*Case Report*

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# Granulomatous lobular mastitis associated with ductal carcinoma in situ of the breast

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**Abstract**

The association of granulomatous lobular mastitis and carcinoma of the breast is very infrequent. We present the case of a 44-year-old woman with concurrent granulomatous lobular mastitis with coryneform bacteria and ductal carcinoma in situ in the same breast.

**Keywords**

Granulomatous lobular mastitis, ductal carcinoma in situ, breast, coryneform bacteria

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## Introduction

Granulomatous lobular mastitis (GLM) is an infrequent chronic inflammatory disease of the breast; however, its frequency seems to be increasing.[1,2] The cause of GLM is still unknown, although it has been linked to multiple infectious and non-infectious causes.[3] On the other hand, mammary carcinoma is the most frequent malignant tumor in women, especially invasive carcinoma; however, the incidence of ductal carcinoma in situ (DCIS) has increased significantly in the recent years, probably due to more frequent breast tumor screening through mammography.[4] The synchronous concurrence in the same breast of GLM and invasive mammary carcinoma is very rare; moreover, the coexistence of GLM and pure DCIS has not been communicated until this report. The following is a case of a GLM with coryneform bacteria coincident with DCIS in the same breast.

## Case

Here, we present the case of a 44-year-old woman with a medical history of coronary disease with myocardial infarction and stent placement at 29 years old. The patient smoked for 10 years until 39 years old and she did not have a family history of heart or breast diseases. She presented with a 2-week history of left breast swelling with a painful and inflammatory mass in the upper-outer quadrant, measuring approximately 7 cm. Under medical evaluation, she received cefadroxil as a treatment. Breast ultrasound revealed inflammatory changes characterized by cutaneous thickening, increasing echogenicity of subcutaneous tissue, superficial laminar collection, and loss of fatty planes due to multiple anfractuous collections (Figure 1(a)). Blood laboratory analysis showed normal leukocyte count and a mild elevation of inflammatory markers (C-reactive protein and erythrocyte sedimentation rate).

After 1 week of treatment, clinical worsening was noticed, with increasing redness and edema. A new ultrasound was performed showing no abscess formation and she underwent an ultrasound-guided core biopsy (Figure 1(b)). On histological examination, the breast tissue revealed an extensive, chronic-active inflammatory process which included granulation tissue with suppurative granulomas, some with lobulocentric pattern, with an optically empty central vacuole and Gram-positive coccobacilli. These findings were present in an area with solid DCIS, nuclear grade 2. The diagnosis of GLM with coryneform bacteria associated with DCIS was established (Figures 2 and 3). Cefadroxil was maintained for

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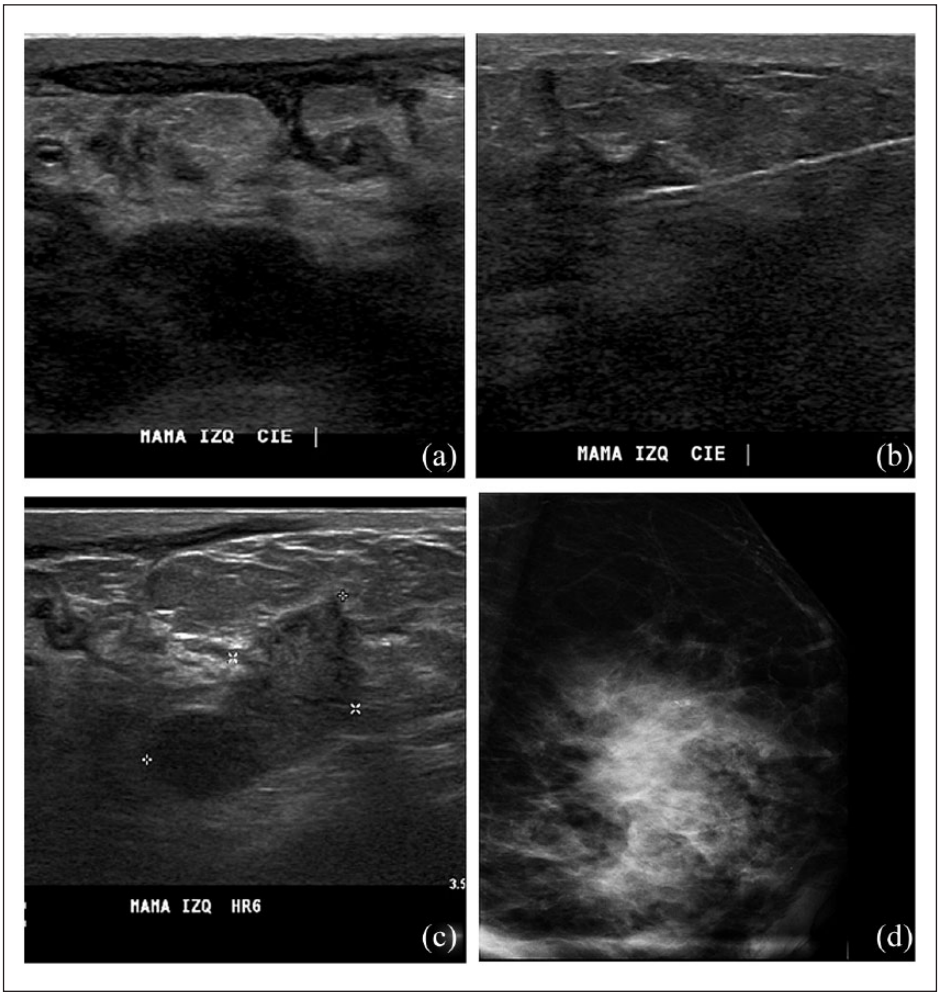


**Figure 1.** (a) Breast ultrasound shows left breast with cutaneous thickening, increasing echogenicity of subcutaneous tissue, superficial laminar collection, loss of fatty planes, and multiple anfractuous collections. (b) Percutaneous biopsy under ultrasound vision shows puncture needle (linear echogenic image) in collection area. (c) Ultrasound control shows no significant changes with respect to inflammatory changes. There is an anfractuous collection in the union of both outer quadrants of the left breast. (d) Digital mammography (localized and magnified plate) shows a diffuse dense breast with focal areas of higher density, related to areas of collections with no evident microcalcifications.

10 days followed by a 6-day course of azithromycin. Prednisone at 1 mg/kg was also started. An initial favorable response to corticosteroids was noticed, but a clinical worsening with increasing pain and fever presented when steroids doses were lowered. Multiple collections in the upper-outer quadrant and lower-outer quadrant developed during next months, requiring multiple antibiotics schemes and several collection drainages under local anesthetics. Cultures taken from the collections were all negative for aerobic germs.

Mammogram and breast ultrasound were performed after 4 months of treatment searching for images suggestive of DCIS involvement, but both studies revealed no microcalcifications or other suspicious images for malignancy (Figure 1(c) and (d)). During the next months, she presented clinical worsening with several inflammatory collections growing in the outer quadrants, and she underwent incisional surgery and drainage of the collections. The pathological and microbiological studies were repeated. The microscopic analysis of the breast tissue obtained revealed similar pathological findings to those described in the first biopsy. Therefore, the diagnosis was similar: GLM with coryneform bacteria associated with DCIS of nuclear grade 2, with intensely positive estrogen and progesterone receptors. With the Ziehl–Neelsen and periodic-acid Schiff (PAS) stains, no infectious agents were found. Aerobic, anaerobic, fungi, and mycobacteria cultures were negative.

The serology for rheumatologic conditions revealed no evidence of inflammatory diseases. Methotrexate was started with a mild and slow improvement of local inflammation, but with incomplete response.

Subsequently, and given the progression of the disease, the patient underwent a lumpectomy. The study showed an extensive zone with DCIS with several surgical edges involved and also associated with GLM. With this information, the patient

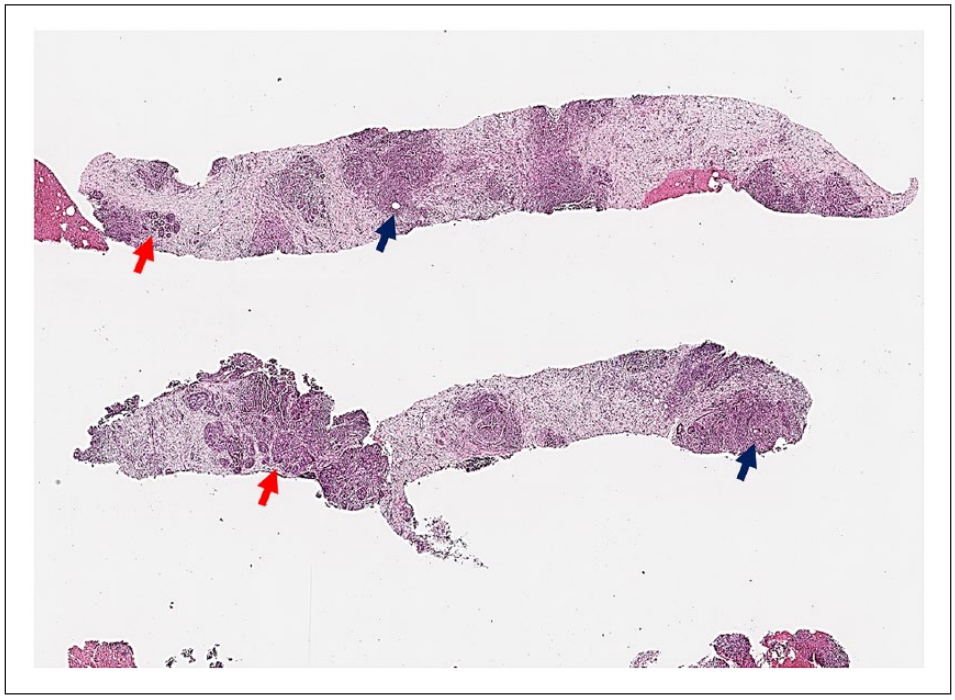

**Figure 2.** Core biopsy of the breast. Note the multifocal distribution of the lesions; there are multiple foci of granulomatous lobular mastitis (blue arrows) and areas with ductal carcinoma in situ (red arrows) (hematoxylin–eosin 4×, original magnification).

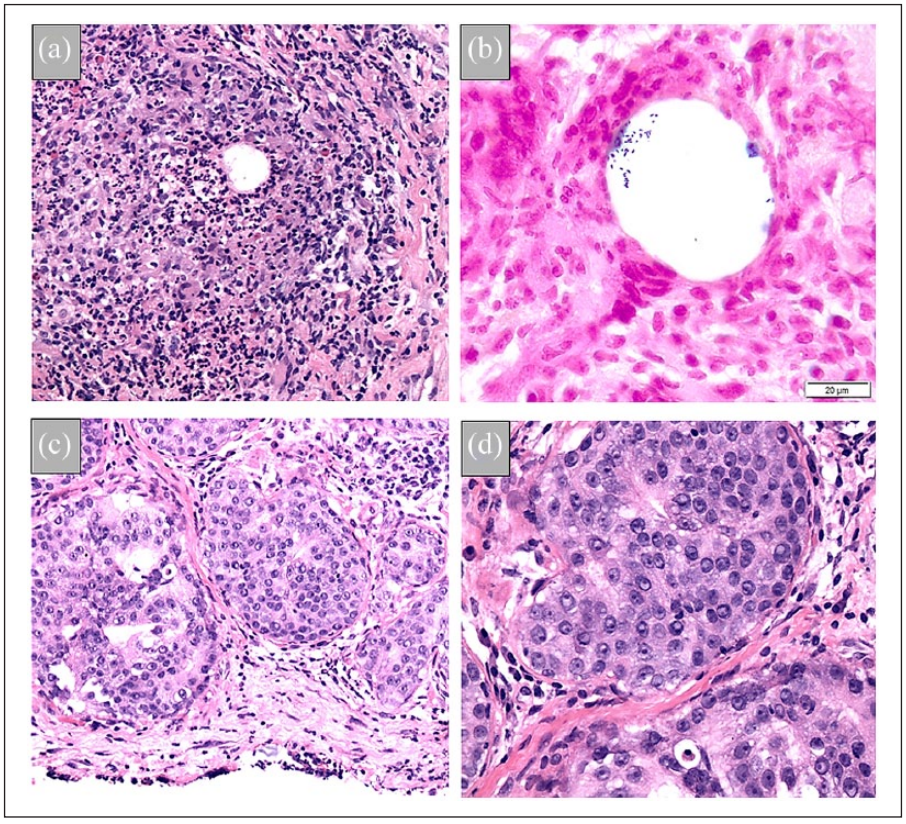


**Figure 3.** Core biopsy of the breast. (a) Granulomatous lobular mastitis (hematoxylin–eosin 10×, original magnification). (b) Coryneform bacteria in granulomatous lobular mastitis (Gram stain 40×, original magnification). (c) and (d) Solid ductal carcinoma in situ (hematoxylin–eosin 10× and 40×, respectively, original magnification).

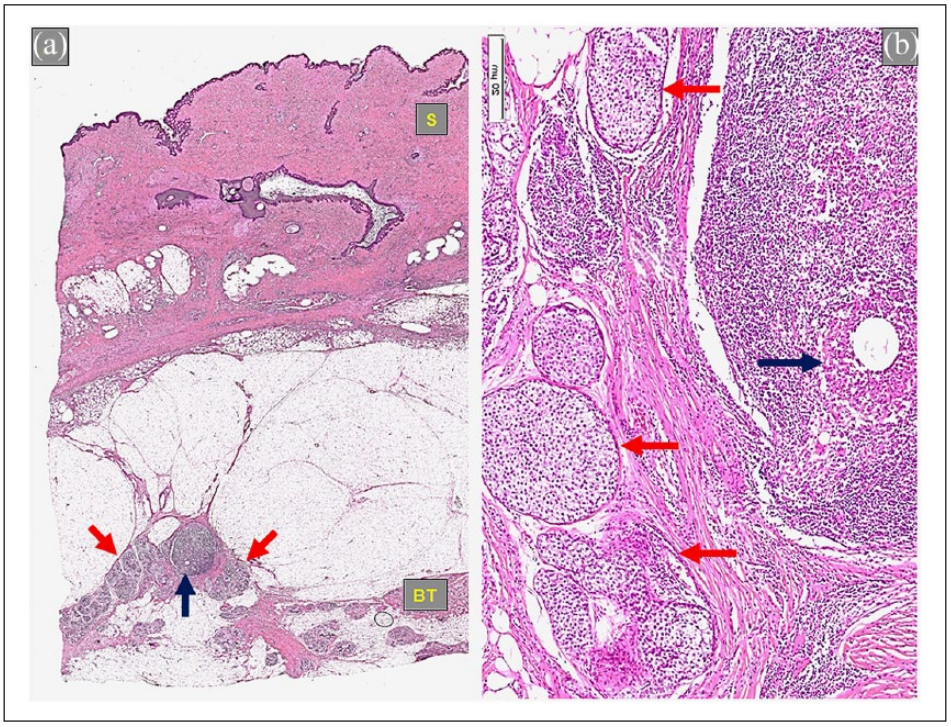


**Figure 4.** Surgical biopsy of the breast. (a) Skin (S) and breast tissue (BT). There are two areas with ductal carcinoma in situ (red arrows) and an area of granulomatous lobular mastitis (blue arrow) (hematoxylin–eosin 4×, original magnification). (b) Ductal carcinoma in situ (red arrows) coalescent with granulomatous lobular mastitis (blue arrow) (hematoxylin–eosin 10×, original magnification).

underwent a total mastectomy; we found, in both external quadrants, a wide region with DCIS, distant to more than 1 cm from all the surgical edges, also in coexistence with GLM (Figure 4).

## Discussion

At the time of this publication, there are only four other cases published in English (PubMed) about the concurrent condition of GLM with mammary carcinoma. In three cases, both lesions were in the same breast, and in two of those the association was with invasive ductal carcinoma.[5,6] In one case, the association was with invasive ductal carcinoma and DCIS simultaneously.[7] In the last case, the disease affected different breasts, but in a synchronous manner; one breast with GLM and the other with invasive ductal carcinoma.[8] In none of the four cases, the presence of bacteria or other infectious agents in the breast tissue was mentioned. Herein, in our case, a concurrent GLM with coryneform bacteria and DCIS was demonstrated in the same breast.

After GLM was recognized as a pathological entity, it was determined that this disease can simulate invasive breast carcinoma by its clinical manifestations and its imaging signs. As a consequence, the breast biopsy, especially the core biopsy, became the most important and irreplaceable method to make a correct diagnosis and perform the most precise treatment.[9–12]

The concurrence of GLM and carcinoma brings the classic and extensively described discussion about the relationship among mastitis, infection, and breast cancer. It raises the theory that inflammation and chronic infection can be related to cancer.

The association between mastitis and mammary carcinoma was described many decades ago;[13] however, unlike several other organs in which the inflammation induced by infectious agents or irritating substances have been strongly linked to the development of malignant tumors, this has not been demonstrated in breast tissue. Despite the fact that the overall risk of breast cancer seems slightly higher in women with history of mastitis, a causal relationship between inflammation and development of tumor lesion has not been well established.[14]

The causes of GLM are not well defined; it has been associated with several possible etiologies and pathogenesis, such as infectious agents (especially, lipophilic corynebacteria), autoimmune processes, erythema nodosum, hormonal alterations, use of oral contraceptives, hyperprolactinemia, pregnancy—birth and lactation, cigarette smoking, alpha 1 antitrypsin deficiency, interferon-alpha therapy, IgG4-related disease, Rosai–Dorfman disease, and even ethnic conditions.[3] It is possible that the anatomopathological picture of GLM is the morphological expression of different causes. Therefore, the treatment of GLM is not standardized and includes surgery, antibiotics, steroidal and nonsteroidal

anti-inflammatory drugs, and immunosuppressive drugs, with most of the cases requiring medical treatment for a long period of time. The persistence and/or recurrences of the disease are frequent.[15,16]

In the case herein we presented, we concluded that the association of GLM and DCIS was coincidental, which added greater complexity to the treatment and follow-up of the patient.

## Conclusion

The rare coexistence of GLM and DCIS in the same breast emphasizes the importance of biopsy in the diagnosis, treatment, and follow-up of patients for this uncommon association. This very rare condition is challenging for radiologists, pathologists, and breast surgeons.

### Declaration of conflicting interests

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### Ethical approval

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

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### Informed consent

Written informed consent was obtained from the patient for her anonymous information and accompanying images to be published in this article. A copy of the written consent is available for review on request.

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