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Idiopathic Granulomatous Mastitis Associating With Erythema Nodosum: A Case Report

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Abstract: Background: *Idiopathic granulomatous mastitis (IGM) is a chronic benign inflammatory disease of the breast that can mimic breast cancer. It is more common in young women of childbearing age and poses a diagnostic of an inflammatory breast tumor.* ***Case presentation:*** *This was a 26-year-old patient who consulted for the management of an inflammatory breast in whom the examination found an apyretic patient with a left breast seat of an irregular smooth edematous and painless mass measuring 4 cm surmounted by crusts and multiple scars of fistulization leaving pus emitting after pressure with a mobile homolateral axillary lymphadenopathy. The contralateral breast was without particularity. The rest of the physical examination found inflammatory erythematous plaques on the 02 legs. A mammography plus breast ultrasound were performed followed by a biopsy, which returned in favor of a fibrous mastopathy. The persistence of the symptoms led to the completion of a lumpectomy whose histology was in favor of granulomatous mastitis. Etiological research has not found an obvious cause. Corticosteroid therapy combined with antibiotic therapy was instituted with complete regression of the symptomatology after two months.* ***Conclusion:*** *IGM poses a diagnostic problem. Its symptoms and imaging remain atypical and non-specific. The biopsy sometimes remains unsatisfactory and the use of a wide excision for histological study is more judicious. A careful investigation would be an asset to avoid mutilating surgeries without interest.*

Keywords: Idiopathic granulomatous mastitis; corticosteroid therapy; lumpectomy

Introduction:

Idiopathic granulomatous mastitis (IGM) is a chronic benign inflammatory breast disease that can mimic breast cancer **[1].**

It is an inflammation of the breast of unknown origin that must be separated from tumors and breast infections, including tuberculosis **[2]**. Its symptomatology is non-specific and the diagnosis is often not obvious. It is a non-well known entity to clinicians and radiologists **[3].**

We report a case in a 27-year-old patient who manifested as an inflammatory breast with multiple fistulizations and erythema nodosum in the lower limbs.

Patient and presentation: A 27-year-old patient living in a rural area who has consulted for the management of an inflammatory left breast. She has been breastfeeding for 7 months and has no particular history.

The beginning of his symptomatology would date back to 4 months before his consultation and would be marked by the recurrent occurrence of purulent nipple discharge associated with a skin modification with type of inflammatory cupboards and multiple fistulization of the breast that motivated a local application of herbicide. Given the persistence of the symptomatology and the appearance of reddish erythematous nodular lesions to the limbs, the patient decided to consult in our structure for management.

The general examination had found an apyretic patient in good general condition.

Breast examination found asymmetrical breasts with an inflammatory left breast seat of an irregular edematous and painless smooth mass measuring 4 cm topped by crusts and multiple fistulization scars leaving pus to pressure with lymphadenopathy mobile homolateral axillary. The contralateral breast was without particularity. The rest of the physical examination found reddish, warm, oval inflammatory erythematous plaques on all 02 legs

A mammogram performed found an opacity with ill-defined contours of interest to the outer supero quadrant (QSE) and the junction of the upper quadrants (JQS) of the left breast associated with a thickening of the skin coating next to the QSE.

Breast ultrasound found a heterogeneous hypo-echogenic range of interest to the outer superior quadrant (QSE) and the junction of the upper quadrants (JQS) of the left breast, associated with skin thickening and tumor-like axillary lymphadenopathy requiring histopathological confrontation and Left retro areolar ductal dilation to be monitored with a right sein without detectable abnormalities.

The biopsy of this hypo échogenic range returned in favor of fibrous mastopathy and the cytopuncture of homolateral axillary lymphadenopathy returned in favor of PIRINGER lymphadenitis evoking toxoplasmosis or infectious mononucleosis. Further research of the genome BK par PCR on the biopsy of lymphadenopathy had come back negative.

A prelevment of pus that flowed at the pressure of the breast was made and came back sterile.

A dermatology opinion was performed for lesions of the lower extremities and a skin biopsy of erythematous plaques on the 2 legs was performed and the histological result was in favor of predominantly septal panniculitis with histiocytic granulomas compatible with erythema nodosum.

Given the appearance of new lesions in the left breast and the lack of diagnosis explaining the clinical picture, a breast ultrasound was redone and it described an ultrasound aspect in favor of several collections in the left breast with homolateral axillary lymphadenopathy.

The patient's file was thus discussed in a multidisciplinary consultation meeting and a decision to make a surgical excision of the mass of the left breast was taken with a more exhaustive etiological assessment. A re-examination of the biopsy of left axillary lymphadenopathy concluded that there was reactive adenitis.

The patient received a lumpectomy of the left breast during which the exploration found pus encysted with a very crumbly mammary gland.

The histological study of the lumpectomy piece found a mammary parenchyma made of regular ducto-lobular structures with palleal tissue dissociated by epithelioid and gigantocellular granulomas centered by foci of suppurative necrosis without tumoral proliferation describing granulomatous mastitis with suppurative necrosis without tumor proliferation.

The etiological balance was without particularity. It consisted of serologies (HIV, hepatitis B, C and syphilis), BK research, neutrophil anticytoplasm antibodies (NAFC), C3 and C4 supplements, assessment of renal function (urea, creatine and proteinuria) and fasting blood glucose.

The final diagnosis of idiopathic granulomatous mastitis combining erythema nodosum was retained.

The patient was then put on medical treatment based on oral corticosteroid (prednisone 20mg) for 6 weeks and antibiotic therapy based on amoxicillin plus clavulanic acid 1g / 8 h for 21 days and metronidazole 500mg per 8h for 14 days. The dosage of the corticosteroid was 1mg /kg/day either : 60 mg per day for 15 days, then 40 mg per day for 15 days, then 20 mg per day for another 15 days.

The patient was monitored weekly for the first 15 days of treatment and then monitored every 15 days until treatment was discontinued. During each appointment she received a clinical breast examination assessing the regression of the symptomatology. 2 months after the end of treatment, there was a complete regression of the symptomatology. The control breast ultrasound described a total disappearance of the existing lesions before treatment.

Discussion: MGI is a benign inflammatory disease of the breast. It would be an idiopathic disease due to the influence of certain environmental stimuli in genetically predisposed subjects **[1]**. The pathological mechanism is still poorly understood. To date, three main hypotheses have been raised to explain this disease: autoimmune genesis, infectious disease and hormonal disorders **[4,5]**. This pathology occurs mainly in women during periods of genital activity **[6]** but can also occur in postmenopausal women. In a study **[7]** carried out on a series of 20 cases, the authors reported that MGI represented 2% of breast pathologies managed in their structure with an average age of 38.1 years and 55% of women belonged to the age group of 30 to 39 years.

Clinically, the master symptom of MG is a painful mass and about 50% of patients develop erythema and tumefaction as symptoms of inflammation of the affected breast. Other symptoms are hyperemia, area retraction, fistula and ulceration. About 37% of patients show signs of abscesses **[8]..** Sometimes MGI can manifest itself as a hardening of the breast with collections of pus in a non-febrile context evoking a cold abscess of the breast and leading to a multitude of consultation and attempted management **[3].** This symptomatology is still non-specific.

On imaging, the lesions described do not differ from those described in some forms of breast cancer. In our specific case, mammography described opacity with ill-defined contours and breast ultrasound found a heterogeneous hypo echogenic range associated with skin thickening as well as axillair lymphadenopathy with many focus of collections.

The diagnosis of certainty is based on the histology of either a biopsy fragment and/or a lumpectomy piece. The histological study of our case found a mammary parenchyma made of regular ducto-lobular structures with palleal tissue dissociated by epithelioid and gigantocellular granulomas centered by foci of suppurativous necrosis without tumor proliferation.

Therapeutic management is essentially based on surgery, which consists of a wide removal of lesions, preceded by corticosteroid therapy to reduce lesions **[6].**

Conclusion: Idiopathic granulomatous mastitis is a rare clinical entity that poses a diagnostic problem as we see in this case. Its symptomatology and imaging remain atypical and non-specific. The biopsy is sometimes unsatisfactory and the use of a large excision for histological study is more judicious in case of biopsy result not consistent with the symptomatology.

Competing interests: Authors do not declare any competing interests.

Authors' contribution: All authors participated in the patient's care, writing and proofreading of the manuscript. All authors have read and approved the final version of the manuscript.

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