Intravascular histiocytosis: case report of a rare disease probably associated with silicone breast implant

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Abstract
Intravascular histiocytosis is a rare condition characterized by the aggregate of histiocytes within dilated dermal vessels. The diagnosis is mainly histopathological and immunohistochemical. We describe a case of a 55-year-old female patient presenting erythematous/purple patches on the breasts, back and limbs. She previously presented ductal carcinoma in the right breast in 2006 which was treated with mastectomy and proceeded to silicone breast implant in 2009. Clinical hypothesis was telangiectatic metastatic carcinoma. Histopathology showed vascular ectasia, thrombosis and recanalization of upper dermis small vessels. On immunohistochemistry, intravascular cells were CD68+ and negative for estrogen and progesterone receptors, CK7, EMA and AE1/AE3 and endothelial cells were CD64+, leading to the diagnosis of intravascular histiocytosis.

KEYWORDS
Cell proliferation; Histiocytosis; Immunohistochemistry; Prostheses and implants

Introduction
Intravascular histiocytosis is a rare condition characterized by the presence of histiocytes within the dilated lymphatic vessels of the dermis. It can be primary or secondary to systemic diseases such as rheumatoid arthritis and metallic prosthesis. The pathogenesis of the disease is still unclear.

Case report
A 55-year-old female patient referred the onset of asymptomatic erythematous/purple patches in the breasts with progression to the back and proximal limbs one year and 6 months ago (Fig. 1). The patient presented with infiltrative ductal breast cancer in the right breast treated in 2006 and proceeded to the placement of silicone implant in 2009. The main clinical hypothesis were telangiectatic metastatic...
breast carcinoma and three cutaneous biopsies were performed in the left breast and one in the right thigh. The histopathology revealed ectasia, thrombosis and recanalization of upper dermal vessels and the presence of histiocytoid cells inside of those vessels (Fig. 2). The intravascular cells were negative for estrogen and progesterone receptors, CK7, EMA and AE1/AE3 in the immunohistochemistry, which excluded the hypothesis of telangiectatic metastatic breast carcinoma. The endothelial cells were CD34 positive (Fig. 3). The CD68 antibody was then utilized and was positive in the intravascular cells, confirming the histiocytoid origin and leading to the diagnosis of intravascular histiocytosis (Fig. 4).

**Discussion**

Intravascular histiocytosis is a rare condition first described in 1994. It is characterized by the presence of histiocytes within the dilated lymphatic vessels of the papillary and reticular dermis. Since 2003, the disease has been included on the spectrum of reactive cutaneous angiomatosis, as well as reactive angioendotheliomatosis. Intravascular histiocytosis may be primary or secondary mainly to rheumatoid arthritis, but also to other inflammatory diseases such as Crohn’s disease, dermatomyositis and monoclonal gammopathy, metallic prosthesis, chronic infections and neoplasms, specially breast cancer. However,
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Intravascular association of histiocytosis may rarely be observed in breast implant silicone, which usually presents as a metastatic tumor. Histology reveals intravascular proliferation of tumoral cells that can be positive for estrogen and progesterone receptors.

The course of the disease is chronic and there is not a specific treatment. In the secondary cases it is necessary to treat the overlying disease. Some patients evolved into cure after removal of metallic prosthesis. Therapy with infliximab, methotrexate, pentoxifiline, radiotherapy, corticosteroids and topic tacrolimus have variable response and frequent relapses. Our case presented an exuberant disseminated disease and possible association with silicone breast implant, for its beginning in the breasts, which association has never been described in the literature.

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Authors’ contributions
Yasmin Gama Abuawad: Conception and planning of the study; elaboration and writing of the manuscript; obtaining, analysis, and interpretation of the data; critical review of the literature.

Ticiana de Andrade Castelo Branco Diniz: Conception and planning of the study; obtaining, analysis, and interpretation of the data; critical review of the literature.

Priscila Kakizaki: Approval of the final version of the manuscript; conception and planning of the study; obtaining, analysis, and interpretation of the data; effective participation in research orientation; intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the literature; critical review of the manuscript.

Neusa Yuriko Sakai Valente: Approval of the final version of the manuscript; conception and planning of the study; obtaining, analysis, and interpretation of the data; effective participation in research orientation; intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the literature; critical review of the manuscript.

Conflicts of interest
None declared.

References