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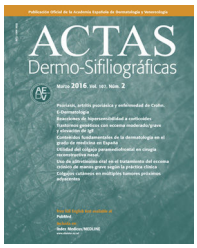

CASE AND RESEARCH LETTERS

Atypical Ulcerated Lesions in a Patient With Cogan Syndrome

Lesiones ulcerativas atípicas en un paciente con síndrome de Cogan

Dear Editor

Cogan syndrome (CS) is a rare vasculitis characterized by recurrent interstitial keratitis and audiovestibular symptoms including hearing loss, tinnitus, and vertigo. To date, cutaneous lesions have been only rarely described associated with this autoimmune disease.[1] We present the case of a patient with CS and multiple ulcerated lesions.

A 39-year-old white Spanish woman diagnosed with CS at age 31 years, with stromal keratitis and severe bilateral hearing loss, consulted for a 2-week history of ulcers that had started as pustules on the legs and then spread to the thighs, arms, abdomen, and back. At the time of onset, the patient was on treatment with methotrexate (25 mg/wk), prednisone (10 mg/d), and tozilizumab (8 mg/kg/mo). Physical examination revealed numerous pustules and ulcerated necrotic lesions on the legs, thighs, arms, back, and abdomen (Fig. 1A). On suspicion of ecthyma, topical antibiotics and oral cloxacillin were prescribed. Smear cultures for bacteria, fungi, and mycobacteria were all negative. Additional tests were within normal limits and the chest radiograph was normal. Two weeks later the patient presented further pustules and enlargement of those previously present. At no time did she report fever or malaise. Histopathology of a biopsy from an ulcer on the left thigh revealed an ulcerated epidermis with epidermal necrosis and pustules at the border, with an abscessified area of skin and a diffuse dermal inflammatory infiltrate formed by neutrophils, histiocytes, lymphocytes, and occasional multinucleated giant cells (Fig. 2A and B). No clear signs of vasculitis were observed. Immunofluorescence was negative. Bacterial cultures from the cutaneous biopsy and new smears were positive for *Pseudomonas aeruginosa*/*Staphylococcus haemolyticus*, and *Streptococcus pyogenes*/*Pseudomonas putida fluorescens*, respectively. Oral cefuroxime and ciprofloxacin were started but the lesions showed no improvement. It was then that prednisone (at a dose of up to 1 mg/kg/d) was prescribed. The patient responded favorably, and in 2 weeks the lesions had completely re-epithelized except for an ulcer in the surgical wound of the biopsy and a linear ulcer on the left arm that had developed after trauma a week earlier, suggesting a pathergy phenomenon (Fig. 1B). At follow-up, no lesions were observed. At the time of writing, the patient remains asymptomatic and continues her usual therapy.

Diagnosis of the cutaneous lesions in our patient was difficult, and ecthyma was our initial provisional diagnosis. The lack of response—and even a deterioration—of the lesions after antibiotic therapy, the pathergy phenomenon, and the favorable response to steroids led us consider a pyoderma gangrenosum (PG)-like neutrophilic dermatosis. However, the multiple lesions and the histopathology were not conclusive for either PG or other forms of neutrophilic dermatosis. Multiple lesions have rarely been described in PG.[2] In 70% of cases, PG is associated with an underlying disease such as inflammatory bowel disease (IBD), inflammatory arthritis, or a hematologic malignancy.[3–5]

Tirelli et al.[6] found about 250 reports of patients with CS, only 13 of whom had concomitant chronic IBD; of these, none experienced improvement after therapy.

The etiology and pathogenesis of CS are unknown. Initially, the disease was thought to be caused by an infection, but it is now considered to be an autoimmune disorder.[7] In addition to the ocular and audiovestibular involvement, numerous systemic manifestations have been reported in CS, most commonly of cardiovascular, neurological, or gastrointestinal origin. Approximately 70% of patients have an underlying systemic disease. Vasculitis is considered to be the pathological mechanism[8]; however, even though swollen endothelial cells and focal fibrinoid deposits were seen in the biopsy from our patient, it was not possible to make a conclusive diagnosis of cutaneous vasculitis.

In a review of the literature, we have found that CS has rarely been associated with skin manifestations, and in most cases such manifestations are reported as rash or ulcers, with no further detailed clinical description or histopathology study. In a multicenter study that included 32 patients with CS, only 7 showed skin and mucous membrane involvement or chondritis: 3 cases of rash, with evidence of vasculitis in only 2 of them, 1 patient with photosensitivity, 1 with vitiligo, 2 with oral ulcers and 2 with chondritis.[9] Pagnini et al.[1] described 23 children with CS; only 3 presented skin manifestations, all of which were described as rashes. As far as we are aware, these multiple ulcerated necrotic lesions have not previously been described in association with CS.

In summary, we have described an unusual case of multiple ulcerated necrotic lesions in a patient with CS.

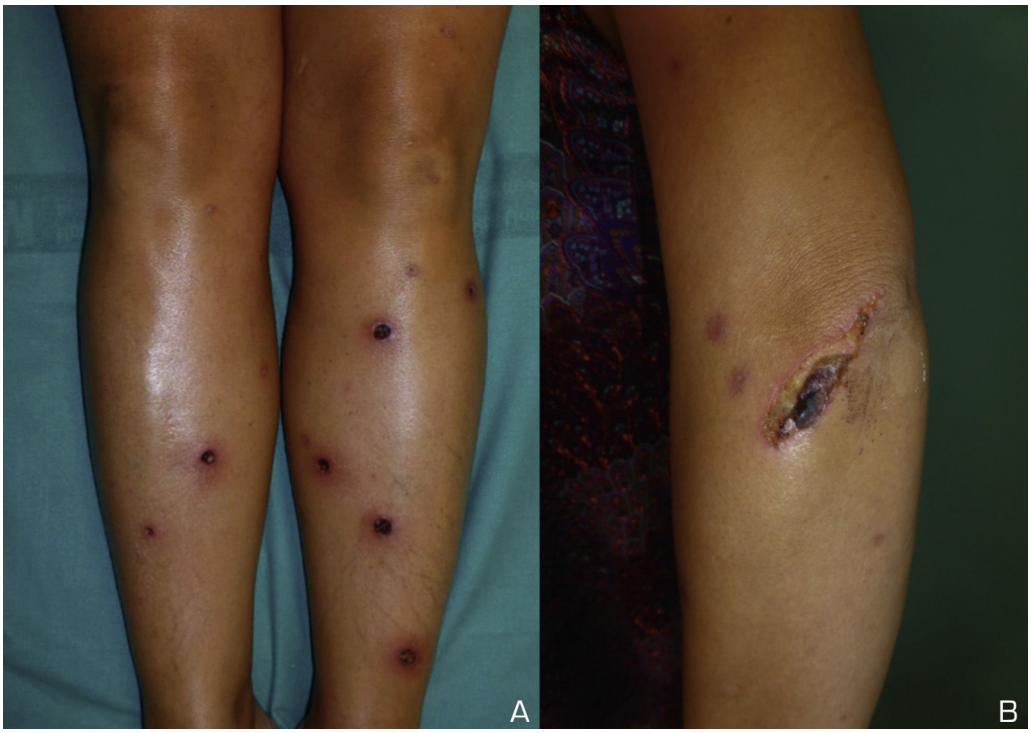

Figure 1 (A) Ulcerated necrotic lesions on both lower legs. (B) Linear ulcer on the left arm exhibiting the pathergy phenomenon.

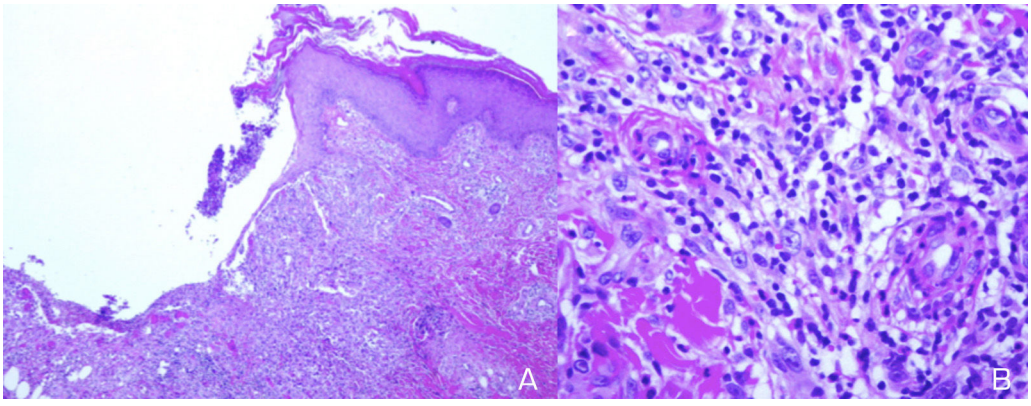

Figure 2 (A) Ulcerated epidermis with a diffuse inflammatory infiltrate in the dermis. (B) Infiltrates formed of neutrophils, histiocytes, lymphocytes, and occasional multinucleated giant cells.

Conflict of interests

The authors declare no conflict of interests.

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Pilot Study to Evaluate Postsurgical Dehiscence After Continuous Suture by Tissue Planes☆

Estudio piloto de valoración de dehiscencias posquirúrgicas tras sutura continua por planos

To the Editor:

The main objective of dermatologic oncologic surgery is the surgical excision of a tumor with sufficient safety margins, allowing correct management thereafter. However, achieving the best functional and cosmetic result is a secondary objective that is acquiring ever greater importance, and that has even been extended to the excision of benign lesions, many of which are not included in the range of services offered by numerous hospitals.[1]

Several different surgical techniques have been developed to avoid wound dehiscence in the immediate and late postoperative periods. The conventional technique for which the best results have been reported is the one described by Weber and Wulc[2] in 1992. That technique was modified and improved by Serrano et al.[3] in 2015, hiding the entry and exit orifices of the suture by performing the subcutaneous vector-running suture along the length of the wound, completely burying the suture.

Based on the concept proposed by Serrano,[3] we perform a continuous suture by tissue planes to prevent dehiscence of the wound and, thus, of the scar.[4] Our wound closure consists of a continuous subcutaneous suture followed by an intradermal suture, demonstrated previously in the surgical videos section of *Actas Dermo-Sifiliográficas*.

This adaptable and simple technique enables us to control tension forces until the wound has healed. It is not necessary to remove the suture material. To avoid the formation of foreign body granulomas that can develop with nonabsorbable sutures, we use a mid-term absorbable suture: Novosyn 3/0, a mid-term absorbable multifilament synthetic suture of braided and coated poly(glycolide-co-L-lactid 90/10). Two additional advantages of this technique are that it is rapid to perform and that no assistant is needed to cut the suture material after each stitch.[5]

Between January and December, 2015, we performed a prospective nonrandomized pilot study in our unit to determine dehiscence in the immediate postoperative period and in the mid-term using the technique described above and the one employed in conventional surgery. The study was approved by the ethics committee of our hospital. All patients were over 18 years of age. The variables gathered were sex, age, clinical diagnosis, length and breadth of the incision, and width of dehiscence at 3 and 6 months. All the lesions excised were on the trunk or root of the thighs.

A descriptive analysis was performed of the overall population and by groups after testing for normality of the continuous quantitative variables using the Kolmogorov-Smirnov test. Variables were compared using the Student t test or Welch test for normal variables and the Mann Whitney U test for non-normal variables. The χ^2 test was used to study associations between qualitative variables. Sixty-five patients were included in the study. Wound closure was performed with the continuous suture by tissue planes in 37 patients and with the conventional Weber technique in 28 (Table 1). No statistically significant differences were found between the 2 groups in the variables age, sex, and length and breadth of the incision. Wounds closed using the continuous suture by tissue planes presented a mean dehiscence of 4.88 mm, whereas mean dehiscence in the conventional suture group was 7.25 mm; this difference was statistically significant ($P = .002$).

No infectious complications were observed in the immediate or late postoperative periods with either of the 2 techniques after strict adherence to the pre- and postsurgical verification protocols applied in our hospital.[6]

The main limitation of our pilot study is that comparison of the 2 surgical techniques was not performed in

Table 1 Comparison of the 2-Suture Techniques Performed in the Study Patients.

	Continuous Suture by Tissue Planes	Conventional Suture	*P* Value
Age, y	54.66 ± 19.93	50.67 ± 15.94	.381
Sex			.880
Male	23	13	
Female	18	11	
Dehiscence, cm	0.49 ± 0.29	0.73 ± 0.27	.002
Incision length, cm	4.48 ± 1.32	4.01 ± 1.48	.203
Incision width, cm	2.19 ± 1.01	2.54 ± 1.18	.251

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