Case Report

Pyoderma gangrenosum after reduction mammoplasty: diagnostic and therapeutic challenges

Pioderma gangrenoso após mamoplastia redutora: desafios diagnóstico e terapêutico

IGOR NAGAI YAMAKI [1]*
CELSO JANDRE BOECHAT [1]
RODRIGO CARLLETI RIZZO [1]
STELLA SOUZA AMORIM [1]
GABRIELA FIGUEIRA ANDRADE [1]
ANNALU PEDROSA REIS [1]

Institution: Hospital Municipal Barata Ribeiro, Rio de Janeiro, RJ, Brazil.

Article received: January 26, 2019.
Article accepted: June 22, 2019.

Conflicts of interest: none.

DOI: 10.5935/2177-1235.2019RBCP0240

■ **ABSTRACT**

Pyoderma gangrenosum (PG) is an inflammatory disease of the skin that may develop spontaneously. It is associated with certain systemic and neoplastic diseases, including those of the breasts. PG is also associated with surgical trauma. It has been increasingly reported, along with the increase in the incidence of reduction mammoplasty procedures. The clinical manifestation of ulcers is characteristic of PG and it should be considered in cases of poor healing with intense inflammatory reaction, tissue loss, bloody and/or purulent secretion, granular background, and lesions with high edges. We describe a patient who developed PG after reduction mammoplasty. She has since responded to systemic corticosteroids and has had no relapse to date.

Keywords: Pyoderma gangrenosum; Mammoplasty; Postoperative complications; Autoimmune diseases; Differential diagnosis.

[1] Hospital Municipal Barata Ribeiro, Rio de Janeiro, RJ, Brazil.

■ **RESUMO**

O pioderma gangrenoso (PG) é doença inflamatória da pele, que pode se desenvolver espontaneamente, associado a certas doenças sistêmicas e neoplásicas, ou ao trauma cirúrgico, incluindo os das mamas. Há relatos cada vez mais frequentes, considerando o aumento desse procedimento nos dias atuais. A manifestação clínica das úlceras é característica e deve ser lembrada nas evoluções cicatriciais desfavoráveis com intensa reação inflamatória, perdas teciduais, secreção sanguinolenta e/ou purulenta, fundo granuloso e bordas elevadas. Relata-se o caso de paciente que teve pioderma gangrenoso após mamoplastia redutora. Respondeu ao corticosteroide sistêmico, e vem evoluindo sem recidivas até o momento.

Descritores: Pioderma gangrenoso; Mamoplastia; Complicações pós-operatórias; Doenças autoimunes; Diagnóstico diferencial.

INTRODUCTION

Pyoderma gangrenosum is a rare, recurrent, and destructive skin disease of unknown etiology with a predominance in females aged 25–55 years[1]. The diagnosis is based on the evolution of the disease and the exclusion of other diseases. It is triggered by trauma (pathergy phenomenon) in up to 25% of the cases [2,3,4].

Although it manifests in various clinical forms, we will concentrate on the presentation of pyoderma gangrenosum characterized by the appearance of ulcers after breast surgery in this report. The recognition of its' lesions, predisposing factors, and surgical risks, as well as the early administration of appropriate treatment can mitigate the extremely serious outcomes for affected patients.

CASE REPORT

A 50-year-old healthy Caucasian female patient was admitted to the Plastic Surgery service of Barata Ribeiro Municipal Hospital, Rio de Janeiro for reduction mammoplasty surgery.

Initial examination revealed large dense breasts with grade II ptosis, which induced postural discomfort and back pain. The patient underwent a surgical procedure using the Wise technique with type I Liacyr Ribeiro pedicle. She had no untoward events during hospitalization and was discharged on the second postoperative day.

On the seventh postoperative day, the patient was referred for an examination, and the condition of the breasts was good. A small degree of edema and small areas of hyperemia were observed. She reported having mild pain, and pain relievers were prescribed.

From the seventh to the tenth postoperative day, the patient experienced a significantly worsening clinical picture. She complained of intense pain and presented with ulcerated and confluent lesions in the lower poles of both breasts. A cranial progression of the lesions was observed over several days, preserving only the nipple-areola complex (Figure 1). The lesions had elevated edges and an exudative fundus with fibrin and secretion with blood and pus, reminiscent of a soft tissue infection.

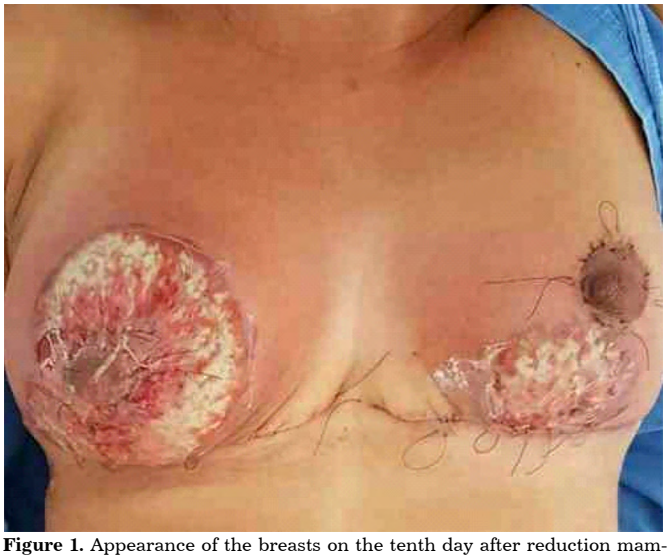

Figure 1. Appearance of the breasts on the tenth day after reduction mammoplasty.

The patient was readmitted to hospital and samples were collected for examination, including swab and culture. Antibiotic therapy with cephalexin was initiated. Subsequently, 800 mg/day of ciprofloxacin and 3 g/day Clavulin were added to the regimen to control the probable infection.

Laboratory results revealed that the patient had elevated systemic markers for inflammation, (erythrocyte sedimentation rate and reactive C protein, without leukocytosis).

Given the unsatisfactory results of antibiotic therapy and the negative results of the cultures of both breasts, the diagnosis of pyoderma gangrenosum was determined and corticoid therapy with prednisone 100 mg/day and local skin care was initiated.

Dressings were applied daily to remove excess exudative content, using 0.9% saline solution and polyhexanide aqueous solution, followed by coverage with calcium alginate. The objectives were to keep the wound as clean as possible, without increasing the trauma at the site and thus avoid the progression of the lesions. The use of chemical debriders containing collagenase was prescribed, and surgical debridement was not performed. The option of grafting was also rejected at this time considering the possibility of triggering a recurrence.

Within a few weeks of beginning corticoid therapy, the ulcers regressed substantially. With the reduction of the lesions, resolution of edge involvement and healing progression, the corticoid therapy was slowly reduced to a maintenance dose of 10 mg/day. The less purulent and superficial wounds were covered with non-adherent cellulose acetate mesh.

DISCUSSION

Pyoderma gangrenosum is a disease of unknown etiopathogenesis, and in more than 50% of the cases, it is associated with systemic diseases[3,4,5]. While some lesions spontaneously appear, other manifestations appear only after surgical trauma, which is frequently reported after breast surgery. It is mainly characterized by the presence of a pimple with a necrotic center surrounded by bluish-red tissue. This quickly evolves into an ulcer whose cultures are negative. The condition usually evolves from 5 days to 10 years and requires continuous monitoring of the clinical presentation of the skin ulcers to exclude infectious, autoimmune and neoplastic diseases[6].

The patient in this report initially had multiple pustule lesions, which spread rapidly through the breasts, generating an exudate containing blood and pus. There was rapid dehiscence of the suture with elimination of the threads, intense pain, and perilesional growth with erythematous-violaceous edges simulating an infectious condition. The preservation of the nipple-areola complex[7,8], lack of response to antimicrobials, and negative lab cultures were the determining factors for the diagnosis of pyoderma gangrenosum.

Laboratory findings are neither specific nor diagnostic, and rates of erythrocyte sedimentation, C-reactive protein levels, and leukocytosis are always high[9]. Histopathology may also aid in the exclusion of other causes of skin ulcers, commonly revealing sterile neutrophilia associated with lymphocytic vasculitis[10].

Because there is no specific etiology of the pyoderma, the treatment varies widely, being based, in most cases, on an immunosuppressive agent[10]. Prednisone (60 mg/day to 80 mg/day) is the initial treatment of choice. There are several alternatives for cases that do not respond to corticosteroids. Thus, clofazimine, sulfasalazine, cyclophosphamide, mofetil mycophenolate, thalidomide, rifampicin, dapsone, gamma globulin, plasmapheresis, infliximab, tacrolimus, and methotrexate, among others, are prescribed [6,9]. In the case described, a significant improvement was obtained with the administration of prednisone 100 mg/day alone (Figure 2), with subsequent gradual weaning.

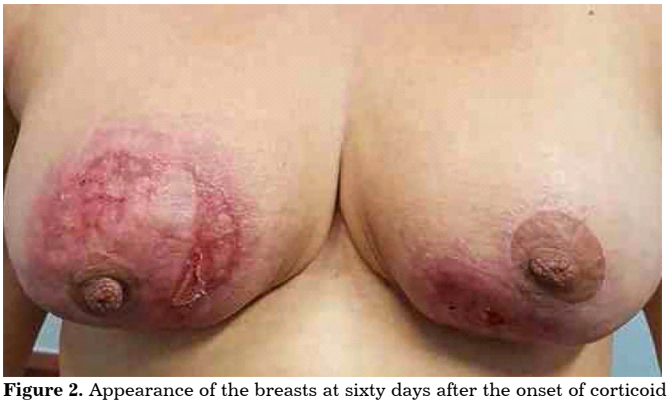

Figure 2. Appearance of the breasts at sixty days after the onset of corticoid therapy.

Potassium permanganate, povidone iodine, silver nitrate, topical rifampicin, intralesional triamcinolone, benzyl peroxide, 0.9% saline solution, hydrocolloids, calcium alginate, and the use of a hyperbaric chamber, among other therapies are reported for administering local skin care. In this context, it is important to remember that debridement should be postponed until the histopathological diagnosis of the active ulcerated area is obtained. In most cases, the patient's condition worsens significantly after surgical procedures. Thus, chemical and surgical debridement and grafting should be avoided when pyoderma gangrenosum is suspected[9]. When grafting is necessary, the administration of corticosteroids before the procedure may minimize the development of the disease.

In situations where the diagnosis, prognosis and clinical presentation are uncertain, the patient should always receive psychological support. The patient's commitment to maintaining long-term treatment with immunosuppressants is fundamental. Another dilemma to be faced is the repair of the scarring sequelae, which in some cases may generate new tissue aggressions and recurrence of the condition.

In conclusion, pyoderma gangrenosum is a rare skin disease with unpredictable development and course. Suspecting the diagnosis of pyoderma gangrenosum is extremely important, as the onset of the treatment will determine the outcome[10]. The use of corticosteroids and/or immunosuppressants should be the first measure to control the progression of the lesions and to minimize final scarring (Figure 3). This should be associated with careful local cleansing in order to remove the exudative content without increasing tissue aggravation. Thus, chemical or surgical debridement should be avoided in this condition.

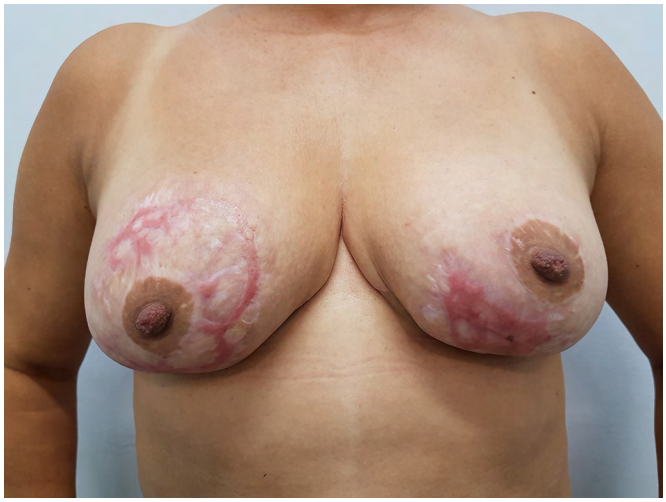

Figure 3. Appearance of the breasts at one year after reduction mammoplasty.

COLLABORATIONS

INY Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing

CJB Final manuscript approval, Project Administration, Supervision, Visualization, Writing - Review & Editing

RCR Data Curation, Project Administration, Supervision, Validation, Visualization, Writing - Review & Editing

SSA Data Curation, Realization of operations and/or trials, Visualization, Writing - Review & Editing

GFA Realization of operations and/or trials, Supervision, Visualization

APR Realization of operations and/or trials, Supervision, Visualization, Writing - Review & Editing

REFERENCES

1. Bennet ML, Jackson JM, Jorizzo JL, Fleischer Junior AB, White WL, Callen JP. Pyoderma gangrenosum. A comparison of typical and atypical forms with an emphasis on time to remission. Case review of 86 patients from 2 institutions. Medicine. 2000 Jan;79(1):37-46.
2. Vasili E, Shkodrani E, Labinoti L, Xhaja A. A case of atypical pyoderma gangrenosum. J Dermatol Case Rep. 2010 Apr;4(1):18-21.
3. Ravic-Nikolic A, Milicic V, Ristic G, Jovovic-Dagovic B. Pyoderma gangrenosum associated with Sjögren syndrome. Eur J Dermatol. 2009 Jul/Aug;19(4):392-3.
4. Yamanaka K, Kuniyuki S, Maekawa N, Yoshida Y, Teshima H. Pyoderma gangrenosum with myelodysplastic syndrome treated with analogous bone marrow transplantation. Acta Derm Venereol. 2009;89(1):105-6.
5. Santos M, Rabelo RF, Chirano CA, Talhari C, Schetini APM, Talhari S. Pioderma Gangrenoso: apresentação clínica de difícil diagnóstico. An Bras Dermatol. 2011;86(1):153-6.
6. Konopka CL, Padulla GA, Ortiz MP, Beck AK, Bitencourt MR, Dalcin DC. Pioderma gangrenoso: Um artigo de revisão. J Vasc Bras. 2013 Mar;12(1):25-33.
7. Tuffaha SH, Sarhane KA, Mundinger GS, Broyles JM, Azoury SC, Seal S, et al. Pyoderma Gangrenosum After Breast Surgery: Diagnostic Pearls and Treatment Recommendations Based on a Systematic Literature Review. Ann Plast Surg. 2016 Aug;77(2);e39-44.
8. Busato WMM, Pontes LT, Velho PENF, Magalhães RF. Pioderma gangrenoso da mama – relato de caso e aspectos relevantes para o diagnóstico precoce. Diagn Tratamento. 2016;21(2):65-9.
9. Portinho CP, Miguel LMC, Morello ER, Herter AHR, Collares MVM. Tratamento de pioderma gangrenoso após mamoplastia reutora com imunossupressão e oxigenoterapia hiperbárica: relato de caso e revisão de literatura. Arq Catarin Med. 2014;43(Supl 1):17-19.
10. Ahronowitz I, Harp J, Shinkai K. Etiology and management of pyoderma gangrenosum: a comprehensive review. Am J Clin Dermatol. 2012 Jun;13(3):191-211.

***Corresponding author:** **Igor Nagai Yamaki**
Rua Carlos Góis, 390, Leblon, RJ, Brazil.
Zip code: 22440-040
E-mail: igoryamaki@gmail.com