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CASE REPORT

Unsual Presentation of Pemphigus Vegetans Mimicking Condyloma Latum

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ABSTRACT

Pemphigus vegetans (PVeg) is a rare variant of pemphigus, characterized by chronic, vegetating lesions primarily affecting skin folds and mucous membranes. The diagnosis of PVeg is established through clinical, histological, and immunological findings. It can be challenging due to its clinical resemblance to other conditions, such as condylomata latum, pyodermatitis pyostomatitis vegetans and Hailey-Hailey disease.

This case report presents a 39-years-old female with genital papulo-nodular lesions evolving over five months. The patient, with a history of epilepsy and no sexual risk behaviors, was initially suspected to have condylomata latum. However, after negative serology for sexually transmitted infections, histopathological analysis and immunological testing a diagnosis of pemphigus vegetans was confirmed. The patient was treated with prednisone and rituximab, resulting in complete resolution of the lesions within one month.

This case highlights the importance of considering pemphigus vegetans in the differential diagnosis of vegetating genital lesions, particularly in the absence of sexual risk behaviors.

KEYWORDS

Pemphigus vegetans; Autoimmune disease; Condylomata latum; Genitalia

INTRODUCTION

Pemphigus vegetans (PVeg) is a rare variant of pemphigus, characterized by chronic, vegetating lesions predominantly affecting the skin folds and mucous membranes [1,2]. It affects mostly middle-aged women with

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2 clinical forms, Neumann PVeg and Hallopeau PVeg with different clinical presentation and prognosis [1]. PVeg diagnosis can be challenging, especially when the lesions mimic other conditions. This case report presents the case of unusual presentation of genital PVeg in 39-years-old female patient who presented with genital flat-topped papulo-nodular and vegetating lesions mimicking condylomata latum. This case underscores the importance of considering pemphigus vegetans in the differential diagnosis of vegetating genital lesions.

CASE PRESENTATION

A 39-years-old female patient was admitted to the emergency room for papulo-nodular lesions of the genital area evolving 5 months prior to her admission. She had a history of epilepsy on lamotrigine. She didn't report sexual risk behaviors or recent medication use. Dermatological examination revealed multiple papillomatous and vegetating flat-topped papular and nodular lesions and hypertrophic plaques, located bilaterally on the genital mucosa, pubis, and groins with history of pustules evolving gradually to papulo-nodular lesions with no history of ulcers prior to the appearance of papillomatous vegetations (Figure1).

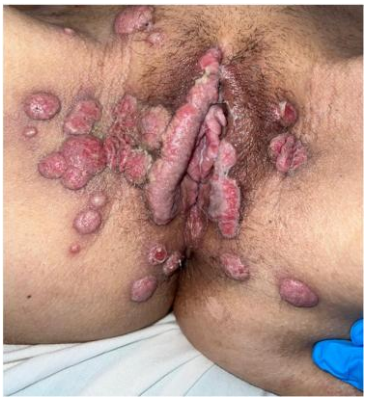

Figure 1: Pemphigus vegetans: Multiple flat-topped papular and vegetating lesions located on genitalia mimicking condylomata latum.

Some lesions showed superficial erosion with oozing of purulent fluid. Other skin folds were normal. Dermoscopy of genital lesions showed the presence of warty surface and dotted vessels. The patient was in otherwise good general condition. Given the warty-like appearance of the flat-topped papular lesions and their location on the genitals and perineum we first considered condyloma latum diagnosis and carried out TPHA-VDRL serologies, which were negative, as well as other sexual transmitted infections serologies that were also negative (HIV, HCV, HBV).

Differential diagnoses including pemphigus vegetans, pyodermatitis pyostomatitis vegetans, condylamata acuminate and halogenoderma were considered.

The patient did not present with any digestive symptoms and underwent an upper gastrointestinal endoscopy and colonoscopy, both of which did not show any signs suggestive of inflammatory bowel disease.

Biopsy sample was obtained from a genital flat-topped papular lesion. Histopathology revealed suprabasal splitting, acanthosis and papillomatous epidermis without cytonuclear atypia as well as acantholytic cells grouped sometimes in clusters floating in the blister along with neutrophils and eosinophils (Figure 2 and 3).

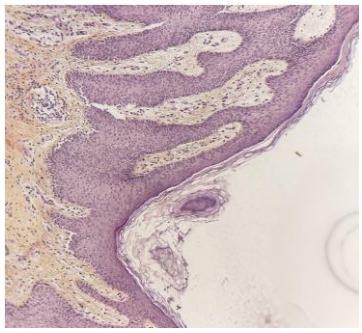

Figure 2: Histopathology of pemphigus vegetans: Skin tissue showing epidermal thickening with orthokeratotic hyperkeratosis and a subtle suprabasal splitting (H&E × 10).

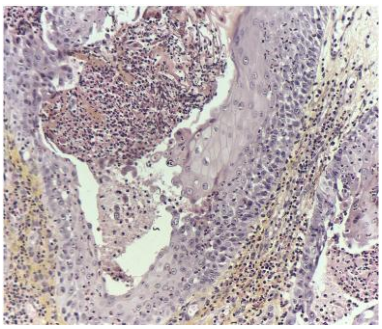

Figure 3: Histopathology of pemphigus vegetans: The epidermis exhibits exocytosis of inflammatory cells, predominantly neutrophilic leukocytes. This is associated with eosinophilic abscesses accompanied by acantholytic cells (H&E × 20).

ELISA revealed a high circulating anti-Desmoglein3 antibodies value estimated at 7 (<0,5) but no anti Desmoglein1 antibodies were found.

In this case, direct immunofluorescence (DIF) was unavailable; however, the diagnosis of PVeg was confirmed based on clinical and histological findings, as well as serologic detection of anti-desmoglein 3 antibodies.

Treatment with Prednisone was administered at a dose of 1.0 mg/kg/day, gradually reduced over a period of 6 months, in combination with two infusions of Rituximab, each 1 g, given two weeks apart, followed by a 500 mg dose at the 6-months with spectacular evolution and complete healing of all lesions 1 month after treatment initiation (Figure 4).

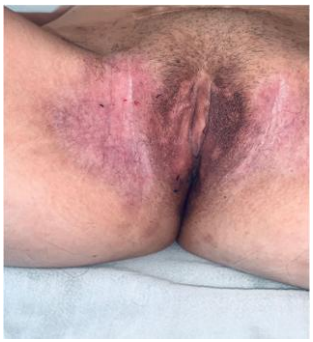

Figure 4: Pemphigus vegetans after treatment: Complete healing of all lesions after prednisone and rituximab treatment.

DISCUSSION

PVeg is a rare variant of pemphigus which represents 2% of all pemphigus subtypes affecting mostly middle-aged women, which is the case of our 39-years-old female patient. PVeg lesions are mostly located in the folds and the oral mucosa [1,2].

We distinguish 2 clinical forms of PVeg with different clinical presentation and prognosis that may coexist in a single patient [2]. Neumann PVeg is characterized by blisters leading to the development of hypertrophic, papillated plaques, whilst the Hallopeau subtype is characterized by multiple pustules that slowly progress into verrucous and papillomatous growths. Hallopeau PVeg follows a more benign course [1,2]. Some studies have raised doubts about this subclassification.3 The main differential diagnosis include Pyodermatitis pyostomatitis vegetans, condylomata lata, Hailey-Hailey disease, and halogenoderma [2-5].

The diagnosis of PVeg is established through clinical, histological, and immunological findings. Histopathological examination of PVeg lesions shows suprabasal splitting associated with acantholysis, intraepidermal eosinophilic abscesses, epidermal hyperplasia, papillomatosis and acanthosis [2,6,7].

The main differential histological diagnosis of pemphigus vegetans is pyodermatitis-pyostomatitis vegetans, as both share similar clinical and histological features; however, the latter typically shows negative findings on immunofluorescence [2]. Pemphigus vulgaris is also a histological differential diagnosis. While it shows acantholysis, it typically lacks neutrophilic and eosinophilic microabscesses, and papillary epithelial hyperplasia is not commonly observed [2].

Direct immunofluorescence is characterized by intercellular IgG and C3 deposition within the epidermis [2-4,8] ELISA serum analysis can detect desmoglein 3 autoantibodies in patients' blood, with high sensitivity and specificity [4].

In this case, direct immunofluorescence was unavailable; however, the diagnosis of PVeg was confirmed based on clinical and histological findings, as well as serologic detection of anti-desmoglein 3 antibodies.

Management of PVeg is the same as pemphigus vulgaris2 we opted for prednisone 1.0 mg/kg/d, in combination with two infusions of Rituximab, each 1 g, given two weeks apart, followed by a 500 mg dose at the 6-months according to the European Academy of Dermatology and Venereology guidelines on the management of pemphigus vulgaris [9].

In our case, atypical clinical presentation of PVeg was challenging, especially because of the flat-topped, eroded, exuding papules appareance found in the genital area and the vulva, this clinical presentation is mostly seen in Condylomata latum, a cutaneous manifestation of secondary syphilis. It is therefore important for the clinician to keep in mind the possibility of genital inflammatory pathology, especially in the absence of high-risk sexual behaviors, and to pursue further laboratory tests in order to refine the diagnosis.

CONCLUSION

In conclusion, this case underscores the importance of considering PVeg when faced with unusual genital lesions, especially in the absence of sexual risk behaviors.

Early and accurate diagnosis, supported by a combination of clinical, histopathological, and immunological findings, is crucial for effective management. In this case, the patient’s favorable response to treatment with

prednisone and Rituximab highlights the importance of timely intervention and personalized care. Clinicians should maintain a high index of suspicion for uncommon dermatological conditions like PVeg, to ensure proper diagnosis and optimize therapeutic outcome.

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