

# Granulomatous mastitis: A case report

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## Abstract

Idiopathic granulomatous mastitis is a rare chronic benign inflammatory mastopathy occurring mainly in young women. With a non-specific imagery, it is considered as a diagnosis by exclusion and has a challenging treatment. Histologically, it is characterized by the predominance of polynuclear neutrophils and the absence of caseous necrosis. The breast carcinoma is the main differential diagnosis at the clinical stage, and imagery plays an essential role in its diagnostic approach. Its treatment combines antibiotics, anti-inflammatories, corticosteroid therapy, and surgery. We report a 42-year-old woman who presented a breast lesion diagnosis as granulomatous mastitis based on magnetic resonance imaging, ultrasound, and histology examinations. She received a treatment with different drugs (antibiotics, anti-inflammatories). The outcome treatment was successful with a good healing of breast lesions.

## Keywords

Granulomatous, mastitis, imaging, histopathology, diagnosis

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## Introduction

Lobular granulomatous mastitis (LGM), also known as idiopathic granulomatous mastitis (IGM), is a rare chronic breast disease occurring mostly in young women. Despite its rapidly increasing incidence in recent years, its etiopathogenesis remains unsolved. Autoimmune disorders have been suggested on several arguments.<sup>1–3</sup> It remains a diagnostic and therapeutic challenge. Breast carcinoma is the main differential diagnosis at the clinical stage. Its imagery is non-specific. Only histology is the key to a positive diagnosis by objectifying lobular necrosis and non-caseous granulomatous inflammation. Treatment and evolution remain conditioned by the underlying etiology.<sup>1,3,4</sup> We report the case of a patient who presented a breast lesion diagnosis as granulomatous mastitis.

## Case report

The patient is a 42-year-old woman, with no prior history of tuberculosis disease or tuberculous contact and no familial neoplasia history. She has five children and adopts a contraceptive device since 5 years ago after her last delivery.

The symptomatology has been evolving 3 months before her admission with the appearance of the right breast swelling. The gradual increase in the lesion volume was the chief complaint during her first consultation.

The clinical examination found a swelling of the right breast, extending to its outer half. It was associated with local inflammatory signs, nipple retraction, and mobile, homolateral, axillary lymphadenopathy, which measured about 2.5 cm. There was no nipple discharge. The patient had no infectious sites, and the infectious biology check-up was strictly normal. An initial ultrasound mammogram found lesions of the supero-external quadrant of the right breast, classified ACR 4 (Breast Imaging Reporting and Data System of American College of Radiology), and the patient

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was given an antibiotic therapy based on fluoroquinolone 200 mg, twice daily for 8–10 days.

After 7 days of treatment, a cutaneous fistulization appeared and the inspection of the breast found a clinical sign described as “orange peel,” resulting from pus to the skin (Figure 1). Then, an additional magnetic resonance imaging (MRI) was requested. The MRI found a lesional range made of multiple oval formations, occupying the entire gland and extending to the skin (Figure 2). The comparison of their fluid signal to the control ultrasound images led to the possibility of an IGM (Figure 3).

Histology, performed on three biopsy cores ranging in size from 5 to 14 mm long, found no signs of malignancy or pathogen on the samples. It showed a fibrous breast parenchyma with a polymorphic inflammatory infiltrate and



**Figure 1.** Pseudo-tumor right breast, with a retraction of the nipple and an appearance of “orange peel” resulting from pus to the skin via a skin fistula (blue circle).

decentered epithelio-gigantocellular granulomas of caseous necrosis (hematoxylin and eosin (HE)  $\times 10$ ) in favor of IGM (Figure 4).

Further treatment was done by antibiotic therapy and wound dressings. The evolution was favorable with a regression of inflammatory signs and a progressive healing.

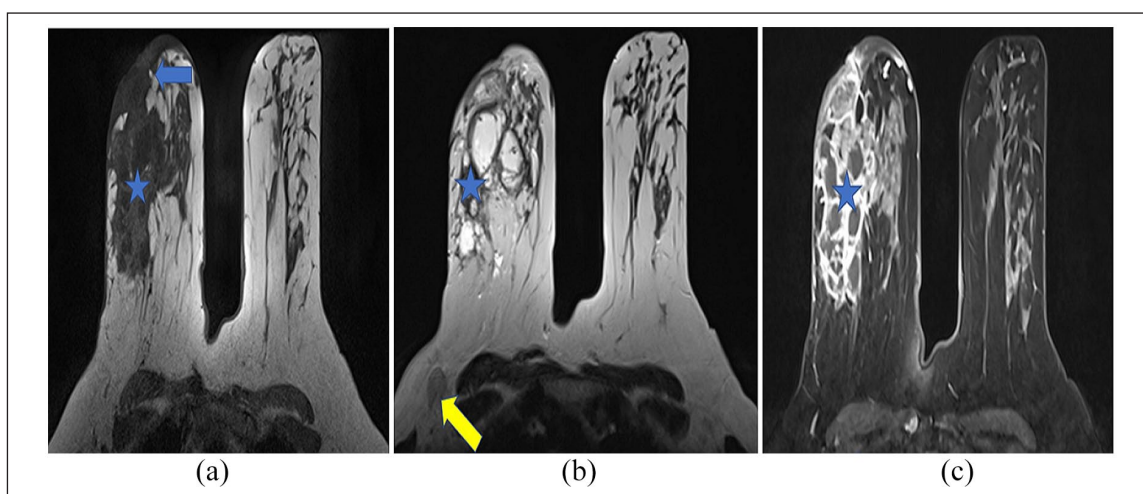
The long-term follow-up was as follows: the patient’s checkups at 3 and 6 months were very satisfactory with a complete regression of the picture and a definitive healing of skin lesions. Because the patient missed the 12 months appointment, she had been called and reassured that she was in good clinical condition.

## Discussion

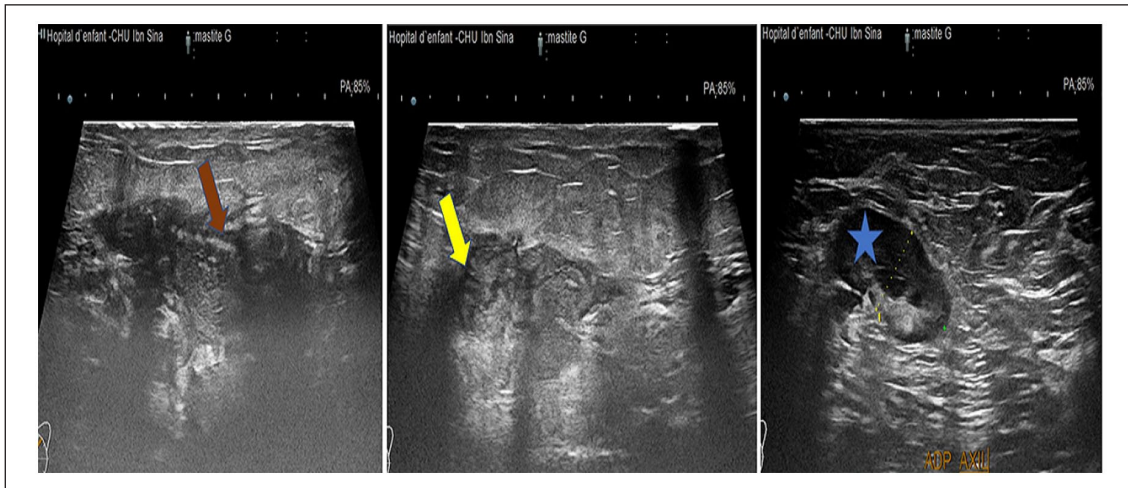
Granulomatous mastitis was first described by Veysiery et al. in 1967.<sup>5</sup> It is a chronic benign breast disease, which affects women of childbearing age with a history of childbirth and breastfeeding. It usually occurs within 5 years of the last delivery.<sup>1,2</sup>

Kessler and Wolloch<sup>6</sup> in 1972 made a clinical, histological, and evolutionary description. Its etiopathogenesis remains little known and diversified. Several hypotheses have been put forward explaining a secondary inflammatory reaction to mechanical, traumatic, hormonal, and metabolic factors. The aggression of the ductal epithelium would be responsible for the extravasation of glandular secretions in the connective tissue of the lobule, creating local inflammation. Immunological disorders related to IgG4 have also been proposed.<sup>1,2</sup> Altintoprak et al.<sup>7</sup> incriminated rare bacterial infections and the polymorphism related to the genetic mutation.

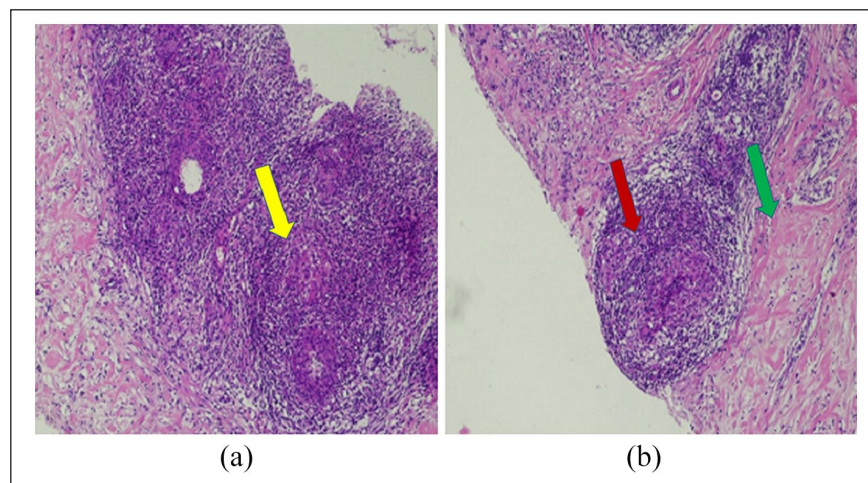
The clinic typically begins with one or more inflammatory nodules of varying size, usually unilateral and extra-areolar. The pain is inconstant, and pseudo-tumor forms are spontaneously observed, mimicking cancer with a retraction



**Figure 2.** Breast MRI showing lesional ranges as multiple oval and rounded formations with irregular thick walls in hyposignal T1 (a) and hypersignal T2 (b), with a peripheral enhancement (c) after fat suppression. Note a homolateral axillary adenopathy (yellow arrow).



**Figure 3.** Ultrasound aspect of the right breast showing an abscess in the form of a heterogeneous hypoechoic collection (yellow arrow) with another uncollected part in heterogeneous echogenic form with posterior enhancement (brown arrow). Note an axillary adenopathy (blue star).



**Figure 4.** (a) Fibrous mammary parenchyma having decentered epithelial-gigantocellular granulomas of caseous necrosis (yellow arrow), HE  $\times 10$ ; (b) fibrous mammary parenchyma (green arrow) having epithelial-gigantocellular granulomas (red arrow) not centered with caseous necrosis, HE  $\times 40$ .

of the nipple and an “orange peel” appearance. Nodules can secondarily be complicated by skin ulcerations, micro-abscesses, or fistulization of the skin, leaving visible scars. Inflammatory flare-ups are sometimes accompanied by satellite axillary lymphadenopathy and general signs (fever, asthenia).<sup>1,3</sup>

Generally, there is no inflammatory syndrome, and mycobacteriological samples are sterile. In case of abscess, moderate inflammatory syndrome and superinfection with *Staphylococcus aureus* can nevertheless be observed.

Imaging is polymorphic and non-specific. It confirms mastitis on radiological arguments that are readily confused with those of a mammary carcinoma. Its purpose is to properly assess the lesional extent, the effectiveness of

conservative treatment, and then to make a correct judgment on the breast volume to be removed during surgery and the volume of the fascia flap useful to replace the lesion.<sup>1,8</sup> Ultrasound, in abscess forms, allows evaluating the lesional size and to better understand the type of drainage to be performed (puncture or surgical flattening). It describes a variety of lesions that are often non-specific if not suggestive of malignancy. The most described aspects are hypoechoic irregular masses with blurred contours, hyperechoic, sometimes heterogeneous with galactophoric ectasias. The territories surrounding them are often hyperechoic. According to Engin et al.,<sup>9</sup> multiple heterogeneous images relatively circumscribed with a tubular configuration associated with a hypoechoic large mass should evoke granulomatous

mastitis. Another aspect is that the absorption of ultrasound beam at the posterior part of the lesions described by Van Ongeval et al.<sup>10</sup> and Alper et al.<sup>11</sup> can cause a variable extended parenchymal distortion and variable images of a fistulized abscess.

Mammography often finds an asymmetrical focal density without micro-calcifications (44% of cases). MRI in the study of this mastopathy is a technique that has shown suspicious lesions of malignancy in the form of irregular tissue masses. In 64% of cases, the lesions were rounded, oval, or lobulated. They took the contrast after gadolinium injection in the majority of cases, with a peripheral enhancement (abscess). Only enhancement kinetics after injection of a contrast medium could help in the differential diagnosis with mammary carcinoma. Depending on the literature, MRI would be useful in assessing the extension and reduction of lesions.<sup>1,2,8</sup>

The definitive diagnostic proof is histological. The results show epithelioid granulomas without caseous necrosis associated with a polymorphic inflammatory infiltrate, which are made of plasma cells, lymphocytes, and neutrophils. The usual colorations (Periodic Acid Schiff (PAS), Grocott, Ziehl) are negative.<sup>5</sup>

Clinical differential diagnosis is mainly with carcinoma-mastitis. It can also occur with mammary tuberculosis, especially in endemic areas, but the predominance of neutrophils and the absence of caseous necrosis argue in favor of mastitis granulomatous. Mastitis should be distinguished from bacterial, parasitic, and mycotic infectious mastitis as well as from non-infectious granulomatous lesions (lipophagic granuloma or cytoosteocrosis, sarcoidosis, plasma cell mastitis, lymphocyte mastitis, etc.).<sup>2</sup>

The uncodified and still controversial treatment should be done in stages, after a pathological diagnosis. Antibiotics, anti-inflammatories, corticosteroid therapy, and surgery remain the main treatments that must be carried out early. Evolution is unpredictable, sometimes spontaneously favorable. Typically, it alternates flare-ups of varying intensity, with more or less long periods of remission.<sup>1</sup>

## Conclusion

IGM is a rare breast pathology and has unpredictable evolution over time. The clinical and radiological aspects are variable and pose a diagnostic problem especially with breast cancer. The pathological examination remains the main element of certain diagnosis. The clinical correlated treatment remains controversial.

## Author contributions

O.K.R. obtained the relevant radiologic images, with consent from the patient and produced the manuscript. All authors worked collaboratively to finalize the manuscript, as well as make critical revisions of, and approve the final manuscript.

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## Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

## Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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