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Composite breast implant-associated anaplastic large cell lymphoma (BIA-ALCL) and extra-nodal marginal zone lymphoma (MZL) in the capsule of a silicone breast implant

Dear Editor,

Breast Implant Associated-Anaplastic Large cell lymphoma (BIA-ALCL) is a rare and poorly understood clinical entity associated with mostly textured silicone breast implants.

A 59 years old healthy woman presented with a rapidly expanding left breast in the last 6 weeks. She underwent 11 years ago an elective bilateral breast augmentation with 410 cc textured NAGOR silicone implants (Westfield Industrial Estate, Cumbernauld, Glasgow, UK).

Her physical examination revealed a very large and tensed left breast. Her right breast was in a normal shape. No lymphadenopathy was palpated.

The MRI demonstrated extensive fluid with floating particles around the intact prosthesis on her left breast (Fig. 1) with no lymphadenopathy. The right breast was normal with an intact implant. The PET-CT did not reveal any pathological activity.

A suspicious for BIA-ALCL was raised. Under general anesthesia, a massive turbid fluid and floating particles outburst from the left breast which was sent for cytological examination. The intact prosthesis was removed and a complete excision of the capsule was executed which was sent for histopathological examination. New silicone prosthesis was inserted. The post- operative period was uneventful.

The cytological examination of the seroma fluid demonstrated atypical cells suspicious for lymphoma (CD30+ and negative for ALK-1). The excisional plane (outer surface) of the capsule (O) was infiltrated by small lymphocyte (CD20+, CD10-, CD3-, CD4-, CD8-, BCL2+). Ki-67 proliferation index was 3–4% (Fig. 2A, H&E, X200, white arrows; Fig. 2C, CD20+, X200, white arrow). IgH gene rearrangement assay demonstrated monoclonal peak establishing the diagnosis of extranodal MZL. Additionally, large atypical lymphoid cells with prominent nucleoli adhered to the seroma inner luminal surface (implant) of the capsule (L). These cells were positive for CD30+, CLA, CD4, CD43 and negative for CD20, PAX5, ALK-1, CD8 (Fig. 2A, H&E, X200,black arrows; Fig. 2B, CD30+, X200, black arrow). KI-67 proliferation index was 70–80% and T-Cell Receptor gene rearrangement demonstrated monoclonal peak establishing the diagnosis of BIA- ALCL. PET-CT was normal and bone marrow biopsy had few lymphatic infiltrates of marginal zone lymphoma. The patient was asymptomatic and given the localized disease she is currently being followed closely with no evidence of disease progression.

There are two known subtypes of ALCL of the breast: (a) in situ (indolent), in which disease proliferation is confined to the capsule. (b) Infiltrative (aggressive) mostly associated with a tumor mass with cells infiltrating the capsule and adjacent tissue [1].

The more common theory of pathogenesis of BIA-ALCL is related to the textured silicone shell which degrades over time (silicone bleeding) due to a continuous friction of the implant in its fibrous pocket. The shedding of silicone particles may cause leakage of antigens that elicit chronic inflammation and a host immune response. Chronic antigenic stimulation may lead to recruitment, proliferation and expansion of T-cells, prolonging T-cell lifespan and leading to clonal expansion and eventually to malignant transformation [2]. In addition, a systemic stimulation may be enhanced causing an autoimmune response [3,4,5] which can result in the proliferation of B cells and eventually to a malignant transformation. Bizjak et al. [6] have revealed that silicone implants elicit chronic stimulation of the immune system against the prosthetic material promoting a chronic inflammatory response that may lead in genetically susceptible hosts to the development of lymphoma. Wolfram et al. [7] have revealed a genetic and cytokine profile of interleukin of producing TH17 cells which stimulate the immune response to silicone implants. Coroneos et al. [8] have found in 99,993 women who underwent breast augmentation and reconstruction with silicone implants, a significant increased risk of several rare systemic disorders classified as autoimmune disorders.

To our knowledge, this is the first report in the medical literature of composite existence of low grade B-Cell lymphoma and High grade T-Cell lymphoma in the capsule of a silicone breast implant. This case may shed light on a possible pathogenesis of BIA-ALCL and might be added as another subtype of BIA-ALCL.

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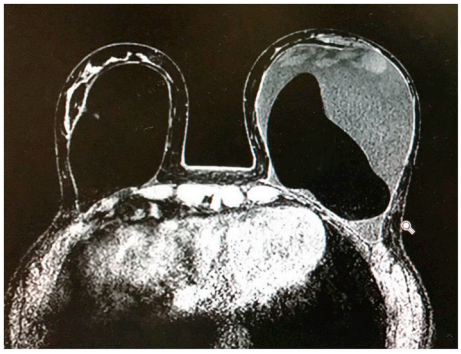

Fig. 1. A pre-operative MRI demonstrating extensive fluid around the intact prosthesis on the left breast with floating particles in the fluid. The silicone prosthesis is intact.

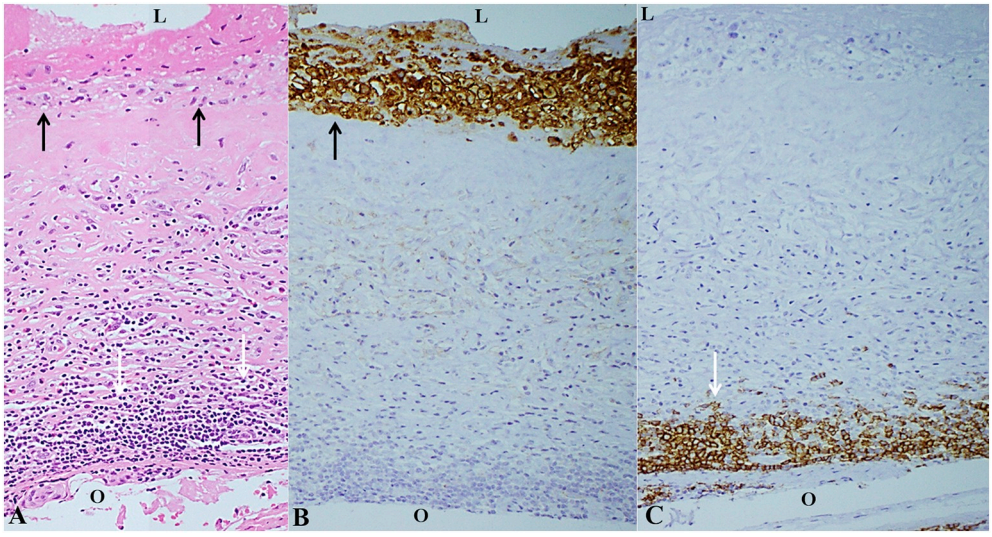

Fig. 2. A- Histological section of the excised capsule demonstrating the composite existence of BIA-ALCL (H&E, X200,black arrows) facing the inner seroma (implant) surface (L) and MZL (H&E, X200, white arrows) facing the outer excision surface (O).
B- Histological section of the excised capsule demonstrating the CD30+ staining of the BIA-ALCL (H&E, X200,black arrow) facing the inner seroma (implant) surface (L).
C- Histological section of the excised capsule demonstrating the CD20+ staining of the MZL (H&E, X200, white arrow) facing the outer excision surface (O).

Conflict of interest all authors

None.

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