OPEN

PRS GLOBAL OPEN

CASE REPORT
Breast

A Case Report of Misdiagnosed Breast Implant-associated Anaplastic Large Cell Lymphoma with Lymphatic Extension

Dino Maglic, MD*
Whitney Moss, MD*
Giovanna Pires, BA*
Archana Agarwal, MD†
Cindy Matsen, MD‡
Alvin Kwok, MD, MPH*

Summary: Breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) is a rare peripheral T-cell lymphoma associated with textured implants and usually presents as a late-onset periprosthetic seroma. We present a 70-year-old woman with a history of left breast invasive ductal carcinoma treated with mastectomy and textured implant-based reconstruction, and subsequent adjuvant chemotherapy due to lymphovascular extension. Eleven years following her reconstruction, the patient developed a periprosthetic seroma. Fine needle aspiration and partial capsulectomy were performed, but appropriate pathologic evaluation was not initially submitted. She then presented with lymphadenopathy, which was biopsied and revealed BIA-ALCL within an axillary lymph node. Despite implant explantation, complete capsulectomy, axillary lymph node dissection, and excision of groin lymphadenopathy, no evidence of primary ALCL was appreciated. This initially misdiagnosed case demonstrates the importance of following the National Comprehensive Cancer Network guidelines when a patient presents with late onset breast periprosthetic effusions. *(Plast Reconstr Surg Glob Open 2021;9:e3916; doi: 10.1097/GOX.0000000000003916; Published online 4 November 2021.)*

Breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) is a rare T-cell lymphoma associated with textured breast implants with an incidence as high as 1:2832.[1] BIA-ALCL patients most commonly present with a late onset of breast swelling secondary to a periprosthetic seroma.[2] The National Comprehensive Cancer Network guidelines state that any symptomatic periprosthetic effusion occurring more than than 1 year after breast implantation should be aspirated and assessed for cytology and CD30 via flow cytometry to evaluate for BIA-ALCL.[3] We present a patient who initially had a missed BIA-ALCL diagnosis who was subsequently found to have lymphovascular extension of disease presenting as axillary lymphadenopathy, though pathologic examination following prosthetic explantation and capsulectomy did not reveal a primary source.

The patient is a 70-year-old woman with a history of left breast cancer diagnosed in 2007. She was initially treated with a lumpectomy and sentinel lymph node biopsy, followed by re-excision, and ultimately a mastectomy with textured implant-based reconstruction. Final pathology showed a 1.3 cm, grade 3 invasive ER+, PR+, HER-2 -, ductal carcinoma with an extensive intraductal component. Sentinel nodes were negative; however, 1 of 4 lymph nodes were found to be positive in the axillary tail of the mastectomy specimen. She received adjuvant chemotherapy without radiation.

In 2018, 11 years following her reconstruction, her left breast doubled in size over the course of 3 days. She was evaluated by a plastic surgeon, with an ultrasound showing a well-defined fluid collection around her implant. This was aspirated and discarded, and then recurred. The plastic surgeon then performed a partial capsulectomy and explant, followed by replacement with a smooth implant. The capsule was sent for pathologic evaluation without flow cytometric evaluation. Postoperatively, she did not have seroma recurrence (Fig. 1).

In May 2019, she presented to her primary care doctor with a chief complaint of a persistent cough. A workup, including a chest CT, was consistent with large and small airway disease. However, there was also a finding of an

*From the *Division of Plastic and Reconstructive Surgery, Department of Surgery, University of Utah School of Medicine, Salt Lake City, Utah; †Division of Pathology University of Utah School of Medicine, Salt Lake City, Utah; and ‡Department of Surgery, University of Utah School of Medicine, Salt Lake City, Utah.*

Received for publication August 27, 2021; accepted September 16, 2021.

DOI: 10.1097/GOX.0000000000003916

Disclosure: *The authors have no financial interest to declare in relation to the content of this article. No funding was received for this study.*

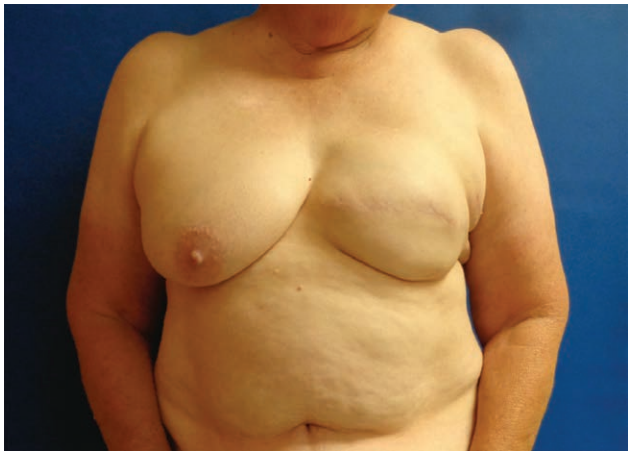

Fig. 1. Patient 11 years after original reconstruction status: post partial capsulectomy and removal of textured implant, followed by replacement with a smooth implant.

enlarged left axillary lymph node measuring 1.8 cm × 1.1 cm (Fig. 2). She reported feeling the lump in her axilla for approximately 4 years but attributed this to a scar from her previous reconstruction. She was then referred to our breast surgery service for further evaluation.

On examination she was noted to have a 3-cm mass in the left anterior axilla. An ultrasound and diagnostic mammogram showed a suspicious morphology of the lymph nodes at the site of palpable concern. A PET-CT scan was then obtained showing at least three hypermetabolic lymph nodes in the left axilla and a hypermetabolic lymph node along the course of the distal right external iliac vessels (Fig. 3).

An axillary core needle biopsy showed reactive lymphoid tissue, and flow cytometry was negative. These findings were felt to be discordant and, due to concern for lymphoma, a left axillary node excisional biopsy was performed. At the time of surgery, there was no fluid collection noted in the axilla or evidence of a periprosthetic fluid collection. Pathologic review revealed maintained

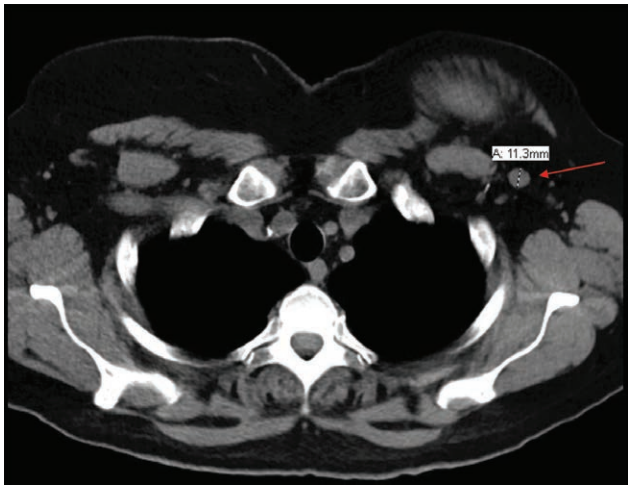

Fig. 2. CT chest scan completed in May 2019 showing an enlarged left axillary lymph node.

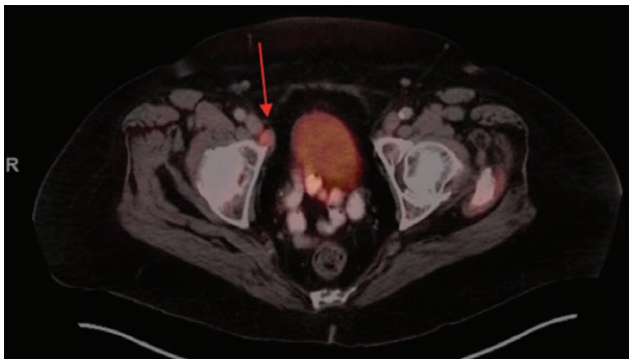

Fig. 3. PET/CT scan completed in June 2019 showing a hypermetabolic lymph node along the course of the distal right external iliac vessels.

lymph node architecture with focal sinusoidal involvement by large, atypical cells positive for CD30, CD4, TIA-1 and IRF-4 and negative for ALK-1, consistent with a diagnosis of BIA-ALCL.

The patient then underwent a bone marrow biopsy, which was negative. An additional PET-CT scan showed scattered mild uptake in adjacent subcentimeter lymph nodes in the left axilla and again demonstrated a 1.9-cm hypermetabolic node around the distal right external iliac vessels. Following a discussion with her multidisciplinary care team, the decision was made to return to the operating room to complete a left axillary dissection and excision of the right iliac node with left breast implant removal and total capsulectomy.

Intraoperatively, there were no gross capsular abnormalities and no periprosthetic fluid collection (Fig. 4). Surgical pathology of the breast capsule, axillary nodes, and the right iliac node showed benign findings. There was no evidence of lymphoma via pathology or flow cytometry.

This case report demonstrates the importance of following the National Comprehensive Cancer Network guidelines when a patient presents with late onset breast periprosthetic effusions. Although the effusion was aspirated several times and the capsule was biopsied, the proper pathologic assessment of flow cytometry, unfortunately, was not performed. It is essential to obtain fine needle aspiration of the effusion and complete cytology and flow cytometry to look for atypical CD30 positive T-cells.[3] If a mass lesion is present, it should be biopsied

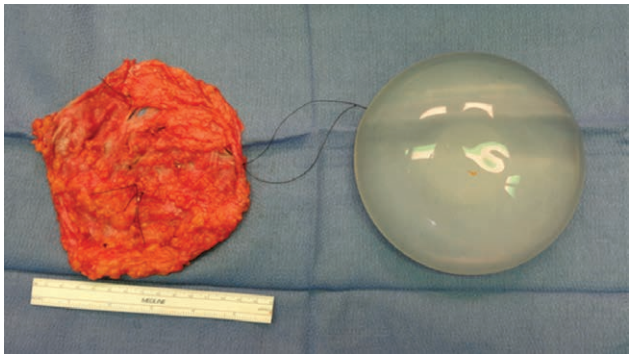

Fig. 4. Left smooth silicone breast implant upon explantation and total capsulectomy.

and evaluated for ALCL by morphology and immunohistochemistry by assessing CD30 and multiple T-cell markers. Initial workup should also include ultrasonography, which is the most sensitive of all imaging modalities.[4]

Although BIA-ALCL is mainly a localized, indolent disease that presents with seroma or mass, it can also present with axillary lymphadenopathy, skin lesions, and B-symptoms. Axillary metastasis has been described in 10%–15% of patients and can be the primary presentation.[5] The intrasinusoidal pattern of involvement, as seen in our patient, is the most common histopathologic pattern of lymph node involvement seen in more than 90% of patients.[6] Moreover, the tumor burden is also variable and ranges from 1% to 95%, emphasizing the need to obtain an excisional biopsy, as a needle core biopsy can easily miss focal and intrasinusoidal involvement.

It is unclear why her axillary lymph node was positive for ALCL but her subsequent pathology including capsule, axillary dissection, and iliac node were negative. One hypothesis is that the primary BIA-ALCL was removed during the initial workup, but that the violation of the capsule allowed for lymphovascular disease extension. Additionally, perhaps there was indolent regression of local disease after textured implant explantation. However, there have been no reports of spontaneous regression of disease following textured implant explantation. As BIA-ALCL continues to be characterized by the plastic surgery community, we hope to reinforce the existing knowledge base to provide the best treatment for patients.

Physicians who take care of patients with breast implants must have a high index of suspicion for BIA-ALCL or breast cancer recurrence and strictly follow the National Comprehensive Cancer Network guidelines if their patient presents with late onset, rapid breast swelling.

Alvin Kwok, MD, MPH
Division of Plastic Surgery
30 North 1900 East, 3B400
Salt Lake City, UT 84132

REFERENCES

1. Van Natta BW. Determining the true incidence of breast implant-associated anaplastic large cell lymphoma (BIA-ALCL): the need for accurate data. *Aesthet Surg J.* 2019;39:NP230–NP231.
2. Brody GS. Anaplastic large cell lymphoma occurring in women with breast implants: Analysis of 173 Cases. *Plast Reconstr Surg.* 2015;136:553e–554e.
3. Clemens MW, Horwitz SM. NCCN consensus guidelines for the diagnosis and management of breast implant-associated anaplastic large cell lymphoma. *Aesthet Surg J.* 2017;37:285–289.
4. Adrada BE, Miranda RN, Rauch GM, et al. Breast implant-associated anaplastic large cell lymphoma: sensitivity, specificity, and findings of imaging studies in 44 patients. *Breast Cancer Res Treat.* 2014;147:1–14.
5. Leberfinger AN, Behar BJ, Williams NC, et al. Breast implant-associated anaplastic large cell lymphoma: A systematic review. *JAMA Surg.* 2017;152:1161–1168.
6. Ferrufino-Schmidt MC, Medeiros LJ, Liu H, et al. Clinicopathologic features and prognostic impact of lymph node involvement in patients with breast implant-associated anaplastic large cell lymphoma. *Am J Surg Pathol.* 2018;42:293–305.