Case Report

Implant-associated anaplastic large T-cell lymphoma (BIA-ALCL): case report

Linfoma anaplásico de grandes células T associado ao uso de implantes (BIA-ALCL): relato de caso

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■ **ABSTRACT**

Recognized by the World Health Organization in 2016, breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) is an uncommon subtype of T-cell non-Hodgkin lymphoma that develops after the insertion of breast implants. The disease is a rare condition that affects approximately one in every 30,000 people with textured breast implants. The main clinical manifestations are late seroma, breast asymmetry, mass, and capsular contracture, with a higher frequency of the former. Explantation of the prosthesis with total capsulectomy may be sufficient to treat ALCL, with resections extended to adjacent sites when necessary. However, in some cases, adjuvant radiotherapy and/or chemotherapy is performed. It is concluded that, for an early diagnosis and effective treatment, women with sudden and late-onset seroma should undergo additional tests to exclude this condition, even with a shorter development time than the average, which is around 10.6 years.

Keywords: Lymphoma, large-cell, anaplastic; Positron-emission tomography; Lymphoma, non-Hodgkin; Seroma; Breast implantation.

■ **RESUMO**

Reconhecido pela Organização Mundial de Saúde em 2016, o linfoma anaplásico de grandes células associado ao implante mamário (BIA-ALCL) é um subtipo incomum de linfoma não Hodgkin de células T, que se desenvolve após a inserção de próteses mamárias. A doença é uma afecção rara que afeta cerca de uma a cada 30.000 pessoas com implante mamário texturizado. As principais manifestações clínicas são o seroma tardio, assimetria mamária, massa e contratura capsular, com frequência mais elevada do primeiro. O explante da prótese com capsulectomia total pode ser suficiente para tratar o ALCL, com ressecções estendidas a locais adjacentes, quando necessário. Entretanto, em alguns casos, é realizada a radioterapia e/ou quimioterapia adjuvante. Conclui-se que, para um diagnóstico precoce e um tratamento efetivo, mulheres com seroma de aparecimento súbito e tardio deverão realizar exames complementares para a exclusão dessa afecção, mesmo com tempo inferior à média de desenvolvimento, que é de cerca de 10,6 anos.

Descritores: Linfoma anaplásico de células grandes; Tomografia por emissão de pósitrons; Linfoma não Hodgkin; Seroma; Implante mamário.

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INTRODUCTION

In 1962, Thomas Cronin and Frank Gerow performed the first surgery to insert a silicone breast prosthesis. Since then, with progressive growth, more than 1.5 million women undergo breast augmentation annually, whether for aesthetic reasons or reconstruction[1]. In Brazil, this procedure has seen a gradual increase over the last 25 years and, in 2011 alone, around 145 thousand Brazilian women underwent this surgery[2,3]. As a result,

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Brazil became the second largest market for breast implants in the world, behind the United States[2-4].

In 1997, Keech and Creech described the first case of breast implant-associated anaplastic large cell lymphoma (BIA-ALCL)[5-9]. However, it was recognized by the World Health Organization (WHO) as an oncological entity only in 2016[5,7-9]. The incidence and prevalence of BIA-ALCL is extremely low, and in 2019, the US Food and Drug Administration (FDA) reported 573 cases in the United States and worldwide, with 33 deaths resulting from this lymphoma[6,10].

The pathogenesis of BIA-ALCL is not fully understood, but it is described as a process that involves multiple factors[5,6,11]. One of the theories found was that textured prostheses have concavities, which results in a wider surface with greater texture. This condition increases the likelihood of causing a chronic inflammatory response, favoring the development of biofilm[1,5,8,11].

As a result of biofilm formation, a growth of gram-negative bacteria, Ralstonia spp, was observed in the microbiome of the capsule of prostheses of patients diagnosed with BIA-ALCL[5,12]. Furthermore, the progression to lymphoma occurs as a result of the malignant transformation of the T cells involved, associated with the period of disease progression, the activation of the immune response, and the patient's genetics[5].

BIA-ALCL constitutes a medical challenge that requires greater understanding and attention, as the use of breast implants is growing exponentially throughout the world, including in Brazil. And, consequently, the probability of new cases tends to increase[1]. In this article, we describe a case of a patient diagnosed with BIA-ALCL five years after breast implant implementation.

CASE REPORT

Patient MSS, 54 years old, female, Caucasian, attends a consultation at a private clinic complaining of post-pregnancy abdominal sagging and small breasts. A patient with no previous history of cancer in the family. Previously performed post-trauma splenectomy, epigastric herniorrhaphy, varicose veins, and two cesarean sections. He has osteoarthritis in his fingers and uses simvastatin and enalapril (Figure 1).

On 12/02/2015, she underwent anchor abdominoplasty and breast augmentation, using high-profile, textured silicone implants: 275cc on the right and 315cc on the left (Figure 2). There were no complications during or after surgery. Postoperative follow-up was carried out until 2019 when he was discharged as an outpatient.

On 10/22/2020, she complained of progressive enlargement of her right breast associated with local pain, with approximately one month of evolution. On physical examination, there were no characteristic signs of capsular contracture (Figure 3). Drainage of 200ml of citrine yellow liquid was then performed, guided by ultrasound, with immediate improvement in symptoms;

The material was sent for oncotic cytology. In the first laboratory, the cytology was negative. Due to the senior author's high suspicion of malignancy, the sample was sent to a new laboratory. This time, positive oncotic cytology and negative culture were found. The patient was immediately referred to the oncologist.

A PET-SCAN was performed, which showed areas of high uptake in the upper and lower pole of the right breast, without lymph node involvement or distant metastases (Figure 4).

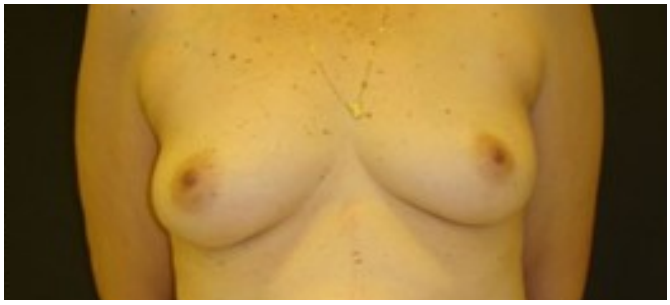

Figure 1. Pre-operative.

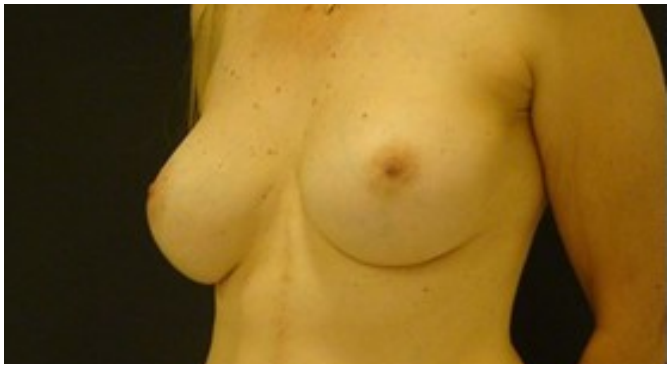

Figure 2. Patient in 2019, with final result of surgery.

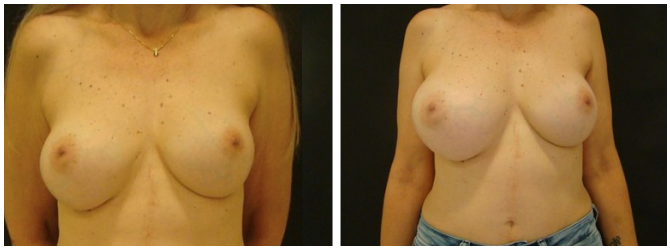

Figure 3. On the left, patient being discharged from the outpatient clinic (2019). On the right, in 2020, patient with right breast enlargement.

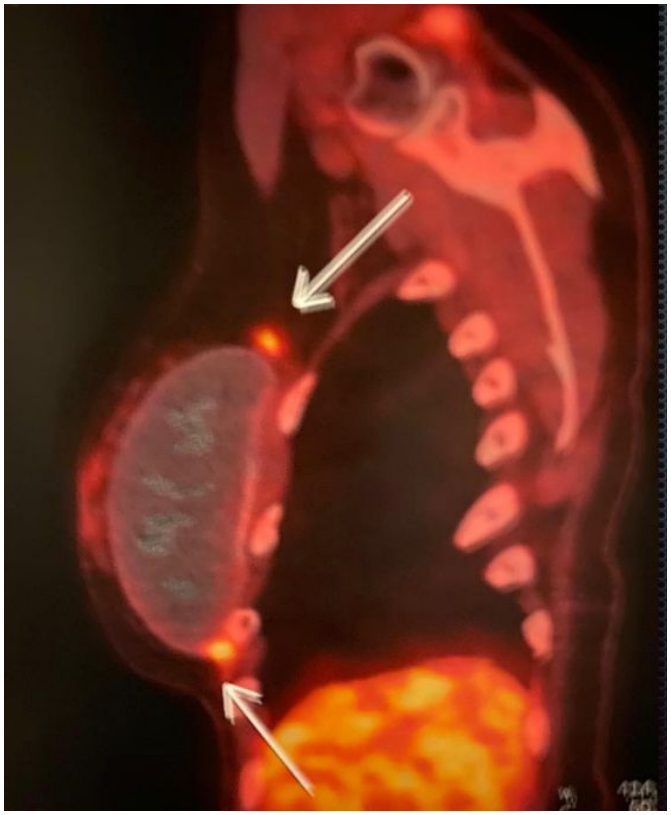

Figure 4. PET SCAN.

The patient agreed to participate in the study and signed the Informed Consent Form.

In December 2020, mastopexy was performed with explantation of the prostheses, as well as extended resection of fat and adjacent muscle (Figure 5). During the surgical procedure, a usual periprosthetic pseudocapsule was visualized in the right breast, with no signs of malignancy (Figure 6).

The anatomopathological study confirmed a pseudocapsule infiltrated by bulky anaplastic lymphoid cells, without extracapsular invasion. Immunohistochemistry showed positive CD30, CD4, CD3, and TIA-1 markers and negative ALK-1 and CD20 markers. Therefore, it was confirmed that it was anaplastic large T-cell lymphoma associated with a right breast implant, restricted to the pseudocapsule.

Three months after the mastopexy with breast prosthesis explantation, a follow-up PET-SCAN was performed, which did not reveal any further areas of hypermetabolism in the patient's right breast. Thus, the patient was free from localized or distant disease. The patient reported that after the operation she had pain in her breasts and bruises in the lower back, but that after a certain time, they went away. Her breasts were different sizes and the post-surgical scars bothered her, and despite this affecting her self-esteem, she was

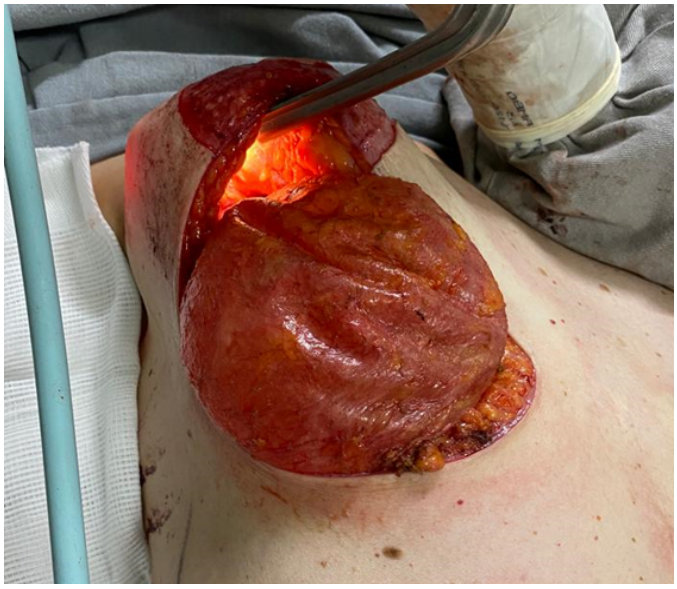

Figure 5. Prosthesis explantation - intraoperative.

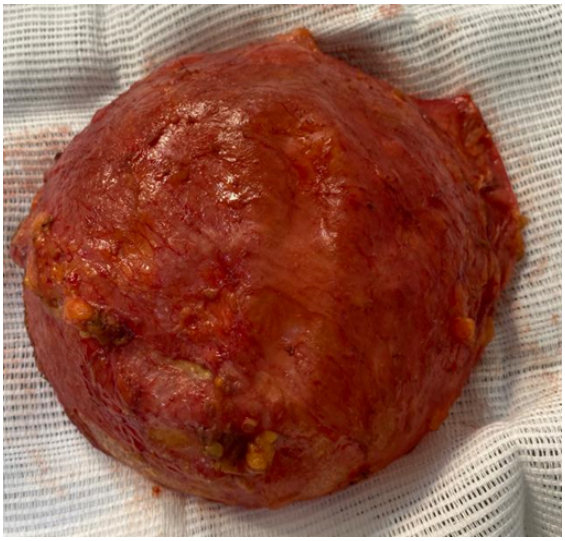

Figure 6. Breast prosthesis with pseudocapsule.

happy with the medical treatment and medical attention throughout the process (Figures 7 to 10).

DISCUSSION

Breast implant-associated anaplastic large cell lymphoma - BIA-ALCL is a rare subtype of T-cell non-Hodgkin lymphoma, characterized as CD30 positive and ALK negative[5].

The disease is a rare condition that affects approximately one person in every 30,000 with breast implants. Polyurethane and textured implants are more associated with the disease due to their greater surface area, which causes a more intense chronic inflammatory

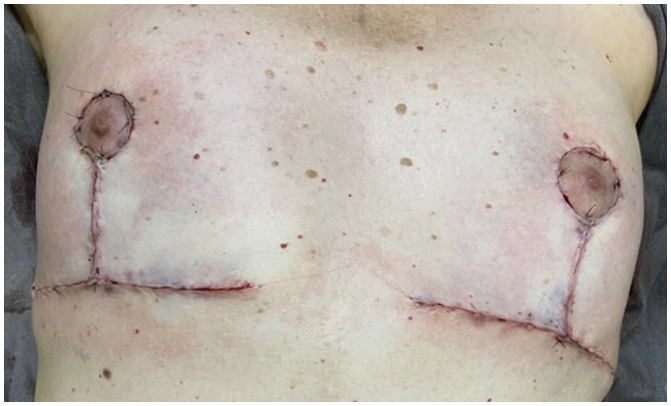

Figure 7. Image demonstrating the immediate postoperative period.

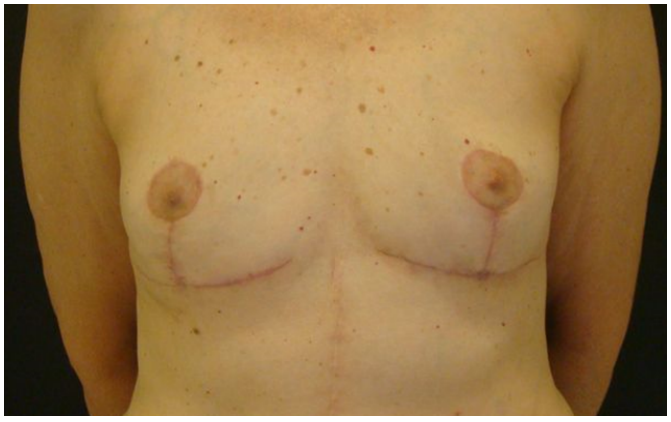

Figure 8. Image demonstrating healing 3 months postoperatively.

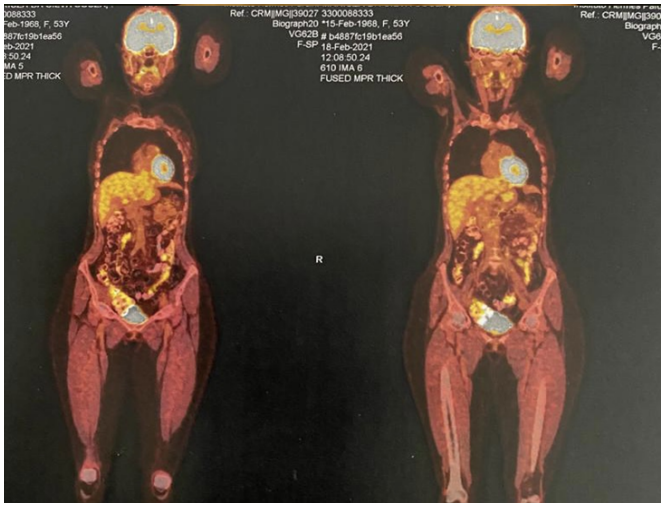

Figure 9. PET-SCAN exam results.

reaction with immune activation mediated by Th1 and Th17[1] lymphocytes.

The main clinical manifestations of the disease are late seroma, breast asymmetry, mass, and capsular contracture, with a higher frequency of the former[1]. In

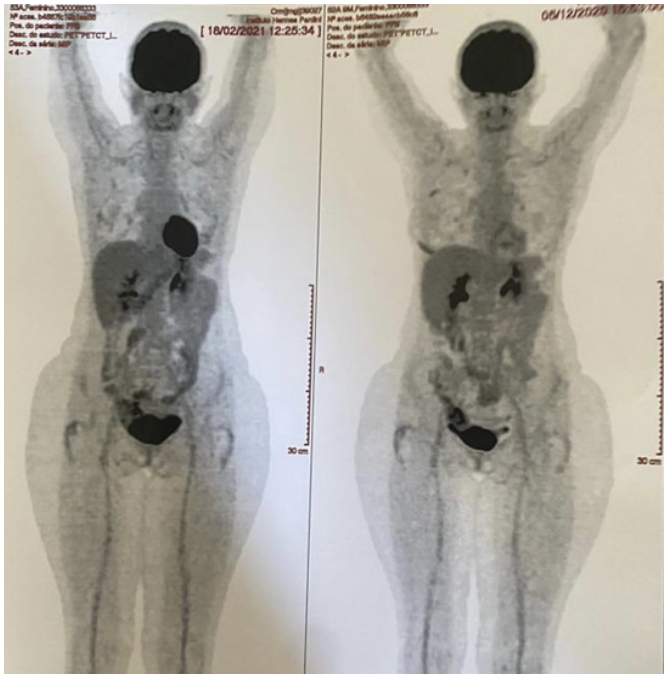

Figure 10. PET-SCAN exam results.

the case reported, the presence of a late and sudden seroma was the first change found.

Explantation of the prosthesis with total capsulectomy may be sufficient to treat BIA-ALCL, with resections extended to adjacent sites when necessary. However, in some cases, adjuvant radiotherapy and/or chemotherapy is performed, for example, if there is regional or distant metastasis[1].

In this case, it was demonstrated that, for effective treatment, women with sudden and late-onset seroma should undergo complementary tests for the earliest possible diagnosis of this condition, even with a shorter development time than the average, of around 10.6 years, as in this case, in which the disease appeared within 5 years[5]. Among the complementary tests that can be useful in the prognosis of the condition, we can highlight the PET-SCAN, which highlights areas of hypermetabolism, corresponding to cancer cells.

The importance of knowing the condition and having it in your range of diagnostic hypotheses is necessary, even if the majority of late-appearing seromas are benign. High suspicion as in the case portrayed, as it is a watershed in early and late treatment, is essential for intervention at the right time and with high cure rates.

CONCLUSION

Anaplastic large T-cell lymphoma is associated with breast implants and, despite being a rare disease, should be suspected in post-breast augmentation patients

who present associated characteristic symptoms. It follows, therefore, that, for an early diagnosis and effective treatment, women with a seroma that appears suddenly and late must undergo additional tests to exclude this condition, even with a shorter than average development time.

COLLABORATIONS

APPN Analysis and/or data interpretation, Methodology, Supervision, Writing - Original Draft Preparation, Writing - Review & Editing.

ALMSM Analysis and/or data interpretation, Methodology, Supervision, Writing - Original Draft Preparation, Writing - Review & Editing.

MJFD Investigation, Methodology, Writing - Original Draft Preparation, Writing - Review & Editing.

MJCF Investigation, Methodology, Writing - Original Draft Preparation, Writing - Review & Editing.

BRSS Investigation, Methodology, Writing - Original Draft Preparation, Writing - Review & Editing.

MVMM Analysis and/or data interpretation, Methodology, Supervision, Writing - Original Draft Preparation, Writing - Review & Editing.

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