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Case Report
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A Geriatric Patient with Bullous Pemphigoid Limited to a Surgical Intervention Area

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Abstract

Background: Bullous Pemphigoid (BP) is uncommon, but becomes more frequent at older age (mainly >70 y).

Case Report: We report a 78-year-old woman who developed blisters, limited to the site of an orthopedic surgical area, occurring one week after the intervention. BP was confirmed by histology but immunofluorescence studies were negative. Risk factors for BP were old age, and the use of a loop diuretic and a dipeptidyl peptidase-4 inhibitor. The course was benign with a good response upon topical steroid therapy.

Conclusion: In the literature, localized cases of BP are reported that occurred, either around surgical wounds, or after topical iodine application. In our patient, the operation area had been disinfected with Isobetadine[R] (Povidone-Iodine hydroalcoholic solution 5%) and had been protected with Opsite[R] adhesive film.

Keywords: Bullous Pemphigoid; Immunofluorescence; Eosinophils

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Case Presentation

A 78-year-old woman, treated for ischemic cardiomyopathy and arterial hypertension (bumetanide, isosorbide, molsidomine, candesartan), atrium fibrillation (acenocoumarol, digoxin, bisoprolol), and type 2 diabetes (sitagliptin, metformin, gliquidon, insulin-glargine) was hospitalised for a hip fracture. Preparing the orthopaedic operation area, the skin was disinfected with Isobetadine[R] (Povidone-Iodine hydro-alcoholic solution 5%) and covered with Opsite[R] adhesive film. After the intervention (dynamic hip screw), her medication schedule was changed and she also received cefacidal, paracetamol, tramadol, furosemide, amlodipine, aspirin, pantoprazole, simvastatin, and rivaroxaban. Seven days after the intervention, itching erosions and blisters emerged around the skin of the operation area, increasing during the next week (Figure 1A). A skin biopsy in this area showed lymphocytic inflammatory infiltration of the dermis, both interstitial and perivascular, with pronounced presence of eosinophils; although immunofluorescence staining (IgG, IgM, IgA, and C3) did not reveal any precipitations, the biopsy was considered as typical for Bullous Pemphigoid (BP). Blood examination revealed eosinophilia (0.744/µL; 7.1%) and hypogammaglobulinemia (6.6 g/L). Treatment with a topical corticosteroid (mometason 0.1%), started after the biopsy, resulted within a few days in a marked improvement (Figure 1B). One month later, all lesions had resolved, the skin remaining slightly red and hyper pigmented.

Discussion

Pemphigoid diseases are characterized by generalized, itching skin erosions and blisters, due to the presence of autoantibodies against components of the epidermal-dermal junction (mainly BP230; BP180). Of this heterogeneous group, BP is the most common variety, occurring mainly above the age of 70, with an incidence of 1.2 to 2.7/10^5/year. Over the age of 80, the incidence rises to 31.2/10^5/year [1,2]. BP is observed more and more frequently, and, given the worldwide aging of the population, its frequency is expected to rise further. Besides age, mainly neurological diseases are associated with BP such as stroke, dementia, Parkinson's disease, and epilepsy [1,2]. Often these neurologic diseases precede BP, which is explained by the presence of analogous neuronal antigens that might circulate and provoke cross-reactivity in the skin. Other risk factors are drugs,

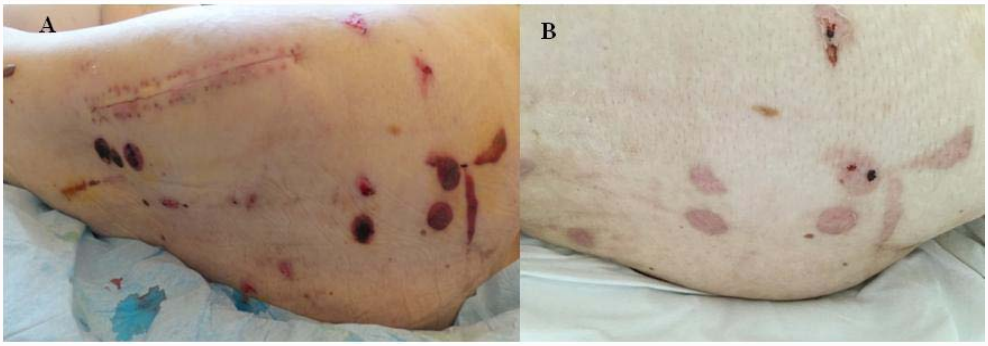

Figure 1: Localized Bullous Pemphigoid of the left hip. A) 14 days after surgery; B) after 7 days of topical corticoid treatment.

either for systemic (loop diuretics, anti-hypertension drugs, NSAIDs, neuroleptics, dipeptidyl-peptidase-4 inhibitors, mainly vildagliptin) or topical use (insect repellents, antitumor ointments).

Localized BP is rarer and has been observed mainly on the lower extremities, sometimes in the aftermath (days to months) and at the site of traumatism, surgical intervention, radiotherapy, or after local application of substances [3,4]. Only rare reports exist on the occurrence of BP after application of Iodine-containing skin treatments [5].

The diagnosis of BP is confirmed by skin biopsy, typically showing splitting in the subepidermal region, and inflammatory infiltration of the dermis by lymphocytes, neutrophils and abundant eosinophils; immunofluorescence examination shows immunoglobulin and complement-C3 precipitation [1,2]. In 10% of the cases, however, immunofluorescence examination is negative, as in our patient. One review reported that localized BP specimen were more often negative for indirect immunofluorescence [3]. In our case, light microscopy, especially the presence of intra-dermal eosinophilic inflammation, pleads in favor of BP. Possibly, the rapid eruption after the localized stimulus (trauma, Iodine, and/or adhesive film), together with the hypogammaglobulinemia, might have resulted in eosinophilic inflammation without auto-immunity.

The detection of circulating anti-BP180/BP230 (by ELISA), recommended as complementary diagnostic tool with a sensitivity of 90%, was not available for our case.

BP is associated with an increased risk for pneumonia and pulmonary embolism, and with higher mortality. Potent topical corticosteroids were shown to be the treatment of choice for BP. Our case illustrates that localized BP reacts well to topical treatment [1,2].

To date, a comprehensive review of localized BP and of BP-inducing physical factors does not exist. With this case-report, we aimed to bring these two aspects under attention.

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