

Breast Implant-Associated Anaplastic Large Cell Lymphoma: A Case Report

Linfoma anaplásico de células grandes associado a implante mamário: Um relato de caso

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Abstract

Keywords

- ▶ allografts
- ▶ plastic surgery
- ▶ anaplastic large-cell lymphoma
- ▶ breast implants
- ▶ breast

Resumo

Palavras-chave

- ▶ aloenxertos
- ▶ cirurgia plástica
- ▶ implante mamário
- ▶ linfoma anaplásico de células grandes
- ▶ mama

Although breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) is a disorder still sparsely described in the literature, the number of reported cases is rising rapidly. Few of the cases published to date involve Brazilian patients. We present a case treated at Instituto Nacional de Câncer (INCA) with an emphasis on the plastic surgery follow-up, which included serial fat-grafting procedures to restore breast volume without new implants.

The prognosis of BIA-ALCL is often favorable, provided its diagnosis and treatment are accomplished early. The present report aims to contribute to the recognition and management of this condition in plastic surgery practices.

O linfoma anaplásico de células grandes associado a implante mamário (BIA-ALCL, do inglês *breast implant-associated anaplastic large cell lymphoma*) é um distúrbio ainda pouco descrito na literatura; porém, o número de casos está crescendo rapidamente. Dos casos que foram publicados até agora, poucos são de pacientes brasileiras. Apresentamos um caso tratado no Instituto Nacional de Câncer (INCA) com ênfase no seguimento realizado pela cirurgia plástica, que incluiu lipoenxertias seriadas para restauração do volume mamário sem uso de novos implantes. O prognóstico do BIA-ALCL associado a implantes mamários é geralmente favorável, desde que diagnosticado e tratado precocemente. Este relato visa contribuir para o reconhecimento e abordagem dessa doença na prática da cirurgia plástica.

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Introduction

Breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) is a T-cell lymphoma. The first case report dates from 1997.¹ As of June 30, 2024, a total of 1,380 cases have been described worldwide.² The epidemiology of BIA-ALCL is limited by the lack of data on the prevalence of implant users, type of implants, and associated adverse events, challenging estimates of incidence and risk. To date, virtually all cases described are related to textured implants, with a mean time to onset of 8 to 9 years after implantation. Diagnosis relies on clinical features, including delayed effusion, mass, swelling, pain, rash, or cutaneous ulcer. Diagnostic confirmation requires cytology and seroma immunophenotyping and/or immunohistochemistry (IHC).³

Objective

The present article aims to report a clinical case of BIA-ALCL, highlighting the diagnostic process, surgical treatment (explantation and capsulectomy), and oncological follow-up. In addition, we demonstrate the reconstructive strategy adopted (breast fat grafting) and emphasize the significance of early recognition of this condition for a better prognosis.

Materials and Methods

The present descriptive observational study is a case report from Instituto Nacional de Câncer (INCA), Rio de Janeiro, RJ, Brazil. Information collection used the following:

- Review of the patient's medical records, including clinical and surgical data and supplementary exams (imaging and histopathology).
- Direct observation of clinical evolution in the pre- and postoperative periods.
- Bibliographic review in databases such as Public Medical Database (PubMed), Scientific Electronic Library Online (SciELO), and Google Scholar, using descriptors in

Portuguese and English (*BIA-ALCL, breast implants, and anaplastic large cell lymphoma*).

- Informed Consent Form for image use and clinical information dissemination with no patient identification.

The institution's Ethics Committee approved this study under opinion number 7.515.140.

We did not use any sampling method as this is a single case described in depth to illustrate the clinical presentation, treatment, and oncological follow-up.

Results

Case Report

A 45-year-old HIV-positive patient was undergoing antiretroviral therapy with dolutegravir, darunavir, and ritonavir, and had no other comorbidity. She had history of breast augmentation in 2014 and reported a sudden, painless increase in the size of the right breast starting in August 2021. A fine-needle aspiration biopsy of seroma in the right breast revealed the presence of markedly atypical cells of lymphoid lineage and was positive for malignancy.

After discussing the case with the mastology team in March 2022, we decided on bilateral explantation with capsulectomy for histopathological diagnosis.

In March 2022, the physical examination revealed a right breast slightly larger than the left, breast implants without contracture, absence of palpable nodules, and a fibroelastic lymph node in the right armpit (►Fig. 1).

Magnetic resonance imaging showed an extensive intracapsular collection in the right breast with heterogeneous content, associated with enhancement and thickening of the fibrous envelope and capsule, consistent with an inflammatory process and capsular contracture. It was not possible to rule out BIA-ALCL. The Breast Imaging-Reporting and Data System (BIRADS) was 4 (►Fig. 2).

On May 2, 2022, a surgical procedure removed the implants (textured device) and involved total capsulectomy.

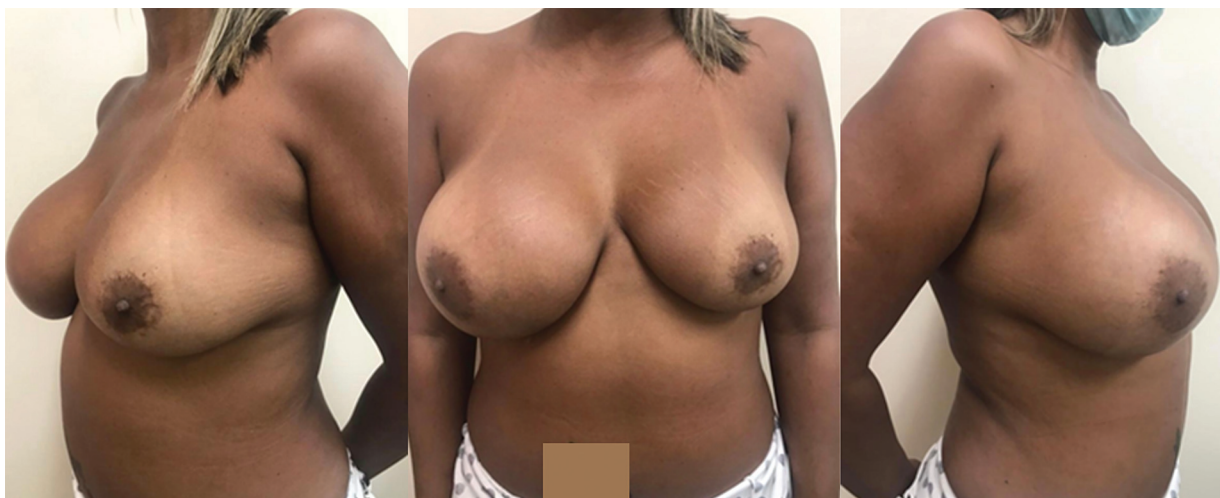


Fig. 1 Preoperative aspect of the patient.



Fig. 2 Breast magnetic resonance imaging of the patient from August 2021 showing single-lumen, retroglandular silicone implants with anterior closure inserts (potentially representing rotation) and no signs of intra- or extracapsular rupture. Large, heterogeneous intracapsular collection on the right side, associated with enhancement of the fibrous capsule. Small amount of pericapsular fluid on the left side, associated with delayed capsular enhancement.

Evolution in the immediate postoperative period was adequate (► **Fig. 3**).

The histopathological study report of the specimen (► **Figs. 4–5**) read as follows: “Anaplastic large cell lymphoma associated with breast implant. The cells line the capsule. Absence of capsular infiltration or solid mass formation.

Associated giant-histiocytic foreign body reaction. Immunohistochemistry was positive for CD30, CD4, CD3, CD2, and granzyme B and negative for ALK, CD20, and CD8.”

The patient had a satisfactory postoperative evolution and remains under follow-up by the INCA oncology and hematology teams. Her monitoring includes blood and imaging



Fig. 3 Immediate postoperative period of bilateral explantation associated with capsulectomy in May 2022.

Res. Clínico :	Paciente com diagnóstico citopatológico de neoplasia linfoproliferativa de mama direita.
MATERIAIS :	A Fragmento de cápsula de mama direita B Cápsula de prótese mamária à direita C Cápsula de prótese mamária à esquerda.
Macroscopia A :	Fragmento pardo claro e friável medindo 0,6 x 0,3 cm.
Conclusão A :	Raras células atípicas em meio a material amorfo eosinofílico.
Concluído por :	Dr. (a) LUCIANA WERNERSBACH PINTO - CRM: 52.60735-3 em 10/05/2022 Dr. (a) FRANCISCO SALES DE ALBUQUERQUE FILHO - CRM: 12.22864- em 10/05/2022 Dr. (a) TATIANA FONSECA ALVARENGA - CRM: 52.86213-4 em 10/05/2022 Dr. (a) YASMIN DE MACEDO MALLON COUTO - CRM: 52.10127-4 em 10/05/2022
Texto conferido e assinado eletronicamente por :	Dr. (a) LUCIANA WERNERSBACH PINTO - CRM: 52.60735-3 em 10/05/2022
Macroscopia B :	Produto de retirada de cápsula mamária direita com prótese, medindo 120 x 110 mm, marcado com fios, exibindo cápsula parda, irregular e espessa. Aberta, presença de tecido pardo claro e friável aderido à prótese mamária e à cápsula.
Conclusão B :	Linfoma de grandes células anaplásicas associado à implante mamário. As células neoplásicas revestem a cápsula. Ausência de infiltração capsular ou formação de massa sólida. Reação gigante-histiocitária do tipo corpo estranho associada. Imuno-histoquímica: Positividade para CD30, CD4, CD3, CD2, granzima B. Negatividade para ALK, CD20, CD8.
Concluído por :	Dr. (a) LUCIANA WERNERSBACH PINTO - CRM: 52.60735-3 em 10/05/2022 Dr. (a) FRANCISCO SALES DE ALBUQUERQUE FILHO - CRM: 12.22864- em 10/05/2022 Dr. (a) TATIANA FONSECA ALVARENGA - CRM: 52.86213-4 em 10/05/2022 Dr. (a) YASMIN DE MACEDO MALLON COUTO - CRM: 52.10127-4 em 10/05/2022
Texto conferido e assinado eletronicamente por :	Dr. (a) LUCIANA WERNERSBACH PINTO - CRM: 52.60735-3 em 10/05/2022
Macroscopia C :	Prótese mamária medindo 140 x 85 x 75 mm e envolta por cápsula de tecido pardo de superfície interna lisa, de paredes finas e opalescentes. Escasso tecido adiposo aderido na periferia.
Conclusão C :	Tecido mamário exibindo fibrose, infiltrado inflamatório linfoplasmocitário com células gigantes tipo corpo estranho, vasos neoformados e sinovialisacão na superfície. Presença de alterações de células colunares sem atípicas.
Concluído por :	Dr. (a) TATIANA FONSECA ALVARENGA - CRM: 52.86213-4 em 10/05/2022 Dr. (a) YASMIN DE MACEDO MALLON COUTO - CRM: 52.10127-4 em 10/05/2022
Texto conferido e assinado eletronicamente por :	Dr. (a) TATIANA FONSECA ALVARENGA - CRM: 52.86213-4 em 10/05/2022

Fig. 4 Histopathological and immunohistochemical report of the case. In red, CD30 positivity and anaplastic lymphoma kinase (ALK) negativity are evident, confirming the diagnosis of breast implant-related anaplastic lymphoma.

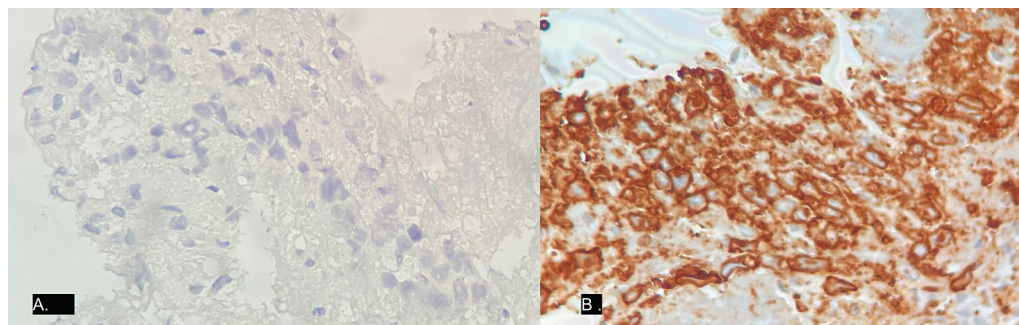


Fig. 5 (A). Negativity for ALK in neoplastic cells. (B). Strong and diffuse expression of CD30 in neoplastic cells.

tests (positron emission tomography-computed tomography [PET-CT]), showing no evidence of active disease or need for adjuvant therapy to date.

After postoperative recovery, the INCA plastic surgery team recommended multiple serial fat-grafting sessions for the patient to increase breast volume. The patient did not wish to undergo a mastopexy surgery or exhibit extensive scars.

The first fat-grafting session occurred in September 2023, and the second session occurred in early December 2024 (→Figs. 6–7).

The patient reports satisfaction with the breast volume obtained after fat grafting. She remains under follow-up with oncology, mastology, hematology, and plastic surgery clinics and presents no evidence of new lesions.

Discussion

According to estimates, more than two million Brazilian women have breast implants, making Brazil the world's second-largest market for these devices. Nevertheless, there are few reports of BIA-ALCL in Brazilian patients.⁴

The etiology of BIA-ALCL is not clear. However, according to the literature, the most common processes for BIA-ALCL development include bacterial contamination of rough/micro-textured surface implants or surface-related friction. These processes would culminate in a persistent immune response, subsequently progressing to lymphoma. Several examples support the theory that cancer may result from infection by organisms such as *Helicobacter pylori* and the Epstein-Barr virus.⁵ Furthermore, there are no reports of BIA-ALCL cases

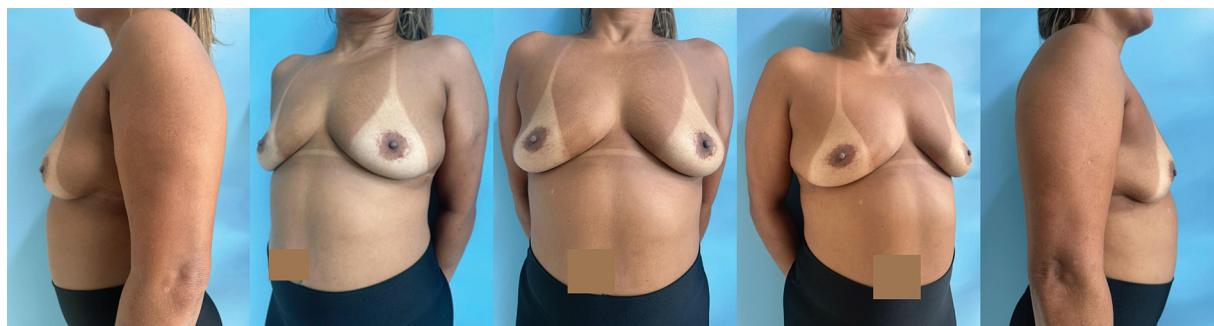


Fig. 6 Aspect three months after the first breast fat-grafting session.



Fig. 7 Aspect on the first postoperative day of the second breast fat-grafting session.

before the textured implant era, even when silicone fluids migrated to surrounding tissues and produced granulomas.⁶

In most cases, BIA-ALCL manifests as late seroma, which occurs 7 to 10 years after implant surgery. Therefore, the onset of a seroma a year or more after implant surgery that cannot be explained by trauma should be considered suspicious for the disease.

Approximately 10 to 40% of patients diagnosed with BIA-ALCL may develop a mass, cutaneous manifestations, capsular contracture, and lymphadenopathy.⁷ In our patient, the sudden increase in size of the right breast was the only evident change.

Diagnosis relies on imaging tests and cytological, IHC, and immunophenotypic analysis of seroma, revealing the presence of large, pleomorphic cells with eosinophilic cytoplasm. The IHC pattern includes CD30 protein expression and absence of anaplastic lymphoma kinase (ALK) translocation protein expression.⁸

Ultrasound is recommended as the first test, followed by aspiration of seroma fluid for immunocytochemistry and cytopathological analysis. The protocol may include computed tomography and magnetic resonance imaging if the ultrasound findings are unclear.

Rare cases, presenting a mass rather than fluid, require a pathological analysis of fragments originating from the capsular mass and skin. In some cases, a pathological evaluation of suspected lymph nodes may detect potential metastases. Our case had no signs of extramammary disease.

Treatment consists primarily of surgical removal of the breast implants and capsule. The National Comprehensive

Cancer Network (NCCN) protocol recommends that plastic surgeons confirm the diagnosis of ALCL before surgery.⁴ Another critical point is the removal of both implants and capsules, as there are cases of disease onset in the other breast, and replacement with textured implants is not recommended.^{9,10} Typical margins used in traditional breast cancer surgery are not necessary. After complete excision of the capsule and implants, surveillance exams should occur every 3 to 6 months, in addition to periodic imaging tests, including chest, abdominal, and pelvic CT or PET-CT every 6 months for the first 2 years. If complete excision is not technically feasible or if surgical pathology reveals incomplete excision or partial capsulectomy, local radiotherapy is often used as adjuvant therapy in the absence of systemic disease.¹¹ In more advanced cases, the patient should be monitored by an oncologist who may perform a lymphadenectomy. Chemotherapy and radiation therapy may also be treatment alternatives.

Breast implant-associated anaplastic large cell lymphoma has a good prognosis, and the complete removal of the capsule is a determining factor for cure. It is also crucial to have an appropriate preoperative plan and treatment in place to prevent the progression of this lymphoma. Reports of death in BIA-ALCL patients are associated with late diagnosis or more aggressive forms of the disease, including tumor nodules, lymph node involvement, bilateral breast involvement, and capsule infiltration.

Breast reconstruction after BIA-ALCL treatment is feasible and can be considered in properly selected patients. The decision regarding the timing and method of reconstruction depends on the extent of the disease at the time of diagnosis,

the completeness of surgical excision, and the patient's preferences and risk factors. Reconstruction options include smooth implant placement, mastopexy, autologous flaps, and fat grafting.¹⁰

Our patient underwent implant removal and bilateral capsulectomy. Both units were textured round implants in a subglandular position. After surgery, the patient was monitored by INCA's hematology and oncology teams, with no indication for adjuvant therapy to date, and is satisfied with the plastic surgery team's treatment plan.

Conclusion

We conclude that BIA-ALCL is a relatively new but highly relevant disease. It requires increasing awareness among patients and the medical community to increase knowledge of its diagnosis and treatment.

Clinical Trials

None.

Authors' Contributions

DV: data analysis and/or interpretation, final approval of the manuscript, data collection, conceptualization, study conception and design, project management, methodology, surgeries and/or experiments; KSM: data analysis and/or interpretation; VA: data collection; BB: investigation, surgeries and/or experiments; DYS: surgeries and/or experiments; FASL: supervision; LWP: software, visualization.

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Conflict of Interests

The authors have no conflict of interests to declare.

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