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CASE REPORT

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Secondary intact capsulectomy with seroma without implant: revision of an incomplete treatment of BIA-ALCL – a case report

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ABSTRACT

Background: Breast Implant Associated Anaplastic Large Cell Lymphoma (BIA-ALCL) is a haematological malignancy which may occur in patients with textured breast implant history. While typically diagnosed at an early stage with good prognosis, it may present with local residual disease due to incomplete surgical excision.

Case presentation: We describe the case of a 42 year-old woman with a history of bilateral breast augmentation for cosmetic purposes 21 years prior, who developed recurring seroma of the left side. She sought help from her first surgeon who performed 2 breast implant exchange procedures placing textured devices and finally a bilateral breast implant removal over the course of two decades. The patient did not receive capsulectomies in the previous implant exchanges, and received sampling from the anterior capsule in the last procedure, where BIA-ALCL was diagnosed on the left side. She was referred to a tertiary cancer center where preoperative workup confirmed presence of local residual disease. Following multidisciplinary team management, she underwent revision of en-bloc capsulectomy of the left side without need for additional treatments. Post-operative course was uneventful with no signs of local recurrences at 18 months follow-up.

Conclusion: Residual disease in BIA-ALCL may be caused not only by tumor characteristics or extent, but also by misdiagnosis or late diagnosis. This case highlights the critical importance of thorough surgical excision in BIA-ALCL. The existence of guidelines and clinical practice recommendations direct surgeons on how to appropriately recognize and manage symptomatic patients in order to treat suspicious cases in a timely manner.

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Introduction

Breast Implant-Associated Anaplastic Large Cell Lymphoma (BIA-ALCL) is an emerging hematologic malignancy arising from the fibrous capsule surrounding breast implants [1,2]. Since its original description in 1997 [3], BIA-ALCL has gained wide recognition, increased awareness and rising attention by the medical community, which has led to significant global changes in the way breast implants are viewed and managed [4,5]. As of 20 April 2024, the Global BIA-ALCL Network reported 1618 unique cases worldwide [6]. Its prevalence which was once considered rare has been recently estimated to range from 1 in 300 to 1 in 914 patients with textured breast implants [7–10]. Its potential for morbidity and mortality have made BIA-ALCL a focal point of ongoing research in virtually all aspects of this malignancy, from etiology to diagnosis and management [11].

The proposal of clinical pathways and the development of guidelines and recommendations have standardized the management of confirmed BIA-ALCL cases [12–14]. In fact, nearly 85% of instances are now identified at an early stage [15]. It has been clarified that complete surgical excision with en-bloc capsulectomy and contralateral total intact capsulectomy is the most effective approach for improving disease-free survival [16,17]. However, some patients might present residual disease caused by incomplete surgical

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resection. Residual disease might be due to tumor extension, anatomical constraints, or other surgical limitations in patients with advanced disease at time of diagnosis [18]. However, there are instances where residual disease is caused by misdiagnosis or suboptimal management with failure to identify BIA-ALCL before undergoing primary surgical treatment. The implications of incompletely resected cases are grave as they include potentially worse clinical outcomes, with risk of recurrence, disease progression, and the possible need for additional adjuvant treatments following surgery [19].

Despite the relevance of this issue, the literature addressing the management of incompletely resected BIA-ALCL remains limited. We present the case of a symptomatic patient inadequately managed with primary surgery, who developed local residual disease requiring reoperation.

Case presentation

A 42 year-old patient was referred to our tertiary cancer center with a history of recurring seroma of the left breast. The patient had undergone bilateral breast implant removal with partial capsulectomy 11 months prior to her first visit at our institution. The patient presented with no notable comorbidities, and had a history of bilateral breast augmentation in the submuscular plane with cosmetic indication at age 21 in the year 2003. No information could be found regarding the type of implants she had originally received. The patient returned to the same surgeon in 2008 complaining of bilateral seroma, to undergo a first bilateral breast implant replacement with textured anatomical implants of 425 cc (Sebbin LSA TF 425). In 2013, she underwent a second implant replacement procedure to increase the size with 480 cc implants from the same manufacturer (Sebbin LSA TF 480). In 2017, the patient returned for a check-up where she complained of pain and breast volume increase bilaterally. At the time, she had received an ultrasound scan which identified periprosthetic effusions bilaterally, with no lymphadenopathy or palpable masses in the breast regions. The effusions were drained without analysis, but recurred over the years, when the patient sought help from the same surgeon and underwent a third revisional procedure in 2022, with a bilateral breast implant removal and sampling of the anterior periprosthetic capsule. The surgeon sent two specimens from the right side (9×1 cm and 3×3 cm) and one from the left (2×1 cm). Histopathological report of the specimen found 'Acellular amorphic proteinaceous material' on the right side, and 'Sclero-hyaline fibrous tissue with lymphocyte-monocyte inflammation, requiring correlation with the clinical presentation' on the left side. Despite the explantation, seroma still recurred on the left side, and was sent for cytopathological testing which identified 'a population of atypical pleomorphic cells with irregular contours suggestive of a lymphoproliferative process with CD30+ elements' for which further diagnostic testing was advised. For this reason, the patient sought treatment at our institution where the case was managed by a multidisciplinary team (MDT) featuring a plastic surgeon, an hematopathologist, a hematologist, an oncologist, a radiotherapist, a surgical oncologist, and a breast imaging radiologist Figure 1. The original histological specimen from the capsule sampling was reviewed by a hematopathologist from our referral center, who identified occasional sparse large atypical CD30+ cells that were deemed compatible with BIA-ALCL diagnosis. Anaplastic cells presented the following phenotype: CD30+, CD3-, CD7-, CD5-, CD4-, CD8-, Granzyme B+/-, TIA1-, ALK1-, PAX5-, CD79a-, CD68-, CD15-/+. The patient received an ultrasound scan with fine-needle aspiration cytology of the recurring effusion which confirmed an atypical CD30+ lymphoproliferative process. Total body scanning with positron emission

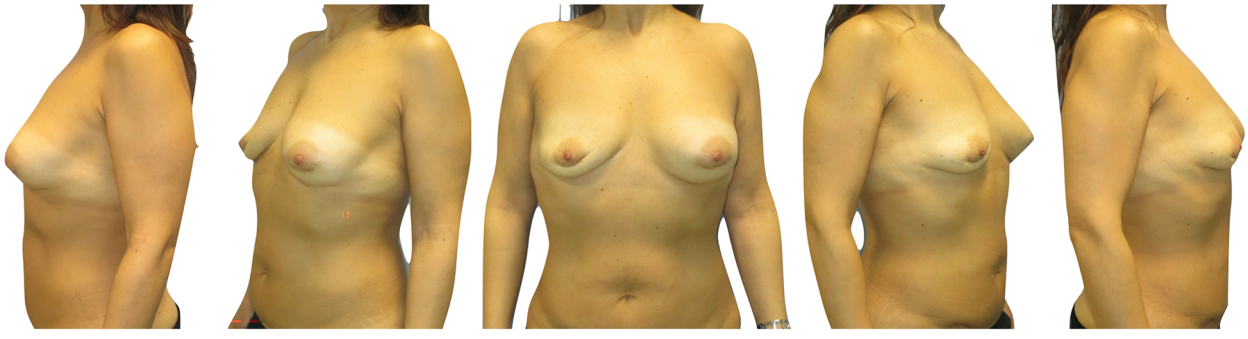

Figure 1. Clinical presentation of the BIA-ALCL patient when referred to our institution, 11 months after bilateral breast implant removal, with recurring left breast swelling, shown in frontal, lateral and oblique views.

tomography/computerized tomography (PET/CT) was performed in a different facility in closer proximity to the patient's domicile, revealing a 67 × 20 mm area of seroma in the left breast region with focal ^{18}F-FDG (Fluorodeoxyglucose) uptake (SUV max 1.8). No other areas of uptake could be located in all other explored body segments. The patient also received magnetic resonance imaging (MRI) of both breasts with contrast medium to complete preoperative staging, confirming the seroma of the left side. Slides of the specimen from the previous surgery were reviewed by the hematopathologist who deemed the findings sufficient for identifying BIA-ALCL with early infiltration of the periprosthetic capsule (pT2 according to the proposed MD Anderson-TNM staging system) [20]. Three weeks after the initial visit, the patient underwent radicalization surgery with en-bloc capsulectomy containing only 150 cc of serous effusion without implant Figure 2. The liquid contents were sent for culture and cytopathological testing while the solid specimen was sent for histopathological examination. Additionally, the patient underwent sentinel lymph node biopsy of 2 axillary nodes identified preoperatively through segmental lymphoscintigraphy. Lymph nodes presented chronic reactive silicone-associated lymphadenopathy with no signs of lymph node metastasis. Histology report of the capsule revealed cell aggregates infiltrating the periprosthetic capsule, but clear resection margins and no signs of infiltration beyond the capsule. As such, the patient received a stage IC (pT3pN0M0).

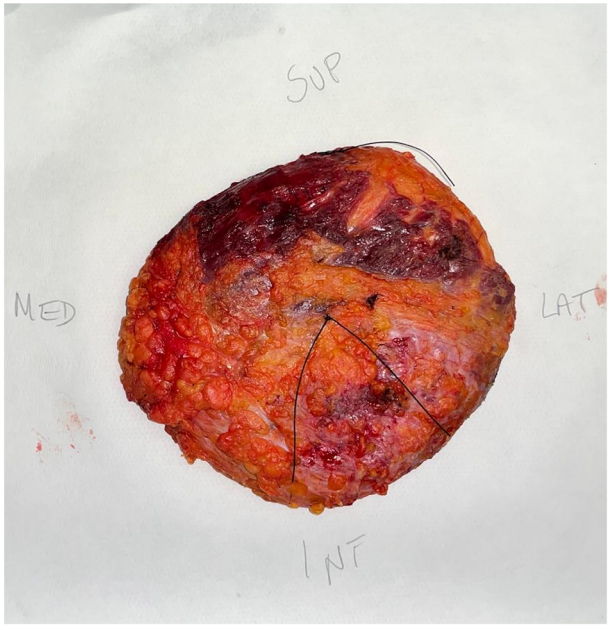

Figure 2. Surgical specimen of the intact secondary capsulectomy with seroma without implant, sent to pathology with landmark sutures.

Post-operative course was uneventful, and no further adjuvant treatments were deemed necessary by the MDT. She continued her stringent monitoring protocol, with post-op PET/CT at 6, 12 and 18 months from surgery showing no signs of local or distant disease recurrence. As it stands, the patient is still continuing her follow-up visits and scans, with a disease free survival beyond 18 months.

Discussion

The patient was diagnosed with BIA-ALCL never having undergone adequate surgical treatment: she had implant removal with partial capsule sampling which was supposedly not diagnostic for BIA-ALCL in the center where the histological sample was first analyzed. The reassessment of the slides in a specialized referral center demonstrated that the samples were diagnostic for the malignancy. In fact seroma still built up despite the implant removal. The combination of these elements suggest that our patient had a residual/inadequately managed disease. Findings from literature corroborate the fact that residual disease in BIA-ALCL patients may sometimes be caused by misdiagnosis or diagnostic delayed owed to inappropriate management, similarly to what was presented in our case. In fact, we are aware of one other description of local residual disease where one symptomatic patient had recurrent seromas 2 years after replacement of a textured implant with a smooth one, with incomplete removal of the capsule. Diagnosis was obtained after the latter surgery, although it was reported that the patient was successfully treated with re-explantation and complete capsulectomy [21]. To avoid similar cases, it is of pivotal importance to recognize the signs and symptoms, as the management of symptomatic cases has been made abundantly clear in national and international recommendations and guidelines [11,12]. Suspicious cases with a clinical presentation that is compatible with the malignancy such as recurrent seromas should receive complete diagnostic testing including morphology, CD30 immunohistochemistry and flow cytometry. Sending samples of the effusion for analysis in suspicious cases should be of paramount importance to exclude malignancies before any surgical intervention [22]. Additionally, surgical specimens from confirmed cases should be submitted possibly fresh and intact to the pathologist only containing landmark sutures, and should be evaluated within 24 h onto 12 strategic regional biopsies in a standardized approach [23]. Cases of equivocal pathology should be managed by seeking secondary review in a tertiary referral center for BIA-ALCL [24]. Nevertheless, it should

be acknowledged how there are contrasting opinions regarding the use of diagnostic imaging for follow-up in patients with implants. In fact, we acknowledge the UK guidelines according to which routine radiological surveillance to assess implant health is not recommended, advocating for only symptom-driven assessments [25]. Yet, we advocate in favor of erring on the side of caution, providing recommendations similar to those from the US Food and Drug Administration's breast implant screening recommendations. The latter state that patients should have a first imaging test at 5-6 years after initial implantation and then every 2-3 years thereafter, ultrasound or MRI, even if they are asymptomatic [26].

It should be addressed how local residual BIA-ALCL may in other instances be a consequence of an advanced malignancy (i.e. tumor extension beyond the periprosthetic capsule), despite timely surgery and a seemingly appropriate treatment strategy in accordance with guidelines and recommendations [27]. Indeed, surgical control of excision margins should always be sought whenever possible as the mainstay of treatment, since surgical management with complete surgical excision is currently the only demonstrated way to achieve optimal event-free survival [17]. However, clear surgical margins may not always be achieved due to chest wall invasion, which has sometimes required invasive chest wall resections and reconstructions that were associated with relevant morbidity [28,29]. Efforts should be made to achieve oncological radicality, due to its importance for improving disease-free survival [14]. However, this may not always be possible, either because of tumor extension, anatomical constraints, or even patient frailty. In fact, we previously reported a case of focal invasion of the posterior chest wall in a frail patient which could not undergo reoperation but who was successfully managed with adjuvant radiotherapy [15]. According to the National Comprehensive Cancer Network (NCCN)'s recommendations on BIA-ALCL, local residual disease due to incomplete surgical excision should be discussed with a multidisciplinary team, and should take into consideration adjuvant treatment options, or even systemic therapy should radiotherapy not be feasible [21,28]. However, effectiveness of adjuvant strategies in advanced BIA-ALCL cases is currently based on few anecdotal reports and small retrospective studies [16].

Finally, this case report is an example of how human factors may come into play and lead to diagnostic delay of BIA-ALCL. These factors include not recognizing a clinical presentation that was suggestive of BIA-ALCL, not abiding by national or international guidelines by performing ultrasound-guided fine needle aspiration of the effusion, taking the patient to surgery before having reached a conclusive diagnosis, and not having referred the patient to a referral center in case of dubious diagnosis. The patient's implant history spans over a period of approximately two decades. We are unable to identify the exact moment when BIA-ALCL may have been developed as there was no defining trait that distinguished the last episode of late seroma from the first ones. What made the difference was the use of diagnostic tools which had not been used in previous times causing a diagnostic delay. Hopefully, this will serve as a cautionary tale, improving awareness of BIA-ALCL and encouraging practitioners to use current guidelines for the appropriate management of this malignancy.

Conclusion

Misdiagnosing BIA-ALCL or delaying its identification with suboptimal treatment have been associated with residual disease which may cause worst prognosis and progression into advanced disease. Management by a multidisciplinary team is fundamental in managing any suspicious or confirmed case of BIA-ALCL. Whenever possible, reoperation should be attempted to improve disease-free survival. Adjuvant treatment modalities should be discussed in the multidisciplinary setting according to disease and patient characteristics. The best strategy to prevent misdiagnosis of diagnostic delay from happening is to continue improving awareness of the disease, encouraging the recognition of the clinical presentation as well as the implementation of current guidelines for its management. Finally, any dubious case should be referred to referral centers specialized in the treatment of such patients, as secondary salvage procedures may expectedly be more complex that primary ones, threatening patient survival in terms of morbidity as well as mortality.

Conflict of interest disclosure Statement

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