

Case Report

Genital vesicobullous disease-not herpes genitalis!

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ABSTRACT

Genital bullous pemphigoid (BP), a localized subset of bullous pemphigoid, has been identified in children and women in the past. However, we report an adult male presenting with blisters confined to glans and shaft of penis, mimicking herpes genitalis, failing antiviral therapy. The patient responded to treatment with dapsone.

Keywords: Genital bullous pemphigoid, Localized subset, Bullous pemphigoid, Dapsone

INTRODUCTION

Bullous pemphigoid, an autoimmune, vesicobullous skin disease, against the epidermal hemidesmosomes, BP 180 and BP 230, disrupting skin integrity. It mostly affects the elderly, with an incidence of less than 0.5 cases per million population in individuals younger than 50 years of age. The condition has a female predilection.¹ Localized variants of the disease are infrequent, resulting in delayed diagnosis.² We report a case of genital (BP), a rare subset, in a middle aged, male patient, with no significant aggravating factors.

CASE REPORT

A 48-year-old, male, married patient presented to our clinic with complaints of multiple, variably sized and shaped, tense vesicles and bulla on a normal base, containing clear fluid, localized to the dorsal aspect of the penile shaft and glans penis (Figure 1: A and B) for 1 month.

There was no eliciting history of drug intake/trauma/secondary manipulation/application of any local agents. No history of similar complaints in the partner. Nikolsky's sign was negative. Bulla spread sign was inconclusive. The patient was treated on the lines of

herpes genitalis on clinical grounds with valacyclovir with no response. Tzank smear failed to reveal findings consistent with herpes simplex infection. HSV 1 and 2 serology (IgM and IgG) were negative on 2 occasions a month apart. Syphilis, hepatitis, HIV were ruled on respective testing.



Figure 1: (A and B) Tense vesicles and bulla on the shaft of the penis and glans penis.

Histopathology sections revealed a partially re-epithelialized subepidermal blister (Figure 2) containing plasma and inflammatory cells including neutrophils and eosinophils. Epidermis forming the roof exhibited degenerative changes. There was perivascular and

interstitial infiltrate of lymphocytes and eosinophils in the upper dermis (Figure 3) underneath the blister. Cytopathic changes attributed to HSV were not seen in these sections. On clinical and histopathology grounds, a diagnosis of GBP was made, with differentials of linear IgA disease kept in mind.

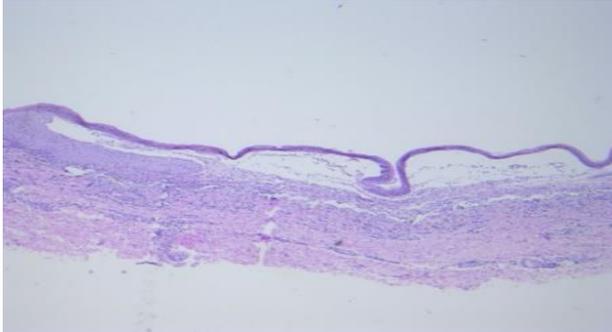


Figure 2: Subepidermal blistering (low magnification).

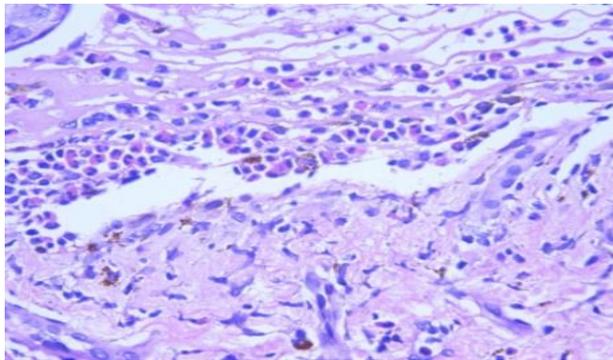


Figure 3: Eosinophilic rich infiltrate in the dermis (high magnification).

The patient was started on oral dapsone (100 mg/day) after reviewing complete hemogram, liver function test and G6PD sufficiency and is currently under follow-up, with resolution of lesions on the shaft within a month of initiation.

DISCUSSION

Bullous pemphigoid may be categorized according to the age of onset, site of involvement or the known triggers. Classically, it presents with blistering, preceded in some by a pre-eruptive urticarial prodromal stage, in the body folds above 70 years of age with genital involvement in 10-30% cases. Lesions in childhood BP has shown to be limited to the genitals in 17% of cases remitting on treatment with topical steroids and/or sulphonamides. Thorough history and examination, histopathology, direct and indirect immunofluorescence studies and serology help confirm clinical diagnosis. BP has successfully been treated with topical and oral corticosteroids, immunosuppressive and immune-modulating agents like methotrexate, azathioprine, mycophenolate mofetil,

dapsone, doxycycline±niacinamide and biologics like rituximab, omalizumab and intravenous immunoglobulin, the choice being individualized to severity of the disease, patient's age, site of involvement and the presence of comorbidities.³

Genital localization of the disease as the primary presentation has been only reported in 4 adult men earlier, as per our research of the literature so far. Various differential diagnoses include anogenital viral/bacterial/fungal/parasitic infections, contact dermatitis, drug reactions, other erosive disorders like linear IgA disease, pemphigus, mucosal pemphigoid, dermatitis herpetiformis, epidermolysis bullosa acquisita, bullous lichen planus. GBP has shown to respond to either topical ultra-potent corticosteroids alone or in combination with tetracycline; oral corticosteroids, mycophenolate mofetil, and doxycycline in the few studies known.⁴⁻⁷ Dapsone, however, remained our choice of drug, in view of the COVID-19 (20) pandemic, BP severity and to spare the genital skin from topical steroid induced local side effects.

Limitations

Due to hesitation in taking the biopsy from the concerned site, the biopsy sample for DIF study was taken from lower abdomen, in vicinity to the site of involvement. However, the negative DIF result on this sample is inconclusive and cannot be accepted as truly negative because the sample was not from perilesional skin. (DIF from perilesional skin in a case of bullous pemphigoid reveals linear deposition of IgG and C3 at the basement membrane zone.)³

CONCLUSION

Localized subgroup of bullous pemphigoid is less commonly encountered and hence, deferring its recognition from the time of presentation. Keeping it in mind as a differential and initiating treatment at the earliest may prevent it from evolving into a generalized form; save unnecessary investigations, distress, structural disfigurement and cost to the patient.

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