

Case Report

Localized Bullous Pemphigoid Associated with Chronic Venous Stasis: A Case Report

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Abstract: Bullous pemphigoid (BP) represents one of the most prevalent forms of bullous diseases. Nevertheless, its localized variant, known as Localized Bullous Pemphigoid (LBP), is relatively uncommon, comprising only 2.5% of all BP cases. LBP is typically associated with trauma and may manifest on any wound or surgical site. Although chronic venous stasis has not been well-documented as a predisposing factor, we present the case of an 80-year-old man who exhibited blistering lesions localized to his left lower limb. He had a history of chronic venous insufficiency (CIV) on both legs, with no improvement after surgery. Clinical-pathology correlation concluded with the diagnosis of LBP. Treatment with clobetasol topical ointment and compression therapy led to complete remission of the skin lesions after one month. This report underscores chronic venous stasis as a contributing trigger for LBP.

Keywords: Autoimmune Disease, Chronic Venous Stasis, Localized Bullous Pemphigoid, Pemphigoid.

INTRODUCTION

Bullous pemphigoid (BP) is an autoimmune disease caused by autoantibodies targeting the hemidesmosomal proteins [1]. Localized bullous pemphigoid (LBP) is a rare BP subtype that manifests after exposure to triggers [2]. Some have been described such as, surgery site, radiotherapy or photochemotherapy and chronic lower limb edema [3]. There are few case reports of LBP in the context of venous stasis and it is hypothesized that the presence of epidermal inflammation could represent an important triggering factor [4]. We describe a case of a male patient diagnosed with LBP in the left lower limb with a history of chronic venous insufficiency (CIV).

CASE REPORT

An 80-year-old Hispanic man presented to the dermatology service with tense blisters on his left lower limb. He had a history of CIV affecting both legs, and surgical intervention with bilateral saphenectomy had been previously performed. No improvement in his left lower limb was achieved. He described an initial lesion characterized by erythematous-edematous plaques that, one week later, evolved into tense blisters accompanied by intense itching. During the physical examination, numerous tense bullae with circular erosions, hematic crust, and perilesional erythema were observed, limited to the left lower limb [Figure 1].

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Figure 1: Primary lesion. Tense bullae with circular erosions, hematic crust and perilesional erythema limited to the left lower leg and limb

Two punch biopsies were taken, one from an intact bulla and another one from perilesional skin. Histopathology revealed a subepidermal blister with perivascular and interstitial eosinophilic infiltration. The basal membrane was visible in the blister's roof using type IV collagen immunohistochemistry. Direct immunofluorescence (DIF) was negative [Figure 2].

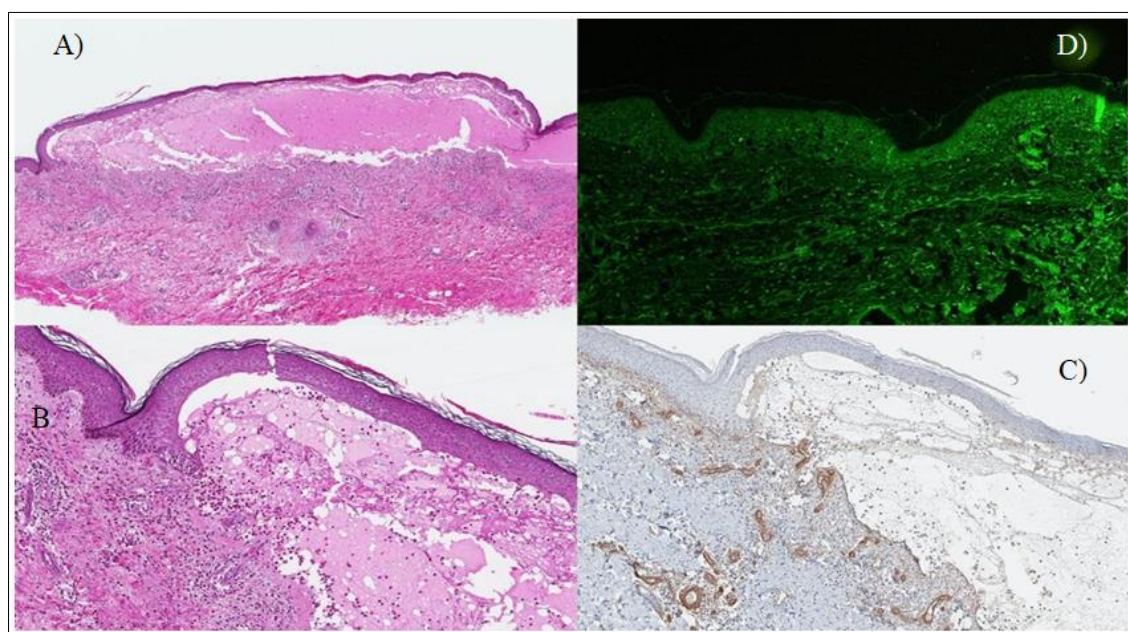


Figure 2: Histopathology findings

- A. Subepidermal blister with perivascular and interstitial eosinophilic infiltration (Hematoxylin and eosin)
- B. Higher magnification (Hematoxylin and eosin)
- C. The basal membrane was visible in the blister's roof using type IV collagen immunohistochemistry.
- D. Negative direct immunofluorescence.

Although negative DIF results, the other findings strongly suggested the presence of LBP. Treatment with clobetasol propionate topical ointment twice daily and compression therapy resulted in clinical improvement, with complete remission of the lesions after one month of treatment [Figure 3].



Figure 3: Complete remission of skin lesion

DISCUSSION

LBP refers to the occurrence of site-restricted clinical presentation of BP in patients with no history of BP, for a minimal period of three months [2]. It usually occurs in male patients older than 70 years as blistering eruptions, tense pruritic bullae and vesiculobullous dermatitis [5]. BP is one of the most prevalent forms of bullous disease. However, it is uncommon in its localized form with a prevalence of 2.5% of all cases of BP [2]. The pathogenesis of BP is caused by circulating autoantibodies that bind to BP230 and BP180, causing complement activation, inflammatory cell recruitment, and the release of proteolytic enzymes [5]. When LBP is caused by trauma, it can appear in any site of the body; otherwise, it is most common in the lower limbs [3]. Radiotherapy has been described as the most common presumed trigger factor, followed by thermal or chemical burns, surgical procedures, ultraviolet radiation and drugs including neuroleptics and diuretics [2-6]. Our patient did not exhibit any of these triggering factors. The association between venous stasis and LBP has been reported in just one previous case. It has been hypothesized that the immunologic changes associated with venous stasis could represent the triggering factor for the generation of localized disease [4]. Extravasation of blood and plasma, which includes mediators of innate and adaptive immune responses to circulating tissues [7], disrupts the immune system causing damage and predisposing to autoantigen presentation, autoreactive T lymphocyte production, and autoantibody-producing B cells [4-8]. Clinical-pathological correlation, as well as serological tests to detect circulating antibodies, are used to make the diagnosis. Although the clinical and histologic findings were consistent with LBP, DIF remained negative which was considered as a false negative. It has been previously described that BP sampling from the lower extremities should be avoided because of the high rate of false negatives. The Weigand study found in a retrospective series of 46 patients with BP diagnosis, a DIF false negative rate of 33% on the lower extremities [9]. Despite the generally benign course of this disease, there is a 36.8% risk of progression to a generalized form [10].

We present an atypical case of LBP associated with venous stasis, underscoring it as a potential triggering factor for the condition. Clinicians should maintain a high level of suspicion for this pathology, promoting early intervention to enhance patient outcomes. Additionally, we emphasize the significance of recognizing the elevated rates of false negatives in DIF when a biopsy is performed on lower limbs. This awareness underscores the need for a comprehensive diagnostic approach.

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