Case Report ISSN 2768-6647

Medical and Clinical Case Reports

Breast Sarcoidosis: Case Report and Review of the Literature

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Received: 17 Jun 2023; **Accepted:** 19 Aug 2023; **Published:** 25 Aug 2023

Citation: El Hachami Fz, Maali I, Imami Y, et al. Breast Sarcoidosis: Case Report and Review of the Literature. Med Clin Case Rep. 2023; 3(3): 1-3.

ABSTRACT

Sarcoidosis is an idiopathic, systemic, granulomatous inflammatory disease of unknown etiology that affects both sexes with predominance in young women. It is characterized histologically by the presence of epitheliocellular granuloma without caseous necrosis. It usually affects the lungs, skin, liver, spleen, eyes, bone phalanges and parotid glands.

Granulomatous mastitis is a benign inflammatory pseudotumor. This rare anatomoclinical entity poses a problem of differential diagnosis with other granulomatoses and especially with breast carcinoma. The coexistence of systemic manifestations should lead to the discussion of sarcoidosis. In our work, we report 2 cases of breast sarcoidosis, which were treated and cured.

***Objective:** To establish the diagnostic criteria of the granulomatous mastitis, and to quote the major elements to eliminate the other differential diagnosis especially the mammary carcinoma.*

Keywords

Sarcoidosis, Breast, Epitheliocellular granuloma, Granulomatous mastitis, Giganto-epitheliocellular nodules, Caseous necrosis, Tuberculoid granuloma.

Patient Et Observation

The patient was 42 years old, married, with no particular pathological history. The history of the disease dates back to 3 months ago with the autopalpation of a mammary nodule in the left breast, the whole evolving in a state of conservation of the general state.

The clinical examination revealed a breast swelling at the level of the QIE with the presence of a fistula and pus emission, the contralateral breast was without particularity, with free lymph nodes. Mammography found breasts of type D density, with the presence of an excess of density opacity in the left breast without any architectural rupture or clearly circumscribed individualizable nodular formation (Figure 1).

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On breast ultrasound, there was no clearly circumscribed mass but there was a hyper echogenic infiltration of the QIE of the left breast. This examination suggested either a lipomatous or hamartomatous formation.

A trucut biopsy was performed showing lesions of chronic inflammatory granulomatous suppurative mastitis on non-proliferative sclerocystic mastosis with no in situ or invasive breast neoplasia and no sign of specificity.

A serological workup was performed showing negative anti-NPN cytoplasmic antibodies (ANCA), angiotensin converting enzyme with a value of 18 CEU, which was normal. The research of anti soluble nuclear antigens antibodies showed: anti Jo 1, anti DFS 70 and antientromere B antibodies were positive. anti RNP/Sm antibodies, anti SS -B antibodies, anti SSA antibodies, anti Ro 52 antibodies, anti Scl-70 antibodies, anti Sm antibodies, anti RIBOSOMAL antibodies, anti HISTONE antibodies, anti MITOCHONDRIES-AMA M2 and anti-dsDNA antibodies were negative. Protein electrophoresis was without abnormality. The

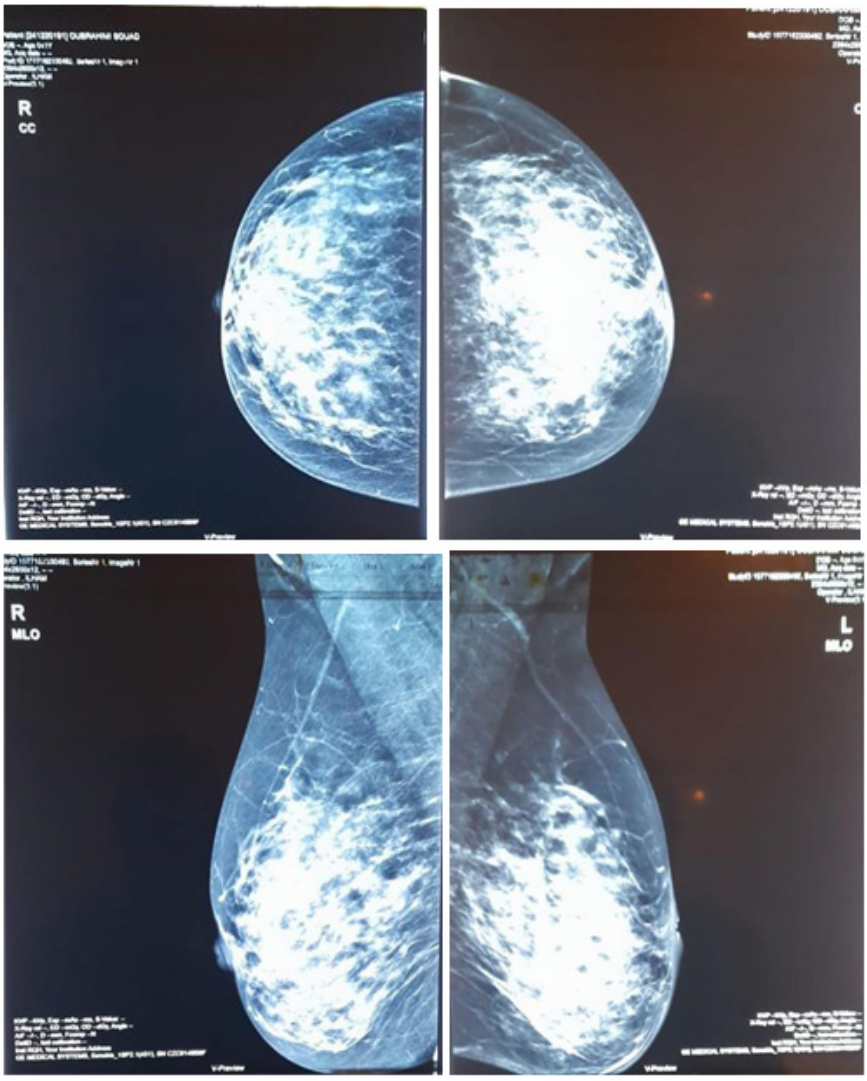

Figure 1: Mammographic appearance.

CRP was negative with a value of 3.9 mg/l. The blood test for interferon gamma production was negative, making M.tuberculosis infection unlikely. A thoracic CT scan revealed no significant abnormalities. The patient was put on corticosteroids with good evolution.

Discussion

The diagnosis of sarcoidosis is based on a combination of clinical, radiological, biological, histological elements, none of which taken in isolation is specific for the disease, and therefore none of which is both necessary and sufficient. According to the currently accepted definition, the diagnosis is based on the combination of several data. Granulomatous mastitis is a rare chronic inflammatory benign mastopathy. It represents 0.5% of breast tumors affecting the proximal subareolar milk ducts, and is found in women during their genital period [1-11].

Clinical Arguments

The usual young age of the subject suffering from sarcoidosis, the absence of any notion of tuberculosis infection, the usual functional latency at least in the recent forms of the disease, the association of multiple and evocative intra-thoracic and extra-thoracic localizations such as: (specific cutaneous, ophthalmological, ganglionary...) expressing the systemic character of the disease[14-15] .

It was first described and named Mastitis obliterans by INGIER in 1909 [7]. In 1951, Haagensen [8-13] named it perigalactophoric granulomatous disease. Two theories are incriminated to explain the etiopathogenic mechanism [9]: According to Haagensen, the first cause is galactophoric dilatation with a stasis of secretory products generating a mechanical or chemical rupture of the ductal epithelium, and thus triggering an inflammatory reaction; according to BOSNER, it is the peri-ductal inflammation that will lead to galactophoric alteration following conjunctival lesions; other theories have been evoked, in particular, an infectious origin or a reaction to a trauma. However, the common lesion remains an aggression of the ductal epithelium causing extravasation

of glandular secretions into the connective tissue of the lobule responsible for the granulomatous inflammatory lesions. This pathology could simulate macroscopically and radiologically a neoplastic lesion. The diagnosis of certainty is histological showing a granulomatous mastitis.la littérature a rapporté uniquement 35 cas de localisation mammaire de sarcoïdose histologiquement prouvés [6,4]. La localisation mammaire peut être primitive [5]. Cliniquement, elle se manifeste par des masses palpables au niveau des seins, dont la taille varie de 0,5 cm à 8 cm [5,6]. Les résultats de l'imagerie en matière de sarcoïdose mammaire ont été rapportés dans un nombre limité de cas [4-6].

Mammography was reported in nine cases. In three cases, the contours of the mass were blurred or spiculated, simulating malignant lesions; two mammograms were negative, and in the other cases, the results were nonspecific; thus, breast sarcoidosis can often mimic breast cancer, with the absence of microcalcifications in most cases [6]. Breast ultrasound was non-specific and histology confirmed the diagnosis Treatment Depending on the case, sarcoidosis may be cured without treatment in about 50% of cases, or it may require treatment that can be definitively effective within 12 months [2]. In some cases, corticosteroid therapy must be extended beyond 12 months or even indefinitely. Prednisone remains the standard of care. Biopsy remains the reference examination showing epithelioid granulomas associated with a polymorphic inflammatory infiltrate consisting of plasma cells, lymphocytes and neutrophils [9]. The differential diagnosis is mainly with inflammatory breast cancer and infectious or non-infectious mastitis (cytosteatonecrosis, sarcoidosis, Wagner's disease) [10].

Granulomatous mastitis can only be labeled idiopathic after having eliminated all causes of granulomatous mastitis and also in order not to neglect a chance of rapid cure by an adapted treatment especially as it is a chronic and lingering pathology. No consensus has been established for the treatment of granulomatous mastitis. At the stage of inflammatory mastitis, treatment is based on antibiotics and anti-inflammatory drugs, but at the stage of collected abscesses, drainage remains the treatment of choice while continuing medical treatment [7-12].

Conclusion

The mammary localization of sarcoidosis, despite its very rare incidence, must be evoked in the differential diagnosis of the mammary nodule, in women with a history of sarcoidosis. MRI can be used to rule out a carcinomatous lesion because of its negative predictive value. However, histology is usually required to confirm the diagnosis.

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