Idiopathic granulomatous mastitis refractory to radical surgical treatment in a patient with late diagnosis of systemic lupus erythematosus: a case report

Natalia Collazos Torres[1*], Ximena Briceño Morales[2,3], Sandra Milena González Rodríguez[2], Sara Marcela Guerrero Vallecilla[2], Verónica López Góngora[2], Ángela María García Sañudo[2], Clara Briceño Morales[4,5]

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ABSTRACT

Background: Idiopathic granulomatous mastitis (IGM) is a benign, chronic inflammatory disease of the breast that predominantly affects women of reproductive age. It is characterized by non-caseating granulomas involving the mammary lobules. Clinical manifestations range from breast pain and induration to cutaneous fistulas and breast deformity. In recurrent or extensive disease, management may require radical surgical approaches followed by reconstruction. We report the case of a patient who experienced an IGM relapse after salvage surgery in the remote donor area (abdomen), questioning the IGM diagnosis and raising other differential diagnoses.

Case Presentation: A 28-year-old woman presented with a 2-year history of multiple emergency department visits for recurrent mastitis, with clinical and histopathological pictures compatible with IGM, and a late diagnosis of systemic lupus erythematosus, who underwent bilateral mastectomy with reconstruction using a deep inferior epigastric perforator flap, and subsequently developed a recurrence of IGM in the non-native tissue.

Conclusion: This case highlights the aggressive clinical course that IGM may exhibit and underscores its potential for recurrence, even after radical surgical management and reconstruction. IGM that presents as a severe, refractory, and recurrent disease, and does not follow the usual clinical course, may be exacerbated by another concurrent illness. Further research is needed to clarify not only the origin of this condition but also the predictors of recurrence, and to propose effective therapeutic options.

Keywords: Granulomatous mastitis, mastectomy, systemic lupus, erythematosus, perforator flap, breast diseases, case report.

Type of Article: Case Report **Specialty:** Breast Surgery/Rheumatology

Correspondence to: Natalia Collazos Torres
*Hospital Infantil Universitario de San Jose, Fundación Universitaria de Ciencias de la Salud, Bogotá, Colombia
Email: natalita_ct7@hotmail.com
Full list of author information is available at the end of the article.

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Background

Idiopathic granulomatous mastitis (IGM) is a chronic, benign inflammatory disease of the breast, characterized by non-caseating granulomas in the mammary lobules. It primarily affects young women of reproductive age, often with a recent history of pregnancy or breastfeeding. Its cause remains unclear, but it has been linked to autoimmune conditions [e.g., systemic lupus erythematosus (SLE) or antiphospholipid antibody syndrome (APS)], hormonal disorders, and infectious processes, among others [1].

IGM often manifests with breast pain, erythema, thickening, cutaneous fistulas, and breast deformity, which has a significant impact on patients' body image [2]. There is no standard medical management because its origin is unknown, although systemic and local corticosteroids, immunomodulators such as methotrexate (MTX), various surgical treatments ranging from limited breast resections to mastectomy, and observation, have been used [1]. A recent meta-analysis shows relapse rates ranging from 4% to 65%, with higher percentages when surgical approaches such as isolated drainage (65%) or combined management with antibiotics and surgery (23%) are used [3].

When dealing with extensive or recurrent disease, radical surgeries combined with reconstructive techniques have been proposed, including mastectomy with autologous reconstruction using local or free flaps, which, in published series, is presented as a salvage treatment [4,5]. To date, no cases of recurrent IGM have been described following radical mastectomy with microvascular free flap reconstruction. In this context, we present what appears to be the first case of a young woman with recurrent disease

despite undergoing bilateral mastectomy with deep inferior epigastric perforator (DIEP) flap reconstruction, and discuss the potential implications of this finding.

Case Presentation

A 28-year-old female with a history of mild cognitive impairment, mixed anxiety and depressive disorder, nonspecific arthralgias, and a fetal loss 3 years ago (26 weeks), presented with a 2-year history of multiple emergency department visits for recurrent mastitis, treated with various antibiotic regimens (Figure 1). On her most recent visit, she was evaluated by breast surgery, which found bilateral inflammatory breast lesions in different stages, ranging from erythematous plaques to healed ulcers. The patient had two pathology reports, taken 2 months apart, which were consistent with IGM. Breast surgery recommended expectant management, nevertheless, due to her history and symptoms that suggested collagen disease, the patient was evaluated by the rheumatology service, which agreed with the diagnosis of IGM, ordered rheumatological studies, and based on the reports, determined that, at that time, she did not meet the criteria for SLE. Treatment was initiated with MTX × 2.5 mg tab (6 tab/week) peroral (PO), prednisolone 10 mg daily PO, folic acid 1 mg daily PO (except on the day of MTX), and calcium carbonate/vitamin D3 (600 mg/200 IU) daily PO. One month later, due to the progression of the breast lesions, the patient received intravenous methylprednisolone pulses for 3 days, and the prednisolone dose was increased from 10 to 30 mg daily PO. The rest of the medical management remained the same.

Within a period of 4 months, the patient experienced severe clinical deterioration, characterized by the appearance of new and multiple breast lesions, abscesses, and cutaneous fistulas. The patient was referred to a Multidisciplinary Medical Board. A bilateral simple mastectomy was agreed upon as the only possible treatment for the recalcitrant and recurrent IGM, which at that time had been ongoing for 3 years, carried out in August 2024. Four months later, the patient underwent delayed breast reconstruction using a DIEP flap (December 2024) (Table 1).

Approximately 4 months after breast reconstruction, and 8 months after bilateral mastectomy, the patient developed new lesions in the free flap from the abdomen, clinically identical to the initial breast lesions, and histopathologically similar to IGM (Figure 2). During the next 4 to 5 months, the patient's condition deteriorated, with the lesions coalescing and the formation of a large dermofat ulcer in the right breast (Figure 3). She was re-evaluated by the rheumatology service, who, based on EULAR/ACR 2019 criteria, made the diagnosis of SLE and APS. By that time, the patient was already showing positive results in the domains of these rheumatological diseases (Table 2). Medical management was adjusted to hydroxychloroquine 400 mg daily PO, MTX 20 mg SC weekly (she does not tolerate PO), prednisolone 20 mg daily PO, folic acid 1 mg daily (except on the day of MTX) PO,

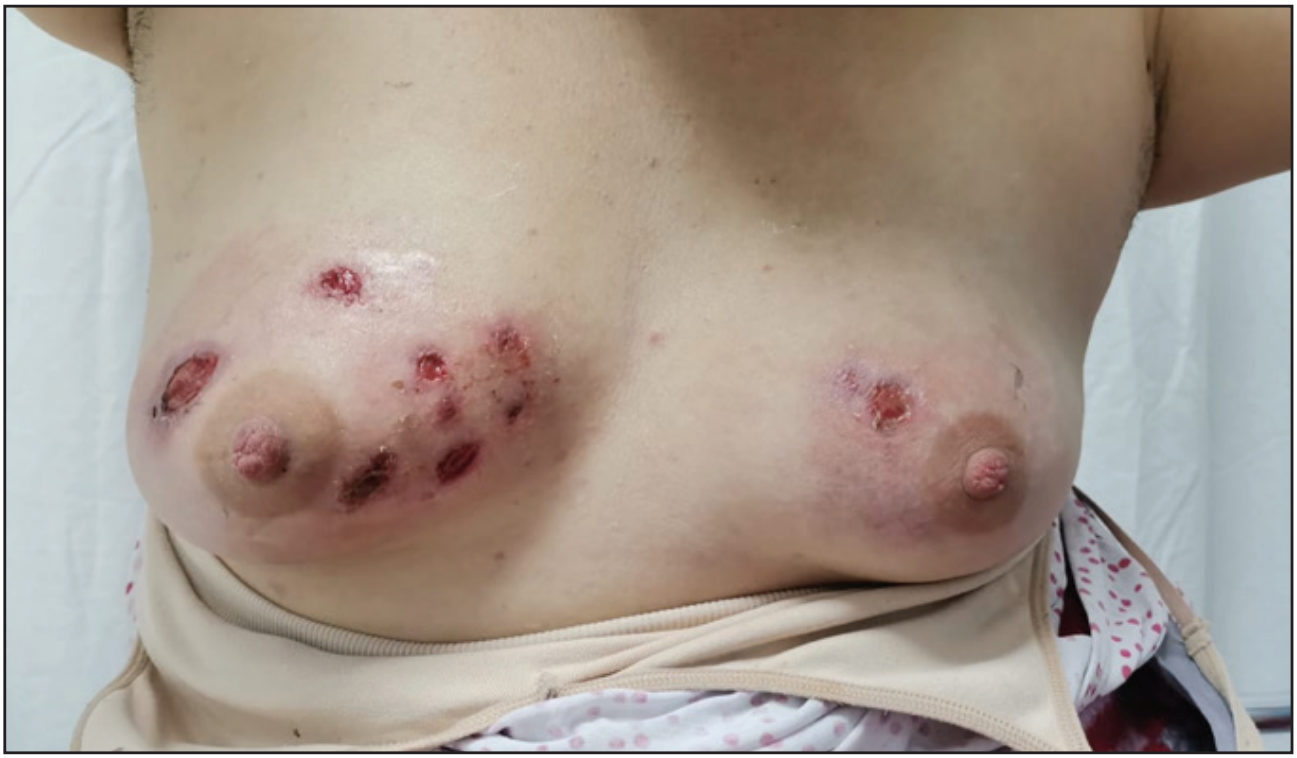

Figure 1. *March 2023. Ulcerated lesions in both breasts.*

Table 1. *Patient's clinical course, including biopsy and histopathological findings.*

INITIAL VISIT	
February 2023	Right breast mass with infectious inflammatory signs. Treatment: oxacillin + clindamycin
March 2023	Right breast biopsy: vacuoles containing clear material surrounded by lymphocytes and histiocytes, with areas of fibrosclerosis extending into the deep dermis, where breast tissue is identified with surrounding lymphoplasmacytic inflammation. Acute ulceration with foreign body inflammatory reaction. Chronic mastitis.
RETURN VISIT	
March 2023	**FOLLOW-UP CONSULTATION FOR ULCERATED LESIONS IN BOTH BREASTS.**
May 2023	Right breast biopsy: Breast tissue showing a granulomatous inflammatory infiltrate composed of lymphoplasmacytic cells with mild polymorphonuclear neutrophilic infiltration. No necrosis, no evidence of malignancy. Acute and chronic granulomatous mastitis.
FIRST BREAST SURGERY	
August 2024	Simple bilateral mastectomy (without reconstruction) Patology: Breast tissue showing fibrotic areas with the presence of dilated ducts, simple adenosis, usual ductal hyperplasia, columnar cell changes, and collagen bundles arranged parallel to the epidermis. In some areas, an inflammatory infiltrate composed of lymphocytes, histiocytes, and scattered neutrophils is identified. Mild chronic and acute inflammation, scar-related changes
SECOND BREAST SURGERY	
December 2024	Breast reconstruction with a DIEP flap
CLINICAL EXACERBATION	
April 2025	The patient developed new lesions in the free flap from the abdomen, identical in characteristics to the initial breast lesions
May 2025	Trucut biopsy: foreign body inflammatory reaction.
Return visit	
August 2025	Left breast skin biopsy: skin tissue showing ulceration with abundant neutrophils, cellular debris, and granulation tissue. Histologic sections demonstrate adipose tissue containing areas of liquefactive necrosis.
September 2025	Multiple punched-out ulcerated lesions with irregular borders, ranging from 5 mm to 4 cm in diameter, with raised borders and mild erythema, clean base, and no evidence of secretions. Biopsy: No cells with neoplastic appearance are identified. Fibroconnective tissue, fibrous stroma, and dense neutrophilic infiltrate are present. No findings compatible with pyoderma gangrenosum are observed.

ASA 100 mg daily PO, and calcium carbonate/vitamin D3 (600 mg/200 IU) daily PO.

In the postoperative period, the patient was unable to receive the medical treatment prescribed by rheumatology to treat her autoimmune diseases, due in part to low tolerance to oral medication, but mainly to the lack of coverage for some subcutaneous medications by her health insurance.

Over the last 9 months, the patient has been hospitalized on multiple occasions for episodes of bacteremia caused by at least two different microorganisms, *Burkholderia cepacia* and *Klebsiella pneumoniae*. She was treated with broad-spectrum antibiotic therapy, including meropenem and ceftazidime/avibactam; she also required multiple breast lavages and surgical debridements. The pathology report from the last surgical procedure, in August 2025, is compatible with "ulceration, granulation tissue, and liquefactive necrosis." Both special stains for acid-fast bacilli and fungal structures were negative, and malignancy was ruled out, too.

In the patient's last known clinical record, she was hospitalized without receiving antibiotic therapy, while awaiting transfer to a higher-complexity referral center for continued comprehensive management in conjunction with the Plastic Surgery team.

Discussion

The reported patient presents a long-standing, severe, highly insidious clinical picture that is refractory to conventional, radical, and rescue surgical management.

The initial histopathological diagnosis of this patient, confirmed by biopsy in March of 2023, was IGM. Among the factors historically associated with IGM recurrence, the study published in 2018 by Uysal et al. [6], which included 720 patients diagnosed with IGM in 22 Breast Units in Turkey between 2011 and 2016, found a higher risk of recurrence ($p < 0.05$) in women with a history of pregnancy and lactation, in those who smoked, and in those with previous infections, both local and systemic [6]. More recently (2025), a study described younger age, surgical treatment, and nipple discharge and inversion as independent risk factors for recurrence ($p < 0.05$); the authors created a nomogram based on these factors that demonstrated a good positive predictive value for the recurrence of IGM, with a sensitivity of 81.7% and a specificity of 66.7% [7].

The described patient is young, has a history of pregnancy and bacteremia, all risk factors for recurrence of IGM, widely described in the literature. Even so, by clinic (Figure 2), our case suggests an atypical presentation of IGM recurrence after radical surgical management with bilateral mastectomy and DIEP reconstruction, a situation that has not been previously reported, particularly due to the recurrence of lesions in non-native breast tissue, such as free abdominal flaps; however, the microscopic description of the histopathological analysis, while reporting "vacuolated histiocytes and multinucleated giant cells, compatible with a foreign body granulomatous reaction", did not show "evidence of ductal epithelium", which would be expected given the patient's history of mastectomy. It is important to highlight that a study conducted by Lermi et al. [8], which included 120 patients from a hospital in Turkey between 2010 and 2022, found that those patients, who underwent surgery as initial treatment had a recurrence rate of 66.1%, which calls into question a surgical approach as a treatment option, at least as a first-line treatment, especially in patients with risk factors for relapse.

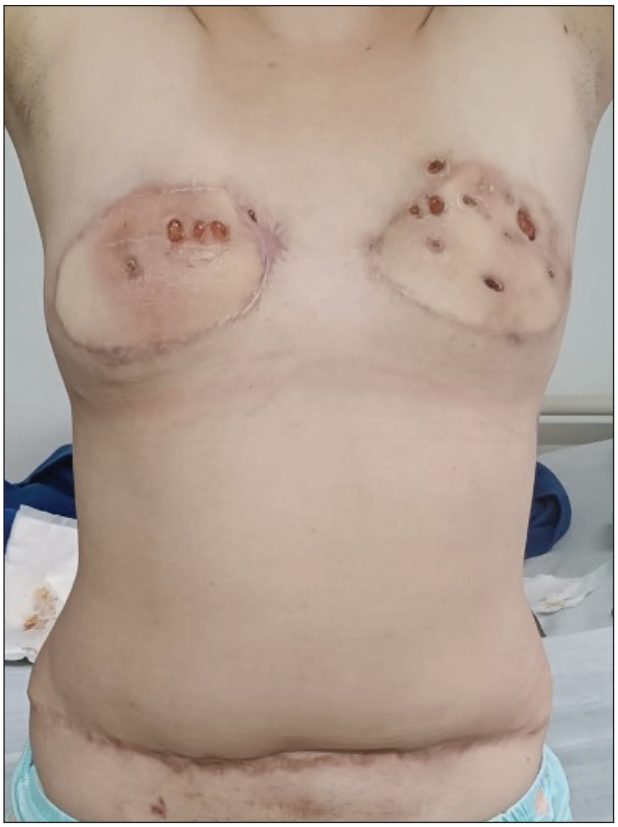

Figure 2. *April 2025. New lesions in the free flap from the abdomen, identical in characteristics to the initial breast lesions, four months after breast reconstruction.*

It is necessary to emphasize the importance of performing an appropriate differential diagnosis when the clinical picture is torpid and recalcitrant, as in our patient's case. Among the differential diagnoses is tuberculous mastitis, which can be indistinguishable in imaging and clinical studies, making histopathology crucial. Other differential diagnoses include breast carcinoma, sarcoidosis, and Wegener's granulomatosis [9]. It is worth considering whether lupus panniculitis (LP) could be a differential diagnosis in the context of the patient's autoimmunity, given its extensive involvement associated with inflammation and relapse at the level of the free abdominal flaps; however, histopathological analysis is also indispensable here, because it shows a mixed panniculitis of the

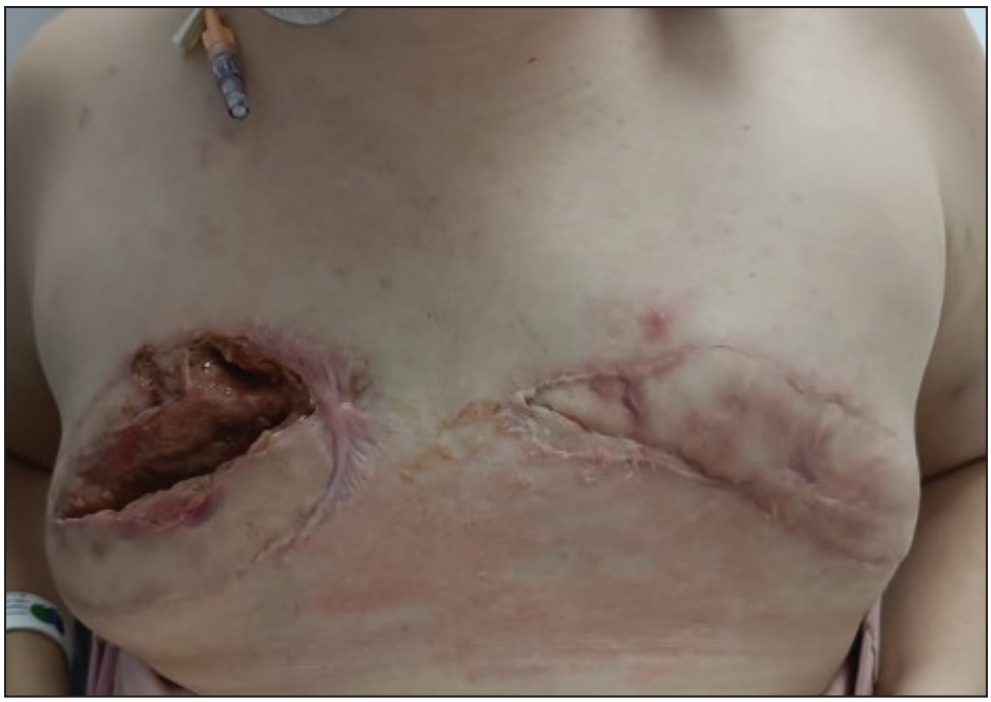

Figure 2. *Between August and September 2025, the patient initially presented with multiple punched-out ulcerative lesions that subsequently coalesced and progressed into a large ulcer involving the skin and subcutaneous tissue.*

Table 2. Rheumatologic disease course and diagnostic workup.

DATE	JANUARY 2023	APRIL 2024	SEPTEMBER 2024	AUGUST 2025	SEPTEMBER 2025
Relevant clinic	Nonspecific arthralgias and a fetal loss	Iron deficiency anemia, recurrent granulomatous mastitis, septic tenosynovitis (Koebner phenomenon?)	Recurrent granulomatous mastitis, hepatomegaly on ultrasound	Appearance of skin lesions on the free abdominal flap of the breast reconstruction, of four months' evolution (April, 2025)	Coalescence, ulceration of skin lesions and formation of a large dermofat ulcer in the right breast
Laboratories	ACL IgM and IgG: negatives	ANAs: negative Anti SSB/La: negative Anti SM: negative Anti RNP: negative Anti SSA/Ro: negative Lupus anticoagulant: positive (not confirmed by Russell's viper venom)	ANAs by IFA: 1/80 titer, AC-4 dense fine speckled patter	*SLE diagnosis: ANAs positive: 1/80, AC-4 fine speckled nuclear pattern; cutaneous domain: malar erythema and diffuse alopecia; articular domain: metacarpophalangeal and proximal interphalangeal polyarthralgia; hematologic domain: complement C4: 8.24 g/l; immunologic domain: Anti-DNA positive: 1/80	*APS diagnosis: Obstetric domain: unexplained fetal death after second trimester; triple-positive immunological domain: IgG ACL: 22.7 U/ml, IgG AB2G: 61 U/ml, AL VVR: 2.2 (ratio)

ACL, anticardiolipin antibodies; ANAs, antinuclear antibodies; Anti SM, anti-Smith antibodies; Anti-RNP, anti-ribonucleoprotein antibodies; Anti-SSA, anti-Sjögren's Syndrome-related antigen A antibodies; Anti-DNA, anti-double-stranded DNA antibodies; AB2G, beta-2 Glycoprotein I antibodies; AL VVR, lupus anticoagulant in Russell's viper venom.
*Based on EULAR/ACR 2019 criteria.

subcutaneous cellular tissue with thickening, fibrosis and lymphoplasmacytic infiltrate, with hyaline necrosis of the fat and lymphoid nodules, associated with lymphocytic vasculitis and dermoepidermal calcification; in addition, when the histopathological findings of LP are equivocal, direct immunofluorescence can help to confirm the diagnosis, demonstrating a deposit of IgG, IgM and C3 in 70%-80% of cases [10]. In our case, we do not have a biopsy that suggests LP.

In the present case, recurrent IGM remains the leading diagnostic consideration based on the overall clinicopathological correlation. The lesions that developed in the DIEP flap were clinically indistinguishable from the patient's original breast lesions, showing recurrent inflammatory nodules, ulceration, and progressive tissue destruction. Furthermore, the patient had a history of multiple breast biopsies over more than 2 years, demonstrating findings compatible with granulomatous mastitis. Repeated pathological evaluations excluded malignancy, while special stains for acid-fast bacilli and fungal organisms were negative. Although the patient subsequently developed episodes of bacteremia, the persistence and progression of the lesions were not entirely explained by infection alone.

Nevertheless, alternative diagnoses were carefully considered. Histopathological examination did not demonstrate the characteristic features of LP, such as hyaline fat necrosis, dense lymphocytic lobular panniculitis, lymphoid follicles, or calcification. Similarly, pathological studies did not reveal findings suggestive of pyoderma gangrenosum, vasculitis, or a specific infectious process. We acknowledge that one biopsy from the reconstructed breast was interpreted as a foreign body granulomatous reaction, and that the absence of residual breast tissue in the specimen limits definitive confirmation of recurrent IGM. Therefore, while recurrent IGM appears to be the most plausible explanation for this unusual presentation, LP, lupus mastitis, foreign body reaction, and other autoimmune inflammatory disorders cannot be completely excluded.

Unfortunately, the patient resides in a developing country with limited resources and poor access to healthcare. Due to her socioeconomic status, it is currently not possible to evaluate her response to rheumatological treatment, for example, or to conduct long-term follow-up. However, the objective of presenting this unusual and insidious clinical case is precisely to question the diagnosis of IGM, even in cases of multiple biopsies that supposedly confirm it (timeline), when it occurs in a patient with a decompensated rheumatological disease. In such cases, other differential diagnoses arise, and these should at least be considered as possible causes, especially in a scenario where the patient's condition is not progressing as expected.

Conclusion

In exceptional circumstances, IGM can present as a severe, recurrent condition refractory to medical and radical/rescue surgical treatments. However, when it does not follow an expected clinical course, the diagnosis of IGM should be questioned, and other differential diagnoses should be considered, but adequately confirmed. In patients such as the one reported, future research should focus on identifying the specific biological mechanisms that cause the

disease, as well as those that predispose to recurrence, in order to eventually find a therapeutic target. Finally, to the authors' knowledge, this appears to represent the first documented case of a patient with recurrent IGM in distant abdominal flaps, following mastectomy and reconstruction with a DIEP flap, in a patient with a late diagnosis of SLE.

What is new?

This is the first reported case of granulomatous mastitis recurring after mastectomy with DIEP reconstruction, involving non-native tissue. It highlights the need to reconsider diagnosis in refractory cases and evaluate underlying autoimmune diseases such as SLE.

List of Abbreviations

APS	Antiphospholipid syndrome
DIEP	Deep inferior epigastric perforator
IGM	Idiopathic granulomatous mastitis
LP	Lupus panniculitis
MTX	Methotrexate
SLE	Systemic lupus erythematosus

Conflicts of interest

The authors declare that they have no conflicts of interest regarding the publication of this case report.

Funding

Nothing to declare.

Consent for publication

Written informed consent was obtained from the patient for the publication of clinical data and images.

Ethical approval

This clinical research adheres to the principles established in the Good Clinical Practice guidelines of the International Committee for Harmonisation and the ethical principles of the Declaration of Helsinki. It also follows the CIOMS guidelines and Resolution 008430 of October 4, 1993, from the Ministry of Health and Social Protection of the Republic of Colombia.

Author details

Natalia Collazos Torres[1], Ximena Briceño Morales[2,3], Sandra Milena González Rodríguez[2], Sara Marcela Guerrero Vallecilla[2], Verónica López Góngora[2], Ángela María García Sañudo[2], Clara Briceño Morales[4,5]

1. Hospital Infantil Universitario de San José, Fundación Universitaria de Ciencias de la Salud, Bogotá, Colombia
2. Departamento de Cirugía, Facultad de Medicina, Universidad Nacional de Colombia—Sede Bogotá, Bogotá, Colombia
3. Instituto Nacional de Cancerología, Bogotá, Colombia
4. Universidad Tecnológica de Pereira, Grupo de Investigación en Ciencias Quirúrgicas, Pereira, Colombia
5. Institución Universitaria Visión de las Américas, Pereira, Colombia

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Summary of the case

1	Patient (gender, age)	28 years, female
2	Final diagnosis	IGM with SLE and APS
3	Symptoms	Recurrent mastitis, abscesses, fistulas
4	Medications	MTX, corticosteroids, hydroxychloroquine, ASA
5	Clinical procedure	Bilateral mastectomy + DIEP flap reconstruction
6	Specialty	Breast surgery/Rheumatology