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(CASE REPORT)

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Twice-triggered localized bullous pemphigoid following orthopedic surgery: A rare presentation

Hajar. Marghadi [1, 2, *], Kaoutar. Elmachichi [1, 2], Maryem. Aboudourib [1, 2], Ouafae.Hocar [1, 2] and Said Amal [1, 2]

[1] *Department of Dermatology, University Hospital Mohammed VI, Marrakech, Morocco.*
[2] *Biosciences and Health Laboratory, Cadi Ayyad University, Marrakech, Morocco.*

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Abstract

Localized bullous pemphigoid (BP) is a rare variant of an autoimmune blistering disease commonly affecting elderly individuals. While BP is typically associated with systemic triggers, its localized form can result from physical trauma or surgical procedures. We present a case of a 60-year-old man who developed localized BP following two orthopedic surgeries on the same limb. Two weeks after femur fracture fixation with retrograde intramedullary nailing, he developed tense, pruritic bullae around the knee and lower leg. Histopathological analysis and direct immunofluorescence confirmed the diagnosis of BP. Lesions resolved after treatment with oral corticosteroids. However, the patient experienced a recurrence of similar lesions after a second procedure for bone grafting due to fracture non-union. The lesions extended above the knee but responded to the same corticosteroid regimen, resolving completely. This case highlights the potential role of surgical trauma and postoperative tissue changes as triggers for localized BP, emphasizing the importance of early recognition and management.

Keywords: Localized Bullous pemphigoide; orthopedic surgery; BP180/BP230 autoantibodies; autoimmune blistering disease

1. Introduction

Bullous pemphigoid (BP) typically affects elderly individuals and is known for its widespread, itchy, tense blisters. However, localized bullous pemphigoid is a rarer variant and can be easily misdiagnosed due to its limited presentation. This case report describes an unusual recurrence of localized BP following two separate orthopedic surgeries in the same patient, emphasizing the importance of recognizing surgical trauma as a potential trigger.

2. Case Presentation

We report the case of a 60-year-old man who underwent orthopedic surgery for a right femur fracture, treated with retrograde intramedullary nailing. Two weeks post-surgery, he developed itchy, tense bullae localized around the right knee and lower leg. Histological examination of a skin biopsy revealed subepidermal blisters with eosinophilic infiltration, and linear IgG and C3 deposits along the basement membrane zone on direct immunofluorescence. These findings confirmed the diagnosis of localized bullous pemphigoid.

The patient was started on oral prednisone (0.5 mg/kg/day), which led to complete resolution of lesions within three months. However, due to a non-union of the fracture, he later underwent a bone graft procedure. Remarkably, just days after the second surgery, similar blisters reappeared, this time extending around the right thigh and knee. The same corticosteroid regimen was resumed, resulting in resolution within two months.

* Corresponding author: H Marghadi

3. Discussion

While bullous pemphigoid is well-recognized, its localized variant is less common and often associated with trauma, burns, phototherapy, or radiation. Post-surgical BP is exceedingly rare, and recurrent episodes after multiple surgeries in the same anatomical region are even more exceptional. It is thought that tissue damage interferes with the humoral mediated immunity, causing alteration of BP180 and BP230 antigens

In this case, the recurrence of BP after two separate procedures strongly supports a link between surgical trauma and disease onset. Although koebnerization—the appearance of skin lesions at sites of trauma—has been proposed as a mechanism, we suggest that localized edema following surgery may have acted as the immunological trigger. This hypothesis is supported by the distribution of blisters beyond the immediate surgical site.

To our knowledge, this is only the second reported case of recurrent localized BP triggered by repeated surgical interventions in the same patient. Dermatologists and surgeons should be aware of this possibility to ensure timely diagnosis and treatment.

Figure 1 Multiple post-bullous erosions on the lower limb

4. Conclusion

This rare case illustrates how orthopedic surgery may trigger localized bullous pemphigoid, even more unusually on two separate occasions. It highlights the importance of maintaining clinical suspicion in postoperative settings, particularly in elderly patients presenting with localized blistering eruptions.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

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