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Pyoderma Gangrenosum Following Breast Reconstruction: Successful Treatment with Steroids

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ABSTRACT

Introduction: Pyoderma gangrenosum is arare cause of skin necrosis after surgery. It often appears with extensive ulcerations and scaring with significant aesthetic aftermaths. The pathogenesis is poorly understood but often associated with inflammatory diseases, however post-surgical idiopathic pyoderma gangrenosum can occur. **Case Report:** We report a case of pyoderma gangrenosum following a bilateral breast reconstruction in a 33-year-old woman, successfully treated with steroids. A week after the surgery wounds started to break down and extensive necrotic ulceration appeared quickly covering both breasts. After dermatopathology evaluation she received antibiotics and prednisone and three months later wounds were completely healed and surgery was not necessary.

Key words: Pyoderma gangrenosum, Breast reconstruction, Steroids.

INTRODUCTION

Pyoderma gangrenosum (PG) is an infrequent ulcerative skin disease.[1] PG is characterized by a quickly progressive necrosis. It is usually a manifestation of underlying systemic disease. PG is also known as Cullen´s postoperative progressive gangrene (PPG). Several cases have been published,[2] but only some of them developed after breast surgery.[3,4]

Our aim is to update the management of *Pyoderma gangrenosum*, reviewing its diagnosis, treatment and prognosis, apropos of a case and according to current literature.

MATERIALS AND METHODS

We report a case of pyoderma gangrenosum following breast reconstruction which was successfully treated with steroids. The etiology and therapy are discussed considering the current literature. Literature review was made searching in PubMed (http://www.ncbi.nlm.nih.gov/pubmed) with the terms "Pyoderma gangrenosum", "breast reconstruction" and "steroids".

RESULTS

We report a case of a 33-year-old Caucasian woman, carrying a BRCA-2 gene mutation, who quickly developed an inflammatory-infectious process on the right breast, following bilateral mastectomies with breast reconstruction in November 2014. Rokitansky syndrome treated surgically at 19-years-old. She was in good health with no history of systemic illness or autoimmune disease.

The surgery consisted of both subcutaneous mastectomies with preservation of nipple-areola-complex and immediate reconstruction (in the same surgical procedure) by bilateral sub-pectoral breast implant. The immediate postoperative period was going well. A week later she complained of pain from the wound on both breasts and lymphadenopathies on the right armpit, but was not valuable. The following week skin lesions appeared on both breasts with extensive necrotic ulceration rapidly covering the right breast. The inflammation increased and the scars started to break down (Figure 1). With clinical suspicion of PG or an infection, assessment was con-

ducted by dermatology, which took a sample for biopsy and microbiology.

We got normal results from the blood count, coagulation, biochemistry, serology, immune study, calprotectin and serum protein. Biopsy from the breast ulceration showed an intensive mixed acute inflammatory infiltrate, mainly neutrophils, with granulation tissue and granulomatous reaction, vessel destruction and necrosis of the epithelium (Figure 2). The injury associated super infection and foreign body reaction.

Dermatopathological and pathological evaluations revealed PG. The patient was appropriately treated with a high-dose of corticosteroids (prednisone 50 mg/daily orally) and antibiotics (doxycycline 100 mg/daily 14 days), continuing with local cures. *Pseudomonas aeruginosa* was detected in culture and treated with ciprofloxacin 500mg / 12 hrs orally for 7 days.

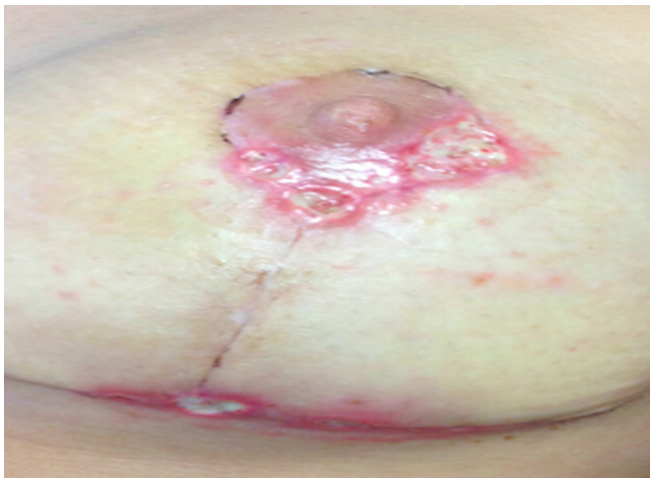

Figure 1: Appearance of the right breast after reconstruction at the time of the diagnosis of Pyoderma gangrenosum. Extensive necrotic ulceration, inflammation and wound dehiscence along scars and nipple areola complex.

Within a week of starting corticosteroids slight improvement of ulcers was observed. By the second week there was a clear improvement in the area of inflammation and some reepithelialisation of the skin with granulation tissue and signs of healing.

The same pattern of corticosteroids was maintained for a month, and then decreased weekly maintaining 5 mg/day on alternate days until the disappearance of the lesions. At the same time, she was treated conservatively with local measures, proteolytic agents to remove the necrotic tissue.

Two months later, the ulcers and inflammation had disappeared. At 3 months they were completely healed (Figure 3). Finally, an aesthetic intervention or breast implant removal was not necessary.

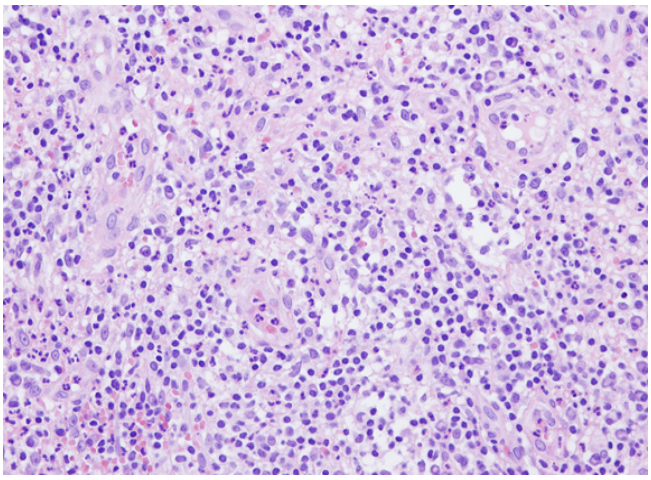

Figure 2: Breast skin biopsy (Hematoxylin and Eosin stain (H&E) 40x).Intensive mixed acute inflammatory infiltrate, mainly neutrophils with granulation tissue and granulomatous reaction, vessel destruction and necrosis of the epithelium.

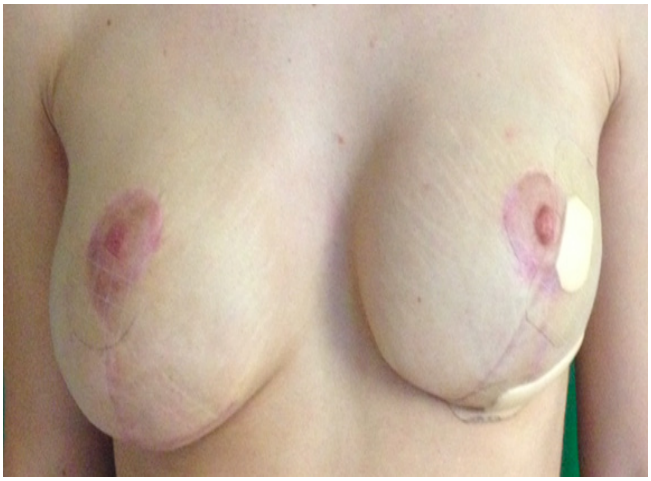

Figure 3: Three months after starting steroids treatment wounds were completely healed with a really good aesthetic result.

DISCUSSION

Pyoderma gangrenosum represents a rare dermatosis[5] that quickly evolves causing severe disruption of wound healing following surgical management.[6] The legs are the most common location of PG, but the disease may also involve other regions[7,8] such as the breasts.[9] Even in typical cases PG is often misdiagnosed or not readily recognized, therefore wrongly treated.

PG starts as an erythematous area that breaks down to form a painless skin irregular and deep ulcer with necrosis and hemorrhagic background. The ulcers may grow fast and result in extensive skin damage; although in rare cases they may remain unchanged for months.[7]

PG is idiopathic but can occur post-surgically.[3] The unknown aetiology and poorly understood pathogenesis often makes it difficult to detect and treat.[10] For about 50% of patients PG is usually a manifestation of an underlying systemic disease,[1] associated with monoclonal gammapathy, inflammatory bowel disease, myeloproliferative disorders, rheumatologic diseases and paraneoplastic syndrome. However post-surgical idiopathic PG can occur in healthy patients.[11] Our patient had an incident with intestinal discomfort years before, bloating and altered bowel habit, compatible with intestinal inflammatory process, but this could not be assured after digestive evaluation.

The pathogenesis remains unclear since there are cases without a preceding surgical intervention associated with a immunological disorder.[9] In most reported cases, the injuries were related to surgical interventions, as the result of *"a pathergy phenomenon"*. A common hapten (immunogenic molecule) between tumour and skin may explain the origin of this inflammatory disease.[5] Biopsies show chronic inflammatory infiltrate and skin necrosis but there was no subcutaneous damage, except in advanced disease.[4] Another theory is based on neutrophil and monocyte dysfunctions (phagocytosis or chemotaxis impaired function).[8]

PG is diagnosed after other possibilities are ruled out, but it can be suspected by clinical appearance and the lack of response to standard initial care.[12] The main differential diagnoses were skin and soft tissue infections including necrotizing fasciitis, Sweet´s syndrome and malignant neoplasms.[13] PG can be difficult to distinguish from infection[1] but PG is a neutrophilicdermatosis with areas of thrombosis, haemorrhage, necrosis or re-epithelisation and absence of inflammatory lymphangitis or lymphadenopathy.[10] Pseudomonas infection detected in our case was considered secondary infection from wounds. Sweet's syndrome is a neutrophilicdermatosis usually associated with autoimmune disorders and malignant hematological neoplasm.[8]

Often, multiple therapies have been employed without success. The treatment used for PG includes systemic corticosteroids, azathioprine, mercaptopurine, sulphasalazine, sulphapyridine,[9] cyclophosphamidem,[13] isotretinoin, immunoglobulins and cyclosporine.[2] The recommended first-line therapy for PG are decreasing dose corticosteroids, between 20 to 30mg/d orally for 3 to 4 months and, if necessary, associated with other immunosuppressive drugs.[8] In our case the associated treatment was a systemic low-dose of corticosteroid therapy with local wound care. Our patient reacted well to systemic steroids and the ulcer completely healed after 3 months. Surgery or radiotherapy are contraindicated, injuries appear again,[8] but Baruch *et al* preferred surgical excision and systemic corticosteroids.[11,13] Also, it should be remembered that treatment includes the management of the underlying disease if required.

Pyoderma gangrenosum has a recurrent nature and may be reactivated by various causes within a period of several years. Duchnowska *et al*[8] reported a case about of recurring PG as amanifestation of locally advanced breast cancer by exacerbation of rheumatoid arthritis. Long *et al*[14] suggested that avoiding epidermal sutures would prevent precipitation of PG at suture sites.

In conclusion, post-surgical idiopathic PG is very rare, but can occur after breast surgery. Early diagnosis and therapy are important to prevent serious clinical consequences. The recommended first-line therapy is medical management with local wound care and antibiotics.

CONCLUSION

Conclusion is previous and last written parragraph. see above. In conclusion, post-surgical idiopathic PG is very rare, but can occur a er breast surgery. Early diagnosis and therapy are important to prevent serious clinical consequences. The recommended first-line therapy is medical management with local wound care and antibiotics.

CONFLICT OF INTEREST

The authors declare that they have no competing interests.

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None

ABBREVIATIONS USED

PG: Pyoderma grangrenosum; PPG: Postoperative progressive gangrene.

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