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Case Report

CO EXISTENCE OF LOCALIZED BULLOUS PEMPHIGOID & VITILIGO – A RARE CASE REPORT

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A R T I C L E I N F O

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A B S T R A C T

Background: Bullous pemphigoid and vitiligo are both autoimmune skin disorders, but their coexistence is extremely rare. Around 16% to 29% of bullous pemphigoid, are of Localized type. **Case report**: A 73 year old male farmer presented with a spontaneously appearing bullous eruption localized to both thighs, and complaints of intense pruritus for 3 weeks duration. He was also a known case of vitiligo who underwent PUVA therapy 10 years back. On examination, there were tense vesicles and bullae present over vitiligo patches and normal skin over posteromedial aspect of bilateral thighs. Clinical, histopathological, immunofluorescence findings were suggestive of Localized bullous pemphigoid. **Conclusion:** This case was unique because of rare coexistence of localized bullous pemphigoid and vitiligo, few bulla developed on preexisting vitiliginous regions, excellent outcome with topical therapy. Bullous pemphigoid is a great mimicker, hence it is mandatory to do skin biopsy, DIF and circulating autoantibodies detection in doubtful cases; for prevention and better prognosis of the disease.

INTRODUCTION

Localized bullous pemphigoid is a rare autoimmune subepidermal blistering disorder, characterized by chronic intermittent eruptions affecting only a defined area of the body. It is more common in elderly. Though it accounts for 16% to 29% of all cases of bullous pemphigoid, the true incidence may be higher as it is often misdiagnosed.[1]

Case report

A 73 year old male farmer presented with a spontaneously appearing bullous eruption localized to both thighs, and complaints of intense pruritus for 3 weeks duration. He was also a known case of vitiligo who underwent PUVA therapy 10 years back. On examination, there were tense vesicles and bullae present over vitiligo patches and normal skin over posteromedial aspect of bilateral thighs (Fig1,2,3). Diffuse hyperpigmented patches with islands of depigmentation & repigmentation present over scalp, trunk, upper limbs and lower limbs (Fig 3,4,5). Differential diagnosis are contact dermatitis, irritative dermatitis, viral infections like varicella-zoster or herpes and other bullous illnesses. Routine haematological and biochemical tests showed normal values. Tzanck test showed few eosinophils and no acantholytic cells seen.

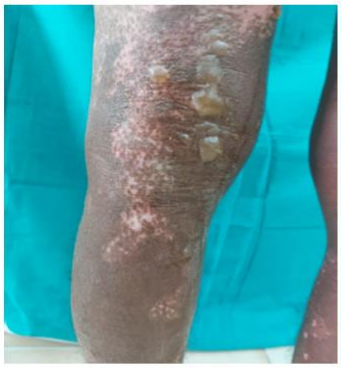

Fig 1

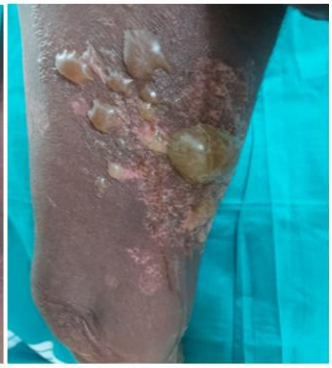

Fig 2

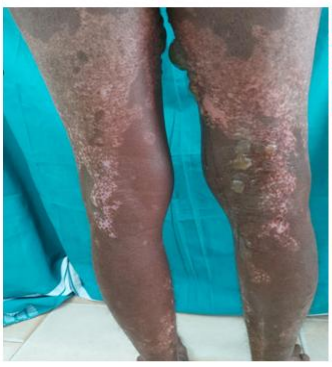

Fig 3

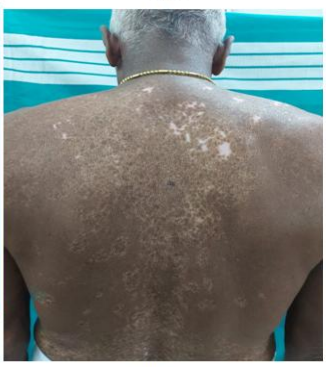

Fig 4

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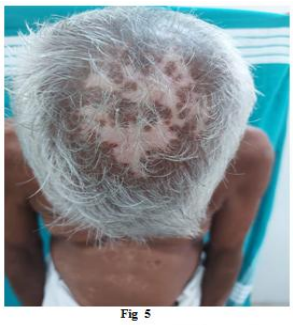
Fig 5

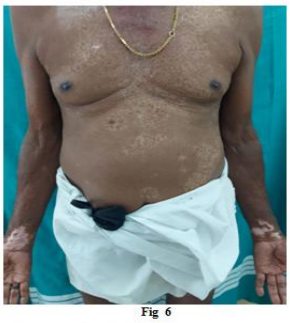
Fig 6

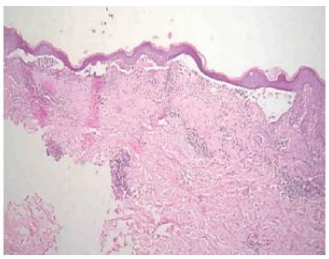
Fig 7

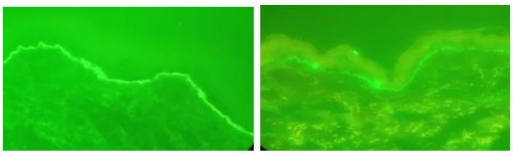
Fig 8

Thyroid function tests were normal; anti-thyroid antibodies and Anti-nuclear antibodies (ANA) were negative. Skin biopsy from blister over normal skin revealed "Subepidermal bulla with fibrin & eosinophils in blister cavity; superficial dermis showing edema, perivascular eosinophil rich infiltrate" (Fig7). Direct immunofluorescence assay of perilesional skin showed linear deposits of IgG and C3 along the basement membrane zone (Fig8) and salt spilt DIF showed IgG on epidermal side of spilt skin. Clinical, histopathological, immunofluorescence findings were suggestive of localized bullous pemphigoid. Patient was started on topical steroids and supportive care. He responded well to this regimen and disease was under control. No recurrence in 6 months follow-up period.

DISCUSSION

Vitiligo is a autoimmune disorder characterized by the destruction of epidermal melanocytes. Other autoimmune disorders like alopecia areata and thyroid disorders are well known to coexist with vitiligo[2]. Only few cases of coexistence of vitiligo and bullous pemphigoid especially localized bullous pemphigoid have been reported till date. The significance of the association of bullous pemphigoid with other autoimmune diseases is still unknown.[3]

Three types of localized bullous pemphigoid are: a) mucous membrane pemphigoid (cicatricial) pemphigoid 2) localized scarring pemphigoid (Brunsting Perry) pemphigoid- affecting head and neck 3) localized non scarring pemphigoid- affecting pretibial region, vulva, breast and the soles [1].

Risk factors of localized bullous pemphigoid include old age, frequent friction, sheer stress, local trauma, burns, radiation, hydrostatic forces and topical medicine[4]. The blister formation could be due to direct mechanical injury of skin, exposing the antigens of the basement membrane zone (normally hidden to the immune system). This induces antibody formation or precipitates bullous pemphigoid in subclinical cases (with low titres of epidermal autoantibodies) [5]. In a study, around 15% of cases, had no precipitating factor of bullous pemphigoid or localized bullous pemphigoid.

In another study on the distribution of bullous pemphigoid antigens BPAG1/ BPAG2 in normal human skin, antigen expression was highest over flexor aspect of arms, legs and thighs. This probably explains the localization of lesions ours thighs in our case.[7]

Localized bullous pemphigoid respond well to topical therapy, hence the prognosis is better than generalized form of bullous pemphigoid. Though localized bullous pemphigoid has a more benign disease course, but need long-term follow-up is important because of risk of developing generalized bullous pemphigoid.

CONCLUSION

This case was unique because of rare coexistence of localized bullous pemphigoid and vitiligo, few bulla developed on preexisting vitiliginous regions, excellent outcome with topical therapy. Most of the cases of Localized bullous pemphigoid are misdiagnosed and a defined set of risk factors for localized bullous pemphigoid is not available. Bullous pemphigoid is a great mimicker, hence it is mandatory to do skin biopsy, DIF and circulating autoantibodies detection in doubtful cases; for prevention and better prognosis of the disease..

References

1. Mehta V, Balachandran C. Localized flexural bullous pemphigoid. Indian J Dermatol 2008; 53:157-8. Vandana Mehta**,** C Balachandran Department of Skin and STD, Kasturba Medical College, Manipal, Karnataka, India
2. Riyaz N, Nasir N, Bindu V, Sasidharanpillai S. Bullous pemphigoid induced by topical PUVASOL. Indian J Dermatol Venereol Leprol 2014;80:363-364.Najeeba Riyaz Department of Dermatology and Venereology, Government Medical College, Kozhikode - 673 008, Kerala India
3. Andrés Tirado-Sánchez ,Griselda Montes-de-Oca. Coexistence of bullous pemphigoid, vitiligo, and thyroid disease: a multiple autoimmune syndrome?**.** Dermatol Online J. 2005 Aug 1; 11(2):20. Department of Dermatology, General Hospital of Mexico, DF, Mexico
4. Chloe Algoet, Sabine Mostinckx,Ivan Theate,Olivier Vanhooteghem. Localized bullous pemphigoid: Four clinical cases and a literature review.Wiley online library. 11 February 2020
5. Rakvit, Pariyawan & Kerr, Alastair & Ibbotson, Sally. (2011). Localized bullous pemphigoid induced by photodynamic therapy. Photodermatology, photoimmunology & photomedicine. 27. 251-3. 10.1111/j.1600-0781.2011.00609.x.
6. V.A Venning,F. Wojnarowska. Induced bullous pemphigoid. Br J Dermatol 1995 May;132(5):831-2
7. David J. Goldberg, M.D., Michael Sabolinski, M.S., and Jean-Claude Bystryn. Regional variation in the expression of bullous pemphigoid antigen and location of lesions in bullous pemphigoid. tile journal of investigative dermatology, vol. 82, no.4 82:326- 328, 1984