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## E-CASE REPORT

# A Case Series of 4 Patients With Peristomal Pyoderma Gangrenosum: Review of Risk Factors and Treatment Response<sup>☆</sup>



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### KEYWORDS

Pyoderma gangrenosum;  
Peristomal;  
Stroma;  
Treatment;  
Tacrolimus

**Abstract** The literature on peristomal pyoderma gangrenosum (PPD) is scarce, and studies to date have included few patients. It is therefore difficult to determine the incidence of PPD, investigate risk factors, or evaluate the effectiveness of the different treatments available. We report on a series of 4 patients diagnosed with PPD at our hospital in 2013 and 2014, and review the clinical characteristics and responses to treatment. Three of the patients had inflammatory bowel disease and 1 had rectal cancer. Three patients responded favorably to initial treatment with 0.1% tacrolimus ointment (administered as monotherapy in 2 cases and combined with immunosuppressants in the other). However, on withdrawal of tacrolimus, the disease recurred in all 3 patients, requiring treatment reintroduction or modification.

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### PALABRAS CLAVE

Pioderma gangrenoso;  
Periestomal;  
Estoma;  
Tratamiento;  
Tacrolimus

**Pioderma gangrenoso periestomal: presentación de 4 casos y revisión de los factores de riesgo y la respuesta terapéutica**

**Resumen** La literatura del pioderma gangrenoso periestomal (PGP) es escasa, y las series publicadas tienen un número de pacientes limitado. En dicho contexto es difícil determinar la frecuencia de la enfermedad, así como sus factores de riesgo, y valorar la eficacia de los distintos tratamientos disponibles. Presentamos una serie de casos de PGP diagnosticados en nuestro centro entre los años 2013 y 2014 y revisamos las características clínicas y la respuesta terapéutica. Se incluyeron un total de 4 pacientes, 3 de los cuales estaban diagnosticados de enfermedad inflamatoria intestinal, mientras que uno de los casos se asoció a carcinoma de recto. Tres pacientes presentaron evolución favorable tras el tratamiento inicial con tacrolimus

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0,1% en pomada (2 casos en monoterapia y uno asociado a otros inmunosupresores), con recurrencia en todos ellos tras la suspensión del tratamiento precisando reintroducción o cambio de tratamiento.

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## Introduction

Peristomal pyoderma gangrenosum (PPG) is one of the most uncommon subtypes of pyoderma gangrenosum, accounting for 15% of all cases of pyoderma gangrenosum.<sup>1</sup> Its characteristics enable it to be differentiated from other types (Table 1). Almost 100% of cases of PPG are associated with inflammatory bowel disease (IBD), although not exclusively,<sup>1,2</sup> whereas pyoderma gangrenosum is associated with IBD in 15%-20% of cases.<sup>2</sup>

The literature on PPG is scarce, and studies to date have included few patients.<sup>1</sup> Therefore, it is difficult to determine the incidence of the disease and its risk factors and to evaluate the effectiveness of the various treatment options.<sup>2</sup> We present the clinical characteristics and response to therapy of a series of 4 cases of PPG.

## Material and Methods

We performed a retrospective observational study of all cases of PPG diagnosed based on clinical and histopathology findings at our hospital during 2013-14. We excluded all dubious cases and cases for which no biopsy evidence was available. After reviewing the clinical histories, we recorded the following variables: sex, body mass index (BMI), age, underlying disease, activity of the underlying disease, type of surgery, treatment, and response to treatment.

**Table 1** Differences Between Pyoderma Gangrenosum and Peristomal Pyoderma Gangrenosum.

	PG	PPG
Incidence	2-3 cases/million inhabitants/y <sup>5</sup>	Incidence in general population unknown 0.6%-0.7% of stomas <sup>2,3</sup>
Site	Mainly on lower limbs, although not exclusively	Peristomal
Association with systemic diseases	50-60%	More than 90%
Association with IBD	15-20%	Almost 100%

Abbreviations: IBD, inflammatory bowel disease; PG, pyoderma gangrenosum; PPG, peristomal pyoderma gangrenosum.

## Results

The study population comprised 4 patients (3 men and 1 woman) aged between 27 and 79 years (Fig. 1 A-D), whose characteristics are included in Table 2. Three of the 4 patients had IBD (ulcerative colitis in 2 cases and Crohn disease in 1 case); 1 patient had rectal carcinoma, although this was not associated with IBD. Time from surgery to onset of PPG varied from 22 days to more than 4 years. Outcome was favorable in 3 patients after initial treatment with 0.1% tacrolimus ointment (monotherapy in 2 cases and combination with other immunosuppressive agents in 1 case). The disease recurred after suspension of treatment, which had to be reintroduced or switched.

## Discussion

The incidence of PPG seems to have increased during recent years.<sup>1</sup> PPG is observed in approximately 2%-4% of patients with IBD who undergo intestinal surgery with stomas and in 0.6% of patients who undergo surgery with ostomy.<sup>1-3</sup> PPG is almost always associated with IBD, although there have been reports of isolated cases associated with cancer, diverticulitis, neurologic dysfunction, monoclonal gammopathy, and collagen disease.<sup>1,4</sup> In the present series, only 1 case was associated with rectal carcinoma, whereas the remainder were due to underlying IBD.

Most studies highlight the greater incidence of PPG in patients with Crohn disease than in those with ulcerative colitis,<sup>2</sup> although this finding seems to have been called into question in recent years.<sup>1</sup> Some authors explain the greater incidence of PPG in patients with Crohn disease by the more common indication of surgery for this condition, which requires partial resection of the intestine. In patients with ulcerative colitis who require surgery, on the other hand, the usual approach is total colectomy, which results in definitive cure of the disease.<sup>2</sup> Only 1 patient in the present series was diagnosed with ulcerative colitis, whereas 2 of those with Crohn disease had undergone total colectomy with conservation of the rectum in order to create an ileoanal pouch. Both had pouchitis (disease activity in this section of the colonic mucosa). Other authors have also suggested the presence of residual activity in the rectum as a risk factor for PPG in patients with ulcerative colitis.<sup>2</sup> The current trend in surgery is to perform reconstructive techniques with ileoanal pouches in order to avoid permanent ileostomy, and this may play a role in the increased incidence of PPG in patients with ulcerative colitis.

In 2012, Wu et al.<sup>1</sup> performed a study on risk factors for PPG in a series of 15 cases. The multivariate analysis revealed independent risk factors and statistically

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