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Alya Binmahfouz, Karin Steinke

ABSTRACT

Introduction: Primary lymphoma of the breast is very rare, comprising 0.5% breast neoplasms. The majority of breast lymphomas are of B cell type, postulated to originate from intramammary lymph nodes or breast lymphatics. Moreover, a still unresolved entity known as breast implant-associated anaplastic large T cell lymphoma (BIA-ALCL) has emerged over the last 20 years, its incidence believed to be on the rise, as the prevalence of women with breast implants is increasing.

Case Report: We report an extremely uncommon presentation of BIA-ALCL in a 55-year-old lady who presented 23 years post-bilateral cosmetic breast implants with a locally aggressive mass in the left breast. The mass invaded the chest wall and was associated with left axillary and internal mammary lymph nodes. We review the medical imaging and histopathologic findings of this mass and briefly discuss the different presentations and recommended treatment options.

Conclusion: This report reinforces the importance of understanding the possible inherent complications and variable clinical presentations associated with breast implants, in order to assist with early recognition and prompt management of this recently emerging, potentially fatal disease.
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Introduction: Primary lymphoma of the breast is very rare, comprising 0.5% breast neoplasms. The majority of breast lymphomas are of B cell type, postulated to originate from intramammary lymph nodes or breast lymphatics. Moreover, a still unresolved entity known as breast implant-associated anaplastic large T cell lymphoma (BIA-ALCL) has emerged over the last 20 years, its incidence believed to be on the rise, as the prevalence of women with breast implants is increasing. Case Report: We report an extremely uncommon presentation of BIA-ALCL in a 55-year-old lady who presented 23 years post-bilateral cosmetic breast implants with a locally aggressive mass in the left breast. The mass invaded the chest wall and was associated with left axillary and internal mammary lymph nodes. We review the medical imaging and histopathologic findings of this mass and briefly discuss the different presentations and recommended treatment options. Conclusion: This report reinforces the importance of understanding the possible inherent complications and variable clinical presentations associated with breast implants, in order to assist with early recognition and prompt management of this recently emerging, potentially fatal disease.

Keywords: Anaplastic T cell breast lymphoma, Breast implant, Implant associated breast lymphoma, Implant complication

INTRODUCTION

Primary lymphoma of the breast is very rare, comprising 0.5% breast neoplasms, most of them being of B cell type, postulated to originate from intramammary lymph nodes or breast lymphatics [1]. Moreover, a still unresolved entity, known as breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) has emerged over the past 20 years [2], with its incidence is believed to be on the rise, as is the prevalence of women with breast implants.

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A 55-year-old female presented to her general physician complaining of left breast swelling, pain and tenderness, first noticed five months prior. No nipple changes or discharge were reported. She has had a bilateral augmentation mammoplasty 23 years prior for cosmetic reasons. She was married, a smoker, a social drinker and had three children. She did not take regular medications, and had no family or personal history of breast cancer or lymphoma.

On examination, the breasts were asymmetric; the left breast was grossly enlarged and tender to palpation with overlying erythema. No nipple retraction, skin ulceration or axillary lymph nodes were noted. The right breast and axilla were unremarkable.

Her most recent mammogram and breast ultrasound scan done 1 year before presentation were unremarkable, apart from bilateral subpectoral intact saline implants. A sonogram of the left breast was performed and showed an irregular contour of the implant suggestive of rupture, with edema and generalized increased vascularity. No definite solid mass or cystic lesions were detected. Bilateral mammography also showed an irregular contour of the left implant, with a suspicious density seen superiorly only on mediolateral oblique (MLO) view, no associated calcification (Figure 1). A contrast-enhanced dynamic bilateral breast MRI scan showed intact implants. However, an irregular, lobulated, thick-walled, rim-enhancing mass with central cystic changes, measuring 7x5 cm was detected posterosuperior to the left implant, invading the left chest wall and pleura and abutting the pericardium (Figure 2). The right breast and both axillae were unremarkable.

The differential diagnosis was malignancy versus aggressive infection, therefore a contrast-enhanced staging CT scan of the chest, abdomen and pelvis was performed, which re-demonstrated an irregular left breast soft tissue mass posterosuperior to the implant extending into the adjacent intercostal spaces, abutting the pericardium (Figure 3). No osseous destruction, pulmonary nodules, or mediastinal/axillary lymphadenopathy were seen at this stage. The abdomen and pelvis were free of disease.

An US-guided core needle biopsy using a 16G automatic gun (Bard Magnum® Biopsy Systems, Tempe, USA) revealed chronic inflammatory cells and reactive changes. No malignant cells were present, so an atypical infection was suspected, the implant was surgically removed, and the patient was commenced on empiric antibiotics. The gross histological specimen measured 7.5x6.0 cm. Histopathology, cytology, and immunohistochemistry results of the removed implant and surrounding tissue showed an intact implant, with evidence of capsular fibro-inflammatory changes, silicone granulomas and CD30-positive/anaplastic lymphoma kinase (ALK)-1-negative abnormal large T cell infiltrate (Figure 4), confirming the diagnosis of BIA-ALCL. Bone marrow biopsy was negative.

A staging ¹⁸F-FDG PET/CT scan was performed showing the persistent left breast mass with intense peripheral uptake and extension into chest wall abutting the pericardium. Mild to moderate uptake in left axillary and left internal mammary lymph nodes was reported (Figure 5).

The disease was stage IIE; the patient underwent surgical resection of the mass and is currently receiving high dose chemo-radiotherapy.

**Figure 1:** Bilateral mammograms MLO views show bilateral subpectoral saline breast implants. The left implant appears irregular and collapsed with global increased density of the left breast. An ill-defined density is seen superior to the left breast implant (arrow).

**Figure 2:** Left breast sagittal T2 fat-suppressed MRI image showing peritumoral edema (arrowheads) (A) and T1 post intravenous contrast administration (8 ml Gadovist®) subtracted image (B) shows an irregular, lobulated, peripherally enhancing mass posterosuperior to the left implant with invasion of the chest wall (arrow).
Primary non-Hodgkin’s lymphoma (NHL) of the breast is very rare with an incidence of 0.5% of breast cancers and is mostly of B cell type [3]. It can arise primarily from the breast or, more commonly, secondarily involves the breast. Sporadic cases of breast lymphoma associated with breast implants have been reported in literature, with around 83 cases documented worldwide. It is frequently of T cell type, specifically anaplastic large T cell lymphoma, which accounts for 3% of NHL.

DISCUSSION

It can occur with either silicone or saline breast implants, yet, all published studies in literature have confirmed the strong link between textured-shell implants and BIA-ALCL [2]. The median age at the time of diagnosis is 52 years, and the time interval from implant placement to diagnosis of ALCL ranges from 1 to 20 years, with a mean of eight years [4].

The pathogenesis is currently under investigation, but is assumed to be an immune response induced by silicone or polyurethane capsular material, which might then trigger an exaggerated reaction and induces a monoclonal neoplasm of activated T lymphocytes [3, 5]. Other postulated mechanisms are an indirect cytokine mediated reaction, and silicone induced toxic damage [6].

The most common clinical presentation is swelling, enlargement, and pain from a delayed seroma that is formed more than a year post implant placement. Occasionally, the lymphoma can present as a mass.

No interrelation between BIA-ALCL and the reason for implant (augmentation mammoplasty versus reconstruction) has been validated. Furthermore, a history of breast cancer or lymphoma does not worsen the prognosis of BIA-ALCL [2].

The first line imaging modality is breast sonography, which differentiates between a fluid collection and a solid mass [7]. The effusion is usually confined and adjacent to the implant. The mass, if present, is usually solitary, lobular, with irregular margins. Ultrasound is also used to guide aspiration of seroma or core needle biopsy of a mass. Mammography, on the other hand, can show a circumscribed lesion adjacent to the implant with a sensitivity of 73% [7], but does not determine the nature of the abnormality. Breast MRI scan is the most sensitive imaging modality to assess the lesion, implant, axilla, and contralateral breast. A seroma may appear as peri-implant fluid collection with enhancing wall. In contrast, the mass associated with BIA-ALCL likely appears as a
solid aggressive lesion with irregular margins, and at times areas of central necrosis, with rapid enhancement and early washout of the solid component.

In the given context, the differential diagnosis includes primary breast cancer (newly diagnosed or recurrent), metastasis, atypical infection, or implant-associated mesenchymal tumor - an implant related benign, locally aggressive fibromatosis very rarely can develop into a fibrosarcoma [8].

The final diagnosis is reached by cytological, histopathological, and immunohistochemical analysis of the aspirate or tissue specimen, showing large anaplastic T-cells with abundant eosinophilic cytoplasm, pleomorphic horseshoe shaped nuclei, and prominent nucleoli with frequent mitosis. The neoplastic cells commonly stain positive to CD30 and negative to ALK-1 proteins, which bares similarity to primary cutaneous ALCL as opposed to systemic ALCL. This immunostaining is key in dictating the prognosis of the disease [1,3].

When lymphoma is diagnosed by means of imaging and histopathologic examination, an FDG PET/CT scan is performed for staging of the disease and as a baseline for follow up after treatment.

The disease largely carries a favorable prognosis when it presents with a seroma without a mass, and the treatment consists of implant removal and total capsulectomy. However, if presenting as a mass, as in our case, the prognosis is usually poor and the treatment includes surgery with adjuvant chemotherapy. Local radiation therapy is considered if the disease is persistent after surgery or the patient cannot tolerate additional surgery [2].

A comprehensive study carried out in 2014 recommended surveillance of this group of patients by postoperative clinical follow up every six months for five years, and annual breast ultrasonography for two years [2].

CONCLUSION

This report reinforces the importance of understanding the inherent complications and variable clinical presentations associated with breast implants, specifically the aggressive BIA-ALCL, to assist in early recognition and prompt management of this recently uncovered, potentially fatal disease. This is, to our knowledge, the first case of aggressive BIA-ALCL infiltrating beyond the chest wall.

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Author Contributions

Alya Binmahfouz – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Karin Steinke – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

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Conflict of Interest

Authors declare no conflict of interest.

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