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CASE REPORT

Pyoderma gangrenosum on an amputation stump treated with ustekinumab

Pioderma gangrenoso sobre un muñón de amputación tratado con ustekinumab

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Abstract

Wound care in the post-operative setting is of great importance in the Traumatology patient. Regular cleaning and evaluation are crucial. However, some cases, may evolve with wound dehiscence and ulceration of the skin borders. These patients might benefit from a multidisciplinary collaboration.
Herein, we describe a case of a pyoderma gangrenosum developed above the knee amputation.
This report described the difficulties in the management of this patient and the evolution of the wound once the correct diagnosis was made, as well as highlights the importance of collaboration between different medical fields in the clinical setting.

Key words: Pyoderma gangrenosum, ulceration

Resumen

El cuidado de las heridas durante el postoperatorio es de gran importancia en el paciente de traumatológico, siendo cruciales la limpieza y la evaluación periódica de las mismas.
Sin embargo, algunos casos pueden evolucionar a una herida dehiscente o a la ulceración. Estos pacientes pueden beneficiarse de una colaboración multidisciplinar.
Aquí describimos un caso de pioderma gangrenoso desarrollado sobre un muñón de amputación de la rodilla.
En este caso se describen las dificultades en el manejo de este paciente y la evolución de la herida una vez realizado el diagnóstico correcto, así como se destaca la importancia de la colaboración entre los diferentes especialistas en el ámbito clínico.

Palabras clave: Pioderma gangrenoso, ulceración.

Case report

A 58-year-old male was admitted to the Gastroenterology department in due to fever and abdominal pain secondary to a case of ileitis.

The patient had a history of hypertension, diabetes, chronic ischemic heart disease, chronic lower limb ischemia (treated via a femoral popliteal bypass in previous years), as well as Crohn disease (treated with a daily dose of 40 mg of hydrocortisone).

The patient developed severe knee pain associated with high fever, and presented erythematous plaques and fluctuating nodules, some of which showed spontaneous external fistulisation.

Then he was assessed by Traumatology and Dermatology department, which determined the infectious nature of the lesions as a first diagnostic possibility. Bacterial cultures were positive for *Staphylococcus aureus*. The diagnosis of erythema nodosum and pyoderma gangrenosum, were taken into consideration but ruled out as exclusion diagnosis due to low clinical suspicion after examination and the positivity of bacterial cultures.

Due to lack of improvement after antibiotic treatment, the skin lesions were surgically debrided as subcutaneous abscesses.

In early post-operative period, deep cutaneous ulcers formed (**Figure 1**). The ulcers progressed rapidly in the following weeks, leading to exposure of both the anterior muscular compartment of the thigh and bone at the previously debrided site.

The patient developed worsening of his general state, and showed signs of septic shock, requiring hospitalization in the Intensive Care Unit.

Figure 1: Appearance of the lession before amputation.

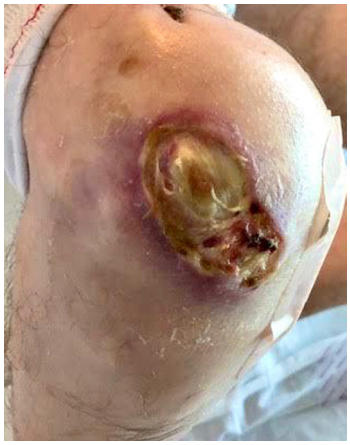


Figure 2: Clinical appearance of the lession after amputation.

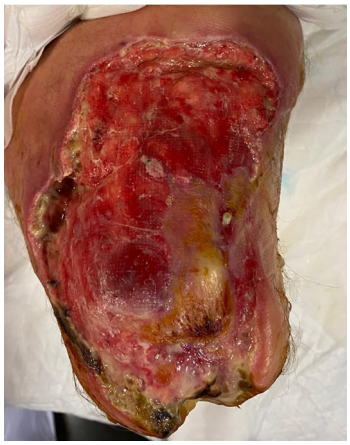
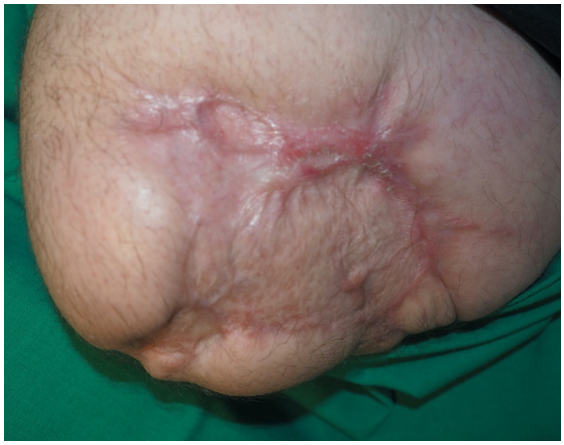


Figure 3: Amputation stump after ustekinumab treatment.



Positive blood cultures for *Klebsiella pneumoniae* were obtained as well as growth of multiresistent *Pseudomonas aeruginosa* and *Escherichia coli* in samples taken from the ulcer. Intravenous antibiotic treatment was started with linezolid and piperacillin-tazobactam.

He was then evaluated together with the Traumatology, Vascular Surgery and Plastic Surgery team. Given that the bypass in the affected leg was non-functioning, the inability to perform knee extension due to quadricipital tendon injury caused by and old injury, and that he was not a candidate for coverage due to a possible active infection, supracondylar amputation was proposed as the best option.

Postoperatively, the patient presented with dehiscence of the surgical wound on the amputation stump, sphacelus and necrotic tissue, and fever.

It was decided to extend the level of the supracondylar amputation. After the second operation, a new ulcerated lesion with violaceous borders appeared. The defect was covered with a meshed partial skin graft, which necrosed. The ulcer persisted and grew. A new medical interconsultation was made to the Dermatology department Under suspicion of pyoderma gangrenosum (PG), prednisone at 60mg/day and topical tacrolimus were started.

A new bacterial culture was positive once more for a multidrug-resistant *Pseudomona aeruginosa*, which was interpreted as a colonization after consultation with the Infectious Diseases unit. The biopsy showed a neutrophilic infiltrate without vasculitis, compatible with PG. Three days after starting prednisone, the ulcer bed began to epithelialize.

As the patient also had decompensation of his Crohn's disease, it was agreed with the gastroenterologists to start ustekinumab as a treatment for extensive PG and Crohn's disease, allowing the prednisone dose to be progressively reduced. Three months after the start of treatment, the patient completely re-epithelialized the ulcer.

He is currently being followed up by different specialists and maintains treatment with ustekinumab every 2 months.

Discussion

Pyoderma gangrenosum is a rare inflammatory disease of reactive origin that belongs to the group of neutrophilic dermatoses. Up to 85% of cases present as a painful ulcer with rapid growth and violaceous borders, although pustular, vegetating, periostomal, superficial granulomatous, malignant, and bullous variants have been described. It most often occurs in the lower extremities. In patients diagnosed with pyoderma gangrenosum, an underlying systemic disease should be ruled out since in about 50% there are other coexisting disorders, mainly inflammatory bowel disease (30%), rheumatoid arthritis (10%) or hematologic disorders (5%)[1,2]. The etiopathogenesis of PG is not well known, but the importance of neutrophils and increased proinflammatory and neutrophil chemotactic factors such as IL-1B, IL-17, TNF alpha, IL-8, IL-6, IL-17 and IL-23 has been widely described. Major and minor diagnostic criteria have been defined, but it should be kept in mind that the definitive diagnosis is always one of exclusion[1,3]. To make the diagnosis of PG, infection must be ruled out, preferably by biopsy. The presence of a neutrophilic infiltrate on histology, in the absence of infection, supports the diagnosis of PG, although a mixed inflammatory infiltrate or leukocytoclastic vasculitis may be seen[3].

Given the absence of complementary tests, the diagnosis of PG can be difficult, as was the case in our patient. Cases have been described in which PG lesions were diagnosed as soft tissue infections (mainly cellulitis or necrotizing fasciitis) or skin tumors, sometimes leading to amputation of the affected limb. It should be considered that surgical interventions can trigger PG lesions, this is since patergia phenomenon can be present in patients presenting this skin lession. The literature reports

cases in which post-surgical PG has even taken years to be diagnosed, which has harmed the patient with unnecessary treatments and prolonged admissions[4].

The assessment of the patient's comorbidities, negative cultures, the evolution of the lesions and poor response to antibiotic therapy can be useful in the diagnosis of PG. It should be considered that the presence of positive cultures does not exclude the possibility of PG and its relevance in the clinical context should always be assessed[4,5].

In our case the patient presented sepsis, whose only apparent origin was the skin ulcers, from which microorganisms had also been isolated in the cultures. We hypothesize that the lesions could be compatible with a case of pyoderma gangrenosum, which would have been overinfected and that acted as the entrance point for the infection.

Imaging tests do not aid in distinguishing with certainty between PG and cases of skin infection[5]. In addition, magnetic resonance imaging of PG lesions may show osteomyelitis (which can be sterile), skin thickening and an increase in the intensity of the subcutaneous soft tissue, among other alterations, which can easily be confused with abscesses, cellulitis, or tumors[6].

For this reason, given the clinical similarity between PG and necrotizing fasciitis, when a patient is suspected of having necrotizing fasciitis, prior to surgery, whenever feasible, the possibility of having PG should be assessed, since the treatments would be the opposite and the performance of debridement in PG would significantly worsen the evolution.

The treatment of choice in PG is corticosteroids, often systemic, and immunosuppressants, which can be complemented with local treatments such as corticosteroids or topical calcineurin inhibitors. Cyclosporine has been frequently used in patients with PG as a corticosteroid sparing agent. In addition, there have been published cases successfully treated with biologic treatments such as anti-TNF alpha, anti-IL1, anti-IL6 or with anti-IL23 drugs, as was the case in our patient, in which we initiated treatment with ustekinumab, an anti-IL-12/23 agent, which is also effective in the treatment of Crohn's disease[2,7,8].

We would like to highlight the importance of multidisciplinary treatment, especially in complex patients with multiple comorbidities, since management between different specialists was the key in our patient to reach the definitive diagnosis allowing subsequent cure.

Ethical approval

Patient´s consent has been obtained before writing this manuscript.

Declaration of interest

The authors declare no competing interest.

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Note

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