Scar Site Bullous Pemphigoid: A Rare Presentation and a Clinician Dilemma

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Received: August 11, 2022, Manuscript No. DMCR-22-18818; Editor assigned: August 15, 2022, PreQC No. DMCR-22-18818 (PQ); Reviewed: August 29, 2022, QC No. DMCR-22-18818; Revised: December 23, 2022, Manuscript No. DMCR-22-18818 (R); Published: January 03, 2022, DOI: 10.4172/2684-124X.8.1.001

Abstract

Bullous pemphigoid is the most common autoimmune subepidermal blistering disease. Its onset is usually after 60 years of age. Characteristically, BP is an intensely pruritic eruption with widespread blister formation. However, in our case, patient characteristically developed lesions on 15th day post total knee replacement surgery and localized around surgical site. This indicates, physical trauma in form of surgical site incision as trigger. Localized BP is rare in itself and can be confused with irritant contact dermatitis, allergic contact dermatitis, and bullous cellulitis. Early recognition of this entity is important so as to determine prognosis and course of disease.

Keywords: Bullous pemphigoid · Subepidermal · Trigger

Introduction

India is a country with population of almost 1.2 billion people. Number of people going for knee arthroplasty in year 2020 was around 2,00,000. It's one of the safest orthopedic operations with high success rate. Trend is on rise due to bell shaped population curve of INDIA. 74% of total TKRs being done in India are posterior stabilized as compared to cruciate retaining implants 11% and Remaining 15% were rotating platform, constrained knees (e.g. LCCK, RHK from Zimmer and TC 3, Noiles Hinge from dePuy J and J).

Common skin complication post TKR includes post operation wound infections, blistering as a result of soft tissue swelling, allergic reactions to dressing material or topical agents. [1] .

Post-surgical occurrence of bullous pemphigoid is a rare but, known entity. However, post TKR only 5 case reports are reported in literature and this being $6^{\rm th}$.

Case Presentation

A 64 years old female, known case of diabetes mellitus and on Teneligliptin 20 mg and Metformin 500 mg 1 BD. Patient underwent total knee arthroplasty with tibial insert fixed bearing curved plus, tibial tray fixed bearing modular titanium cemented, femoral cruciate retaining cemented (all from Depuy, Johnson and Johnson). TKR itself was uneventful and patient was discharged after 24

hours. TKR relieved off patients' knee pain and range of motion gradually increased up to 95 degrees by day 14.

During, stapler removal on day 15th, patient noticed a small single papular eruption in scar line, which was slight pruritic. Treating doctor (orthopaedic) was consulted and cleaning with betadiene 10% was advised. 3 days post suture removal (18th day post OP), patient noticed large number of bullous eruption in and around scar line (more marked centrally) (Figure 1). Bulla was tense, intensely itchy and ruptured to form erosions. For this dermatologist was consulted later, who treated on line of bullous cellulitis and started tab LINID 600 mg 1 BD and supirocinoint tds for 7 days. However, no clinical improvement on lesion was seen. Antibiotic was changed to tab Rifampicin 600 mg 10 D on follow up for next 7 days, but new lesions kept on appearing. Meanwhile, patient was completely afebrile and good range of motion was present in operated knee (Figure 2). Patient was advised to stop dressing the lesion with butadiene 10% and was just asked to apply supirocin ointment.

Nikolsky was negative and intense itching probed us to point towards localized bullous pemphigoid. Afebrile patient, no pain and swelling at site ruled out cellulitis. Recurrent appearance of bulla, excluded irritant dermatitis [2].



Figure 1. Day 18th post op.

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Figure 2. Day 40th post op.

Investigation

Hemogram was performed to rule out developing cellulitis and it revealed total count of 8,000/cmm, DLC (neutrophils-65%, lymphocytes-40%, eosinophils-3%, basophils-0%), CRP-08 mg/L. Biopsy was not performed as consent was denied by patient in fear of secondary infection spreading to joints and risk implant infection.

Treatment

Patient was then, started on low dose deflazacort (12 mg) once daily in morning, cap Doxycycline 100 mg 1 BD, tab Niacinamide 250 mg 1 BD, tab Levocet 5 mg 1 BD, topical FUCIBET cream BD (betamethasone+fusidic acid) for 20 days to which patient responded drastically. No new lesions appeared post 20 days and itching subsided. Steroid was tapered and stopped in next 10 days followed by. No new lesions re-occurred. PIH and crusting at few site was seen on day 30th of treatment and no new lesion occurred. Pt was continued on tab Niacinamide 250 mg BD, Levocet 5 mg BD, FUCIBET HS application and moisturizer was added. Pt is under observation and is supposed to undergo TKR in right joint anytime soon. Word of caution is advised to patient regarding same.

Discussion

BP is most frequent auto-immune blistering disorder which is caused by auto antibodies against BP180 and BP230. Trigger factors include thermal or electrical burns, surgical procedures, trauma, ultraviolet irradiation, radiotherapy, chemical preparations, transplants, and infections. Drugs like thiazides, cephalosporins, ciprofloxacin, NSAID's are common association.

There are only three cases reported in literature with such acute onset. Kim et al. described a case of generalized BP that started 3 days after surgery and spread to the operative knee 1 week later [3]. S. Sudah, et al. [4] described a case of localized BP that started on day 14 and spread to medial side of operated knee on day 17th. Truss, et al. [5] described in which localized BP appeared and spread in 4 days.

In working up for diagnosis in a patient with bullae or vesicles appearing after a TKA, it is of prime importance to consider a complete differential diagnosis, including, postoperative skin blistering secondary to soft tissue edema post operation, allergic or irritant contact dermatitis, bullous cellulitis and localized bullous pemphigoid. Post op wound infection can be devastating as it can put risk of implant failure. It can be ruled out as it will have pus discharge, redness, swelling occasional bullae and patient will be febrile. Other cause of acute bullous eruption is ACD/ICD due to bandage, dressing material or topical agents used for dressing. This presents as pruritic, bullous eruption. However, removal of culprit agent and changing dressing material should result in complete remission. This was not so in our case, as despite ceasing dressing, bullous eruptions continued.

Occurrence of bullous pemphigoid post trauma and surgery is being linked with "theory of immune compromised district" which states that, tissue damage interfering with local immune and cutaneous processes, leaving the potential for secondary disorders such as BP to occur [6,7].

Conclusion

This case, educates clinicians (dermatologist and orthopaedic) to be aware of this rare entity which can hinder proper patient care. Timely diagnosis can improve patients prognosis and reduce psychological trauma to patients.

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