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Anaplastic Large Cell Lymphoma Associated with Saline Breast Implant

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ABSTRACT

Background: Breast implant associated anaplastic large T- cell lymphoma is a rare type of non-Hodgkin lymphoma, with a reported incidence of 0.3% per 100,000 women with breast prosthesis per year. It presents most commonly as a peri-implant seroma, but may also present as a capsular mass, tumor erosion through skin, in a regional lymph node, or found incidentally during revision surgery.

Case presentation: We report a 68-year-old female patient who presented with a four month history of marked pain and swelling of the right breast, who upon implant removal and right sided capsulectomy, revealed pathology consistent with ALK negative, CD 30 positive anaplastic large T- cell lymphoma.

Conclusion: Breast implant associated anaplastic large cell lymphoma, although a rare clinical occurrence is of clinical significance. Prognosis is favorable in the majority of reported cases. Definitive treatment guidelines have yet to be determined after review of long-term follow-up data.

Introduction

Breast implant-associated anaplastic large cell lymphoma (ALCL) is a rare entity with a reported incidence of 0.3% per 100,000 women with breast prosthesis per year.[1,2] It is a rare type of non-Hodgkin lymphoma (NHL) typically found in the capsule surrounding breast implants.[3] It most often presents as a unilateral breast swelling related to late peri-implant seroma, but may also present as a capsular mass, tumor erosion through skin, in a regional lymph node, or an incidental finding during revision surgery.[4] The average age of presentation is approximately 50 years old and appears to be a late complication as symptoms can develop up to 10 years post-surgery.[4] The first cases of ALCL were thought to be associated with breast implants and were published in 1997 by Keech and Creech.[5] Since then, the growing number of cases reported has encouraged an association between the presence of implants and the development of ALCL.[6] A recent systemic review by Brody *et al* in 2014 revealed a total of 79 cases reported in literature.[7] Since then, there has been an addition of five more cases reported in published literature, totaling 84 cases to date.[1,8,9]

We present a case of a textured saline breast implant associated ALCL, adding to the current literature, and review the pathogenesis, presentation, and current recommendations for the management of this disease.

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Case Presentation

A 68-year-old female presented with a four month history of intense right sided breast pain and swelling. The patient reported a history of bilateral

subcutaneous mastectomies with subsequent placement of subpectoral saline implants greater than ten years ago. On exam, the right breast appeared swollen and markedly tender. There was no evidence of a suspicious mass, skin dimpling, or nipple discharge. On ultrasound, a large peri-implant fluid collection was seen. About 300cc of fluid was initially aspirated and sent for cytology and cultures. Cultures returned negative; however, cytology revealed scattered, single, highly atypical cells with irregular and convoluted nuclei suspicious for malignancy. Breast magnetic resonance imaging (MRI) revealed a thickened and enhanced fibrous capsule with a large right sided peri-implant intracapsular fluid collection, without evidence of implant rupture bilaterally (Figure 1). The patient underwent surgical exploration of the right breast that revealed a well formed single capsule and an intact implant. A right sided capsulectomy was performed and the implant was removed. The contralateral left implant was intact and was also removed but without a capsulectomy. The implants were not replaced. Final pathology confirmed features consistent with ALK negative, CD30+, anaplastic large T-cell lymphoma, without evidence of capsular invasion (Figure 2A and 2B). Post-operatively, the patient recovered well. The patient will maintain close follow-up with no further plans for further intervention at this time.

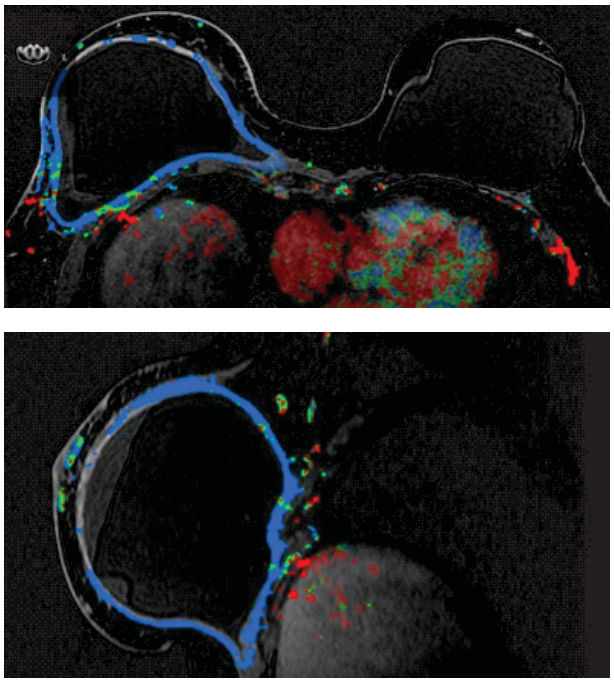

Figure 1. Breast magnetic imaging shows bilateral intact subpectoral saline implants. Large collection of intracapsular fluid surrounds a deformed implant on the right. Thickened and enhanced fibrous capsule with high intensity signal in capsule as well as surrounding skin and subcutaneous fat of the right breast. Absence of lymphadenopathy.

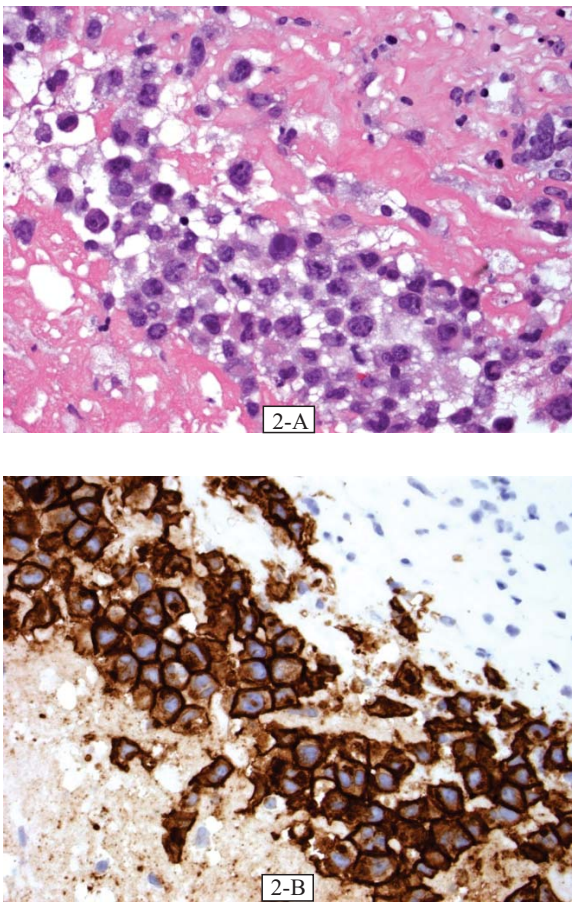

Figure 2. A) Presence of multinucelated cells, presence of "hallmark" cells with horseshoe like nuclei and abundant cytoplasm B) Anaplastic cells exhibiting immunoreactivity of CD30 and CD4 staining.

Discussion

Primary non-Hodgkin lymphoma (NHL) accounts for less than 1% of all breast malignancies. Most NHLs involving the breast are of B-cell origin. Less than 10% are of T-cell origin. ALCL is a rare T-cell lymphoma, accounting for only 3% of adult NHLs and 6% of breast NHLs.[10-13] Different types of ALCL include: systemic ALCL that is either anaplastic lymphoma kinase (ALK) positive or ALK negative, cutaneous ALCL (c-ALCL), ALCL of the breast (non-implant associated), and ALCL (implant associated).[1,3,12] Many authorities argue that implant-associated ALCL is not a disease of the breast parenchyma itself, but rather a disease of the fibrous capsule surrounding the implant as a result of chronic inflammation.[10] It has not yet been determined, however, if disease confined to the capsule will progress to a more advanced stage if left untreated.[14]

Breast implant associated ALCL is typically ALK negative, and is recognized to have a more indolent course secondary to its confinement to the peri-implant capsule, although there are reports of more aggressive presentations.[8, 9, 15, 16] It is noted to affect bilateral breasts equally, whereas primary breast lymphomas have a predilection for the right breast.[3-5, 10, 13, 14, 17] Its onset is often late and can present up to 10

years post implantation.[3,8] Although typical presentations include breast swelling (secondary to seroma or effusion), it can alternately present with constitutional B symptoms such as fever, weight loss, night sweats, general malaise, and lethargy.

It may also present as a palpable mass, or with axillary lymphadenopathy.[6, 9] Histopathologic diagnosis is typically found in microscopically neoplastic cells within the effusion or lining the fibrous capsule surrounding the implant rather than in a gross tumor.[9] Analysis of the neoplastic cells in implant associated ALCL reveals "hallmark" lymphoma cells which feature eccentric, horseshoe, or kidney-shaped nuclei with a paranucelar eosinophilic region, and positive cell surface and cytoplasmic staining for CD30 (Figure 2A and 2B).[3,8]

Definitive guidelines for the management of breast implant associated ALCL have yet to be set forth; however, a recent article published by Kim *et al* compiled recommendations from a multidisciplinary panel based on their interpretation of published evidence. There was universal agreement that seromas occurring more than 1 year after breast implantation should be aspirated and sent for culture, cytology, flow cytometry, cell block, immunohistochemical analysis, and T cell receptor gene rearrangement. With positive cytology, surgical intervention should include removal of the affected implant with total capsulectomy. This alone, is considered adequate treatment. In those with disease confined to the capsule. There is no consensus on contralateral breast implant removal or capsulectomy. All patients should also undergo workup for systemic disease with CT scan of the chest, abdomen, and pelvis, PET scan and occasionally bone marrow biopsy. The addition of chemotherapy and radiation to all patients with breast implant associated ALCL remains debatable, but in the literature, it has typically been reserved for those with a mass, systemic disease, or local, regional or metastatic disease (i.e. stage IE).[8, 14] It is usually not recommended in women with disease confined to the capsule.

Recall, this disease can also present as a mass or with axillary lymphadenopathy. Those presenting with a mass have been shown to have an overall worse survival and higher risk of treatment failure or relapse. For these patients, all suspicious tissue should be excised and sent as a fresh specimen.[8,9] In a case report of a patient exhibiting ALCL with axillary nodal involvement published by Estes *et al*, the addition of radiation therapy and chemotherapy has provided a favorable outcome to date.[9] Radiation therapy has shown some benefit in those with disease extending beyond the capsule, localized recurrent disease, persistent disease after surgery, or patients in whom surgery is not an option.[14] However, a study by Miranda *et al* showed no overall survival benefit in patients who received chemotherapy.[6] It is clear that the modes of treatment and effectiveness of each varies based on presentation; however, capsulectomy, radiation, and chemotherapy are available options for the treatment of breast associated ALCL. Continued surveillance following treatment is also paramount to long term survival. The multidisciplinary panel compiled by Kim, *et al* recommends clinical follow up at least every 6 months for 5 years and annual ultrasound for 2 years. Those with reimplantation of a breast prosthesis may require continued surveillance beyond 5 years.[14]

Breast implant associated anaplastic large cell lymphoma, although a rare clinical occurrence is of clinical significance. It is a diagnosis that can be swiftly identified and adequately treated. Patients presenting with a late forming seroma status post implant placement should undergo prompt ultrasound evaluation with fluid aspiration and cytologic analysis. As seen in the literature, prognosis is favorable in the majority of reported cases. Definitive treatment guidelines have yet to be determined after review of long term follow up data; however, the consensus is clear to perform a total capuslectomy with implant removal of the affected breast, evaluate for disseminated disease, and close follow up.

Conflict of Interest

The authors report no proprietary or commercial interest in any product mentioned or concept discussed in this article.

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