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Botulinum toxin in the treatment of pemphigus vegetans of the scalp

Toxina botulínica no tratamento de pênfigo vegetante de couro cabeludo

Case Report

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ABSTRACT
Pemphigus vegetans is a rare clinical form of pemphigus vulgaris, and scalp involvement is even rarer. Treatment can be challenging, often requiring systemic immunosuppression. Botulinum toxin can be an effective and low-risk alternative for the treatment of pemphigus vegetans unresponsive to conventional treatments. We report an unprecedented case in the literature of successful treatment of scalp lesions through the application of botulinum toxin type A.
Keywords: Pemphigus; Scalp; Botulinum Toxins, Type A

RESUMO
O pênfigo vegetante é uma forma clínica rara de pênfigo vulgar, sendo o acometimento do couro cabeludo ainda mais incomum. O tratamento pode ser desafiador, com frequente necessidade de imunossupressão sistêmica. A toxina botulínica pode ser uma alternativa efetiva e de baixo risco para o tratamento do pênfigo vegetante não responsivo aos tratamentos convencionais. Relatamos um caso inédito na literatura, com sucesso terapêutico das lesões em couro cabeludo através da aplicação de toxina botulínica tipo A.
Palavras-chave: *Pênfigo; Couro Cabeludo; Toxinas Botulínicas Tipo A*

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INTRODUCTION

Pemphigus encompasses a group of autoimmune bullous diseases of the skin and mucous membranes, with variable geographic distribution worldwide, and pemphigus vulgaris is its most frequent subtype.[1,2,3] Pemphigus vegetans is a rare clinical form,[6,8] and scalp involvement is even rarer.[3,6,7,8] Treatment can be challenging, often requiring systemic immunosuppression.[2,3,4,5,6] The present study aims at presenting an unprecedented case in the literature of pemphigus vegetans on the scalp, refractory to conventional treatment, achieving good clinical response to botulinum toxin type A.

CASE REPORT

A 50-year-old male patient, White, previously diagnosed with pemphigus vulgaris, had bullous lesions on his nose, scalp, oral mucosa, trunk, abdomen, arms, and legs. A laboratory assessment found no relevant alterations. The initial therapeutic options were prednisone, azathioprine, and mycophenolate, but they had to be discontinued due to their side effects. Except for the lesions on the patient's scalp, which progressed to a vegetative appearance, all other lesions were controlled by using rituximab (Figure 1A and 1B). Pathologic examination of the scalp lesion found suprabasal acantholysis, hyperkeratosis, acanthosis, and moderate lymphocytic inflammatory infiltrates (Figure 2). Direct immunofluorescence found IgG positivity, with intercellular pattern in the epidermis. Clinical-histological correlation corroborated the diagnosis of pemphigus vegetans. The choice was made to administer botulinum toxin as an experimental treatment for the scalp lesions. After topical application of 4% lidocaine cream (Dermomax®) on the scalp plaques for 30 minutes and antisepsis with 0.2% aqueous chlorhexidine, botulinum toxin type A (Botox®), 2 UI/cm², was injected intradermally (Table 1). After 6 weeks, lesion verrucosity had improved by over 50%. At reassessment, a new dose of botulinum toxin was administered, following the same procedure, achieving progressive improvement (Figure 3A and 3B).

DISCUSSION AND CONCLUSION

Currently, there is no literature on the use of botulinum toxin to treat pemphigus vegetans. Nevertheless, it has shown promise as a low-risk therapeutic alternative for this dermatosis.[9, 12,13,14] The mechanisms of action of botulinum toxin are not yet fully understood. One hypothesis suggests that the inhibi-

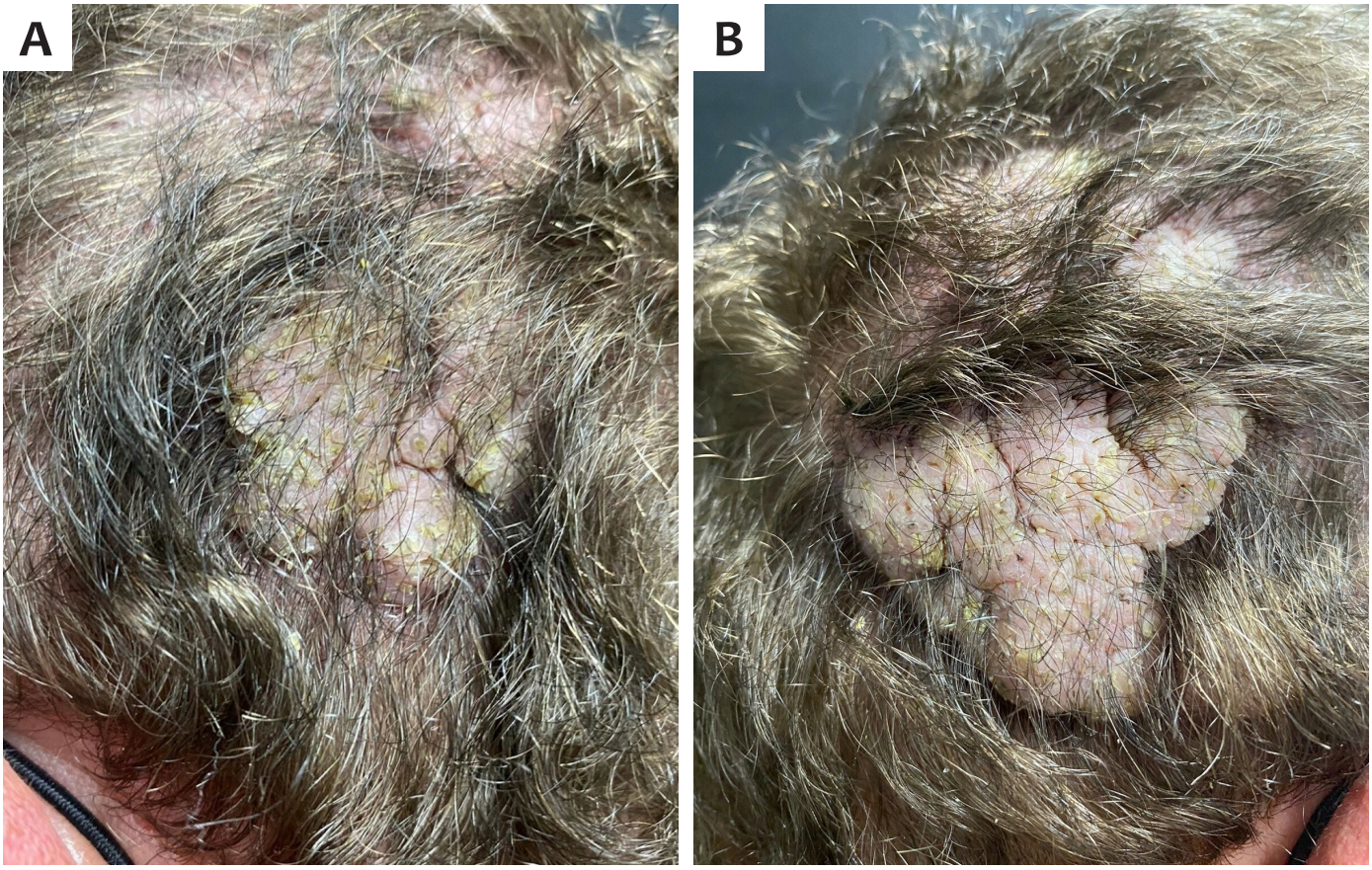

Figure 1: A - Vegetative plaque on the left occipital region before treatment with botulinum toxin; **B -** Vegetative plaque on the right occipital region before treatment with botulinum toxin

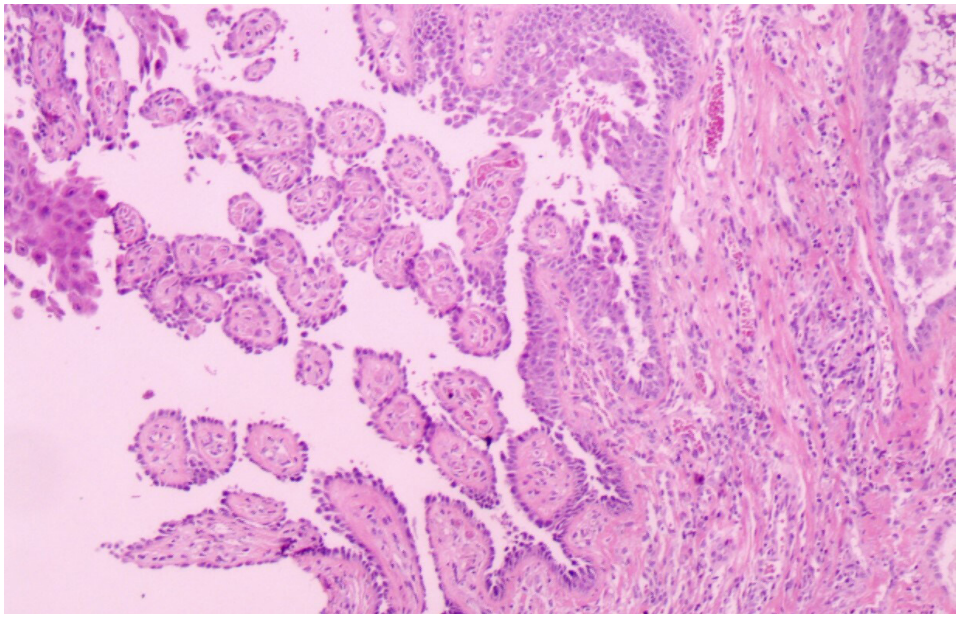

Figure 2: Pathologic examination showing suprabasal acantholysis, hyperkeratosis, acanthosis, and moderate lymphocytic inflammatory infiltrates (hematoxylin-eosin, 200x)

Table 1: Application of botulinum toxin to pemphigus vegetans lesions on the scalp

	Description of application technique
1	Topical application of 4% lidocaine cream (Dermomax®) on lesions 30 minutes before the procedure
2	Antisepsis with 0.2% aqueous chlorhexidine, with full removal of topical anesthetic from application sites
3	Intradermal injection of botulinum toxin type A (Botox®), 2 UI/cm^2, into lesions

Figure 3: A - Left occipital region 6 months after the first application of botulinum toxin; **B -** Right occipital region 6 months after the first application of botulinum toxin

tion of acetylcholine binding to the neuromotor receptors of sweat glands may reduce both sweating and neural activity of the eccrine glands.[12,13,15,16] In addition, it has been proposed that botulinum toxin blocks the release of neuropeptides and other substances involved in the local inflammatory process.[11,14] In the present case, a marked improvement in the lesions was observed, with a reduction of more than 50% in verrucosity in only 6 weeks, and progressive disappearance over a 6-month period. The leading hypothesis is that the anhidrotic and anti-inflammatory effects of botulinum toxin decrease the risk of local infections, both micro and macro, which could otherwise exacerbate the proinflammatory process and worsen the clinical condition. Based on reports of the use of botulinum toxin to treat other dermatoses, we believe that periodic reapplications (every 6 months, for example) may be necessary to optimize outcomes. Botulinum toxin may represent an effective and low-risk alternative for the treatment of pemphigus vegetans unresponsive to conventional treatments. However, clinical trials are still required to clarify the best use and the mechanism of action of this therapy. ●

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Approval of the final version of the manuscript; Conception and design of the study; Preparation and writing of the manuscript; Acquisition, analysis and interpretation of data; Effective participation in the conduct of the study; Intellectual participation in the propaedeutic and/or therapeutic approach to the cases studied; Critical review of the literature; Critical revision of the manuscript

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