CASE REPORT

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# Recurrent Non-Lactational Fungal Abscesses in a Systemic Lupus Erythematosus Patient: Causation or Mere Association? A Case Report and Literature Review

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## ABSTRACT

**Background:** Systemic lupus erythematosus is a multisystemic autoimmune disorder that can present in many different ways that can be debilitating for many patients. These patients are at risk for developing infections following the introduction of immunosuppressive therapy. Breast infections, particularly of the fungal type, in nonlactating patients who are not in an immunosuppressive state are extremely rare. **Objective:** We report a case of recurrent right breast fungal infections manifesting in the form of multiple abscesses in a systemic lupus erythematosus patient. **Case report:** A 39 years old female patient presented with recurrent fungal breast abscesses. She was diagnosed with systemic lupus erythematosus nine years ago and was in remission being maintained with an antimalarial agent without the use of immunosuppressive therapy. Fluconazole was started for her prior to her visit to us, and she had no active complaints. She was not breastfeeding nor pregnant currently nor during any of the previous episodes. Examination was unremarkable, however cultures of samples from her previous lesions demonstrated growth of Candida albicans. A decision to manage her conservatively with the continuation of her antifungal therapy was made. **Conlusion:** Lactation and breastfeeding are well-known risk factors for infectious mastitis and there is sparse literature regarding this condition in the absence of these risk factors. Studies evaluating other risk factors, particularly systemic lupus erythematosus, need to be conducted to determine any relationship and how to best manage this condition in such patients.

**Keywords: Breast abscess, Systemic lupus erythematosus, Fungal infection, Mastitis.**

## 1. BACKGROUND

Systemic lupus erythematosus (SLE) is a chronic autoimmune disorder characterized by multisystemic involvement with various clinical manifestations (1, 2). The worldwide prevalence has a wide variation, ranging from 13 to 7,713.5 per 100,000 individuals (3). Fungal infections among SLE patients, in particular, have been reported previously as a complication following immunosuppressive therapy (4). However, in the absence of predisposing factors such as lactation and immunosuppressive medications, fungal infections, especially involving the breast, remain an extremely rare occurrence (5).

## 2. OBJECTIVE

The aim of this article is to report a case of recurrent non-lactational fungal infection on top of fibrocystic breast disease in an SLE patient who was not on any immunosuppressive medications.

## 3. CASE REPORT

A 39-year-old female, diagnosed with SLE nine years prior, was referred to our breast clinic as a case of recurrent right breast fungal infections manifesting in the form of abscesses that started four years ago. Upon presentation, she was having her fourth attack and was already started on fluconazole and responded to it with no active complaints. The patient had three previous episodes of breast fungal infections confirmed by multiple cultures

| Parameter | Value |
|---|---|
| Complete blood count | |
| White blood count | 4 k/uL |
| Hemoglobin | 9.3 g/dL |
| Platelet count | 231 k/uL |
| Coagulation profile | |
| Prothrombin time | 14.4 sec |
| Partial thromboplastin time | 39.8 sec |
| Inflammatory markers | |
| C-reactive protein | 0.26 mg/dL |
| Erythrocyte sedimentation rate | 50 mm/hr |
| Immunoglobulins & complement | |
| Immunoglobulin A | 320 mg/dL |
| Immunoglobulin G | 1629 mg/dL |
| Immunoglobulin M | 139 mg/dL |
| Immunoglobulin E | 202 kU/L |
| Autoantibodies | |
| Antinuclear antibody | >1280 (Positive) |
| Anti-double stranded DNA | 1:40 (Positive) |
| Anti-Ro/SSA | 70.5 (Positive) |
| Anti-La/SSB | <3.3 (Negative) |
| Anti-ribonucleoprotein | <3.5 (Negative) |
| Anti-Scl-70 | <1.2 (Negative) |
| Anti-Smith antibody | <3.3 (Negative) |
| Complement | |
| C3 | 108 |
| C4 | 18.7 |

**Table 1. Laboratory investigations obtained upon presentation.**

from the lesions, all were treated successfully with fluconazole. The patient had no milky, purulent, or bloody nipple discharge before. Interestingly, she did not complain nor exhibit symptoms or signs of SLE before apart from arthralgia and was not on any immunosuppressive medications to trigger the episodes. The activity of SLE was only managed with hydroxychloroquine 200 mg once daily. Moreover, she was not breastfeeding nor pregnant, and her past history was only significant for recurrent abortions. Local examination revealed no skin changes, tenderness, or presence of any palpable masses in the breast or axilla.

Laboratory investigations obtained upon presentation are summarized in Table 1, including relevant autoantibodies. Previous fine needle aspiration cultures revealed growth of Candida Albicans sensitive to fluconazole. Right breast ultrasound (US) performed on the first attack demonstrated the presence of a fluid filled collection measuring around 1.7 x 3.6 cm located at 1 o'clock, as shown in Figure 1. The lesions were noted to respond to fluconazole in this attack and subsequent ones.

Mammography of the right breast revealed a heterogeneously dense breast parenchyma, representing type C breast density according to American College of Radiology classification, as illustrated in Figure 2. Multiple well-circumscribed right inner breast masses, the largest measuring 10 x 9 mm, were also noted with no evidence of microcalcification. These lesions present at the right breast were hence labeled as Breast Imaging Reporting and Data System 4A; therefore, a histopathological diagnosis was required. US-guided core needle biopsy of the right breast mass revealed necrotizing granulomatous inflammation with presence of fungal hyphae and spores. Acid-fast bacilli stain was negative, and tissue aerobic culture showed no growth.

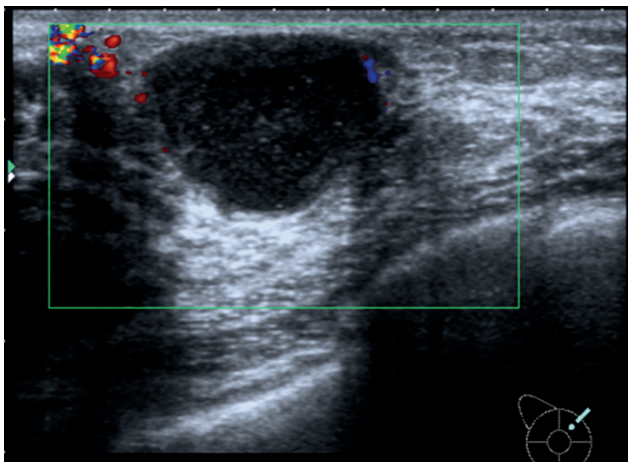

**Figure 1. Right breast ultrasound performed on the first episode demonstrating the presence of a fluid filled collection, measuring around 1.66 x3.56 cm located at 1 o'clock.**

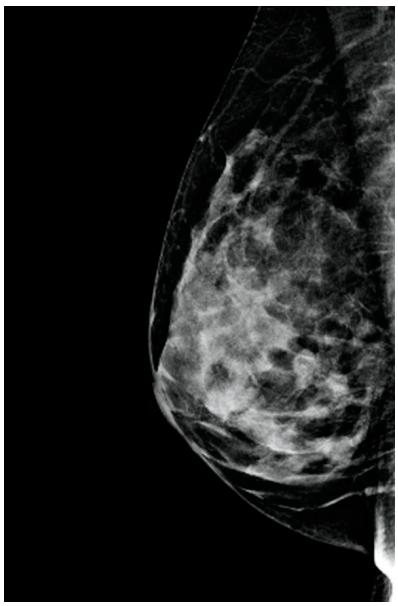

**Figure 2. Mammography of the right breast showing a heterogeneously dense breast parenchyma with multiple well-circumscribed inner breast masses.**

Considering the size of the lesion, a conservative approach was followed thereafter, utilizing antifungal alone to manage the patient's symptoms.

## 4. DISCUSSION

Mastitis is a benign condition of the breast recognized as a cluster of inflammatory changes of the mammary glands caused by either infectious or non-infectious factors, and it most commonly affects lactating women. Breast abscess, on the other hand, is a local collection of purulent material in the breast that is usually a complication of infectious mastitis (6). The main component in the pathophysiology of mastitis in lactating women

is milk stasis as it provides an ideal environment for organism replication (6-8). Breast infections in non-lactating women have not been sufficiently studied, however known causes include duct ectasia and tuberculous mastitis (6). One proposed cause of breast infections in non-lactating women is immunodeficiency in the context of diabetes mellitus (9). Non-infectious causes of mastitis include granulomatous mastitis and lupus mastitis (6, 10). Most cases of lactational mastitis can be managed with supportive therapy in the form of warm compressors and analgesia along with removal of milk from the affected breast. For non-lactational mastitis, supportive therapy along with an antistaphylococcal antibiotic is indicated (6).

Treatment of SLE mainly consists of nonsteroidal anti-inflammatory drugs, immunosuppressive or cytotoxic drugs and antimalarial agents (1, 2). One consequence of immunosuppressive therapy is the increased risk of infections (4). Nevertheless, SLE patients, in the absence of immunosuppressive agent use, are still at an increased risk of infections due to defects in the immune system induced by SLE (5). Well-documented, albeit rare, sources of infection following immunosuppressive therapy are fungal pathogens (4, 11-13). Immunosuppressive medications were found to predispose SLE patients to fungal infections, particularly those affecting the pharynx, esophagus, urinary tract, and soft tissues (4).

Both benign and malignant breast lesions have been reported in SLE patients. One rare manifestation of SLE is lupus mastitis, a condition characterized by inflammation of breast subcutaneous adipose tissue in patients diagnosed with SLE (14). The cause of lupus mastitis is still unknown; however, one theory suggests it is caused by the deposition of immunoglobulin M and C3 (15). Lupus mastitis may present as one or more breast masses, or it may only be seen as a mammographic abnormality, and they may also be firm and imitate malignancy clinically and radiographically (16). Malignant lesions of the breast such as carcinoma of the breasts and breast lymphoma have not been shown to be common in SLE patients and, in fact, those with SLE have been found to be at a lower risk of developing breast malignancies compared to the general population (17). The proposed reason for this decreased risk is the presence of multiple SLE autoantibodies leading to the possible exertion of an anti-cancer effect (18).

## 5. CONCLUSION

SLE and its treatment with immunosuppressive therapy, predispose patients to various complications including recurrent fungal infections. Mastitis in non-lactating women remains very rare. We report a patient with multiple episodes of non-lactational fungal mastitis while not on any immunosuppressive medications. No similar cases to ours have been published in English literature to the best of our knowledge. By reporting this case, we aim to increase awareness of how to approach similar patients with such a presentation. Additionally, we would like to highlight the need for more research in this area.


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- **Consent for publication**: Informed written consent was taken for the publication of this paper.
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