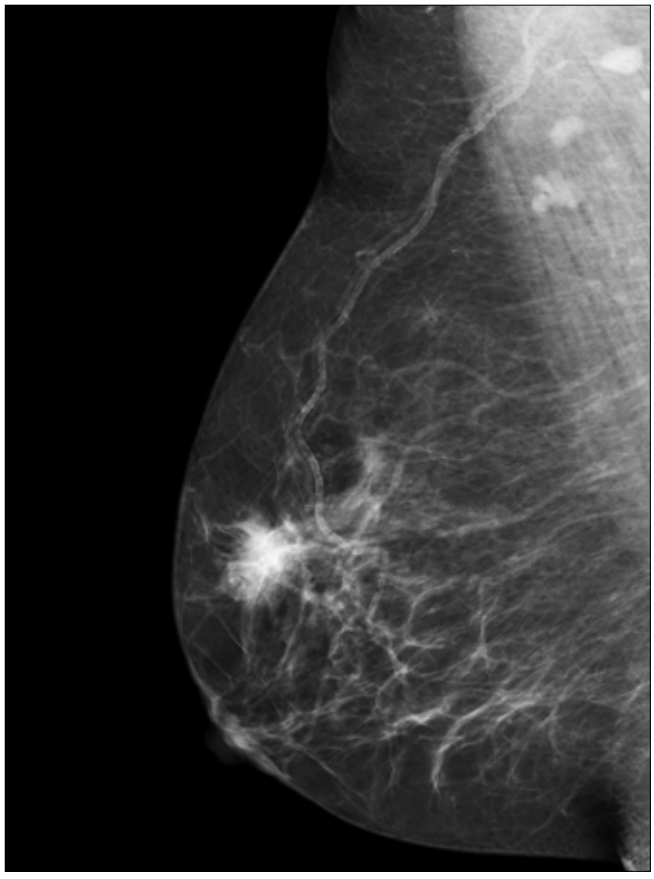

Figure 2. Right mammogram MLO view

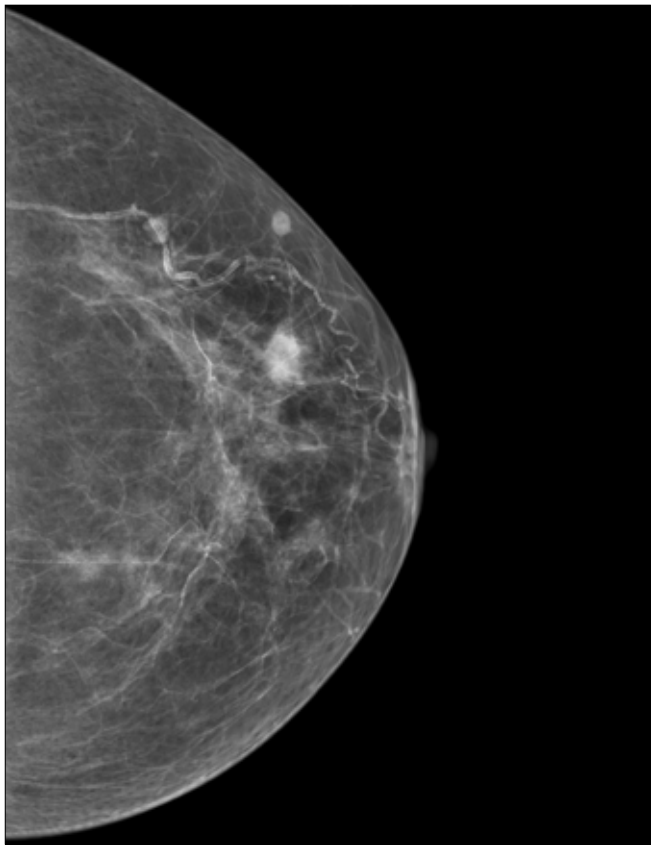

Figure 3. Left mammogram CC view

Table 2. Patients with inflammatory pseudo tumours of the breast

Author	Patient age	Side	Size (mm)	SMA	Surgery	IgG4	ALK	PAN CK	Follow-up (years)
Pettinato 1988	29	R		+ve	e/b				2.5
Coffin 1995	13	R			e/b				1
Bisceglia 1995	38				e/b				
Chetty 1997	16	R	20	+ve	e/b				1
	18	R	20	+ve	e/b				1
	46	R	80	+ve	e/b				0.5
Yip 1997	66	Bil	30/20		e/b				0.75
Gobbi 1999	86	L			e/b				
Sastre-Garau 2002	64	R	20	+ve	e/b		-ve	-ve	
Zardawi 2003	79	Bil			e/b				9
Haj 2003	31	R	60		e/b			-ve	-
Zen 2005	46	L	16		e/b	+ve			1
Ilvan 2005	60	R	10		e/b			-ve	7
Khanafshar 2005	33	L		+ve	e/b		-ve	-ve	
	47	R		+ve	e/b		-ve	-ve	
	75	L		+ve	e/b		-ve	-ve	
Akbulut 2007	38	L	10	+ve	e/b		-ve	-ve	1
Park 2009	47	R	30	+ve	e/b		-ve	-ve	3
Kim 2009	60	L	15	+ve	e/b		-ve	-ve	2
Hill 2010	53	R	50	+ve	e/b	Sparse	-ve		-
Sari 2011	54	L	30	+ve	e/b			+ve	1/3
Vecchio 2011	22 (male)	L	70	+ve	e/b		-ve		10/12
Chougule 2015	66	L	30	-ve	e/b	+ve	-ve		1.5
	45	R	15	-ve	e/b	+ve	-ve		1
Greenleaf 2016	69	R	23		e/b				-
Goto 2016	52	L	30		Steroid				0.75
Present case	73	Bil	30	+ve	e/b	+ve	-ve	-ve	7

e/b: excision biopsy; SMA: smooth muscle antigen; ALK: anaplastic lymphoma kinase; IgG4: immunoglobulin G4; PanCK: pancytokeratin antigen

density (M4) in the upper outer aspect. These findings were confirmed on compression views. Breast ultrasound scan (USS) confirmed 14x15 mm irregular, hypoechoic, highly vascular solid mass (U4) on the right and 12x9 mm irregular, hypoechoic lesion (U5) on the left (Table 1). Core biopsies were taken from both the lesions under USS control. Both biopsies showed features similar with those in the surgical specimen from 2005 of fibrotic breast tissue with a spindle cell proliferation and associated chronic inflammatory cell infiltrate. There was no atypia or mitotic activity. The inflammatory infiltrate consisted of plasma cells, lymphocytes and eosinophils.

Immunohistochemistry showed mild infiltrate of IgG4 positive plasma cells in the right breast lesion (8 IgG4 positive cells per HPF) and a moderate infiltrate in left breast specimen (11 IgG4 positive cells per HPF). Bilateral wide excision of the lesions was performed. Because the right sided specimen was close to all margins a re-excision was undertaken which achieved a clear margin.

Discussion and Conclusion

Only 27 cases have been previously reported in the English literature and the outline details of these and the present case are given in Table 2 (1-20). Median age was 47 years (range 13-86) with 14 aged <50 years and 13 aged ≥50. Of the unilateral cases with known laterality, 13 were right-sided and 10 located on the left so that this does not mirror the slightly increased frequency of left sided breast cancers (1). This is the third case, in whom, after initial surgical excision of IPT from one side, the patient subsequently presented with bilateral recurrence (4, 5). Treatment was almost always excision biopsy but Goto et al. (21) treated the patient with prednisolone after core biopsy with resolution of the lump.

Vecchio et al. (1) recently described the first male case with a large mass occurring after trauma. The most striking histological features was the presence of large pleomorphic cells with hypercellularity and

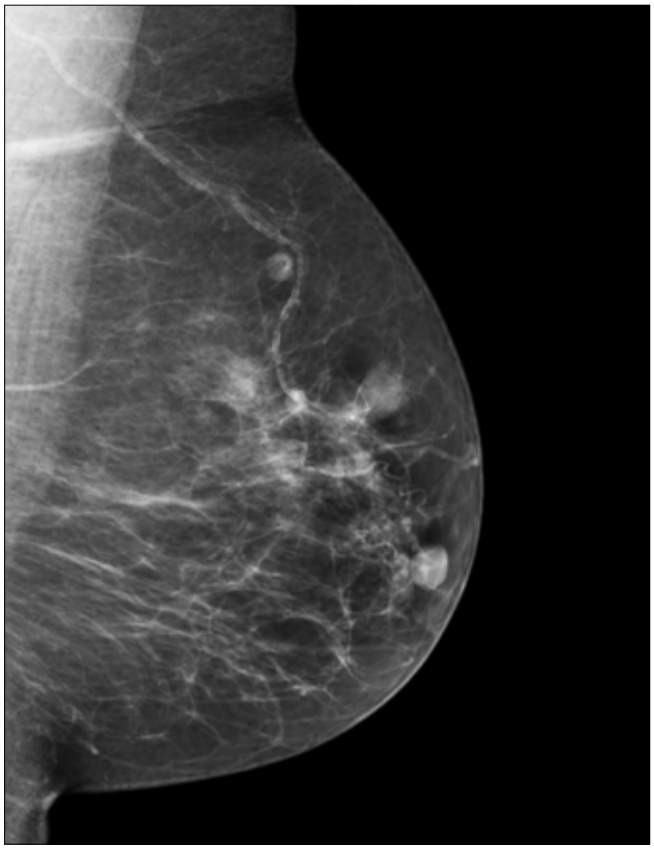

Figure 4. Left mammogram MLO view

a prominent fascicular pattern. The lesion bore a close resemblance to inflammatory myofibroblastic tumor (IMT) with atypical features but was negative for ALK-1 protein on immunohistochemistry. The potential for misdiagnosis and the broader spectrum of morphology in IPT was stressed.

Immuno-histochemistry is important in diagnosis of this entity as histological features are consistent with chronic fibrosis and inflammation. Presence of myofibroblasts has been confirmed in almost all cases by SMA positivity. Serum IgG4 has been shown to be high in IPT of breast (11) however in this case, levels were within the normal range preoperatively. The association between IgG4 and IPT was first reported in pancreatic pseudotumour, also known as sclerosing pancreatitis. Serum IgG4 was shown to be significantly and specifically higher in patients with sclerosing pancreatitis, compared to other pancreatic conditions (22).

Inflammatory pseudotumour has generally been regarded as a benign condition, but is known to recur. In some cases, there is local inflammation with systemic symptoms that resolve after resection. Sastre-Garau et al. (9) carried out a cytogenetic analysis of a breast IPT and found that the lesion was clonal with a 9p deletion and suggested that this was a low grade malignant lesion. Reis-Filho (23) however pointed out that clonal origin was not necessarily an indication of malignancy and that because the tumour described by Sastre-Garau et al. (9) had been incompletely characterised in terms of p63, maspin, E- and P-cadherin expression, it was possible that the lesion was a low-grade spindle cell metaplastic breast carcinoma (SpCMBC) (24). Hence differentiation from malignant condition along with wide excision to achieve clear margins should be the diagnostic and therapeutic aims in the management of this rare benign breast condition

Informed Consent: Written informed consent was obtained from patient who participated in this study.

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