Spinal Anesthesia in a Healthy Parturient Causing Suspected Bullous Pemphigoid Lesions: A Case Report

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Case Report

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Abstract

We present a case of suspected bullous pemphigoid lesions after spinal anesthesia in a previously healthy parturient presenting at term for an elective secondary cesarean section. On day 1 post-partum, pruritic red fluid-filled clustered bullae highly resembling those of bullous pemphigoid were noted around the spinal anesthesia injection site. Fusidic acid/betamethasone skin cream was prescribed by a dermatologist for twice-daily application. The bullae decreased in size gradually and became crusted 1 week post-partum with mild residual pruritus. At 6 weeks post-partum, the bullae completely disappeared without a scar. Checking the site of neuraxial blockade on the first day postoperatively allowed early detection of such unique skin lesions.

Background

During pregnancy, bullae can occur secondary to either common skin disorders or specific dermatoses of pregnancy. A heterogenous group of pregnancy or postpartum related dermatoses include, however is not limited to, pemphigoid gestationis, polymorphic eruption of pregnancy, dermatitis herpetiformis, erythema multiforme, and allergic contact dermatitis among others.

An accurate understanding of these conditions allows for proper management and follow-up of the patient. Some conditions have been associated with adverse obstetric outcomes, such as increased risk of prematurity and small-for-gestational-age babies seen in fetuses born to patients with pemphigoid gestationis, putting emphasis on the importance of making the correct diagnosis.1

Although pregnancy induces an immunosuppressive state, a prevalent T-helper 2 (Th2) cellular profile can lead to an aggravation of bullous pemphigoid especially during the first and second trimesters. However, in the third trimester, the clinical picture can improve due to increased production of endogenous corticosteroids by the chorion.2

We did an up-to-date review of all previous cases in the field trying to find a relationship between bullous pemphigoid-like lesions, pregnancy, and regional or neuraxial anesthesia, but we didn’t find such similar case presentations.

Case Presentation

A 31-year-old female, gravida 2, para 1, with a second pregnancy complicated by gestational hypertension and polyhydramnios, presented at 37 weeks and 3 days of gestational age for an elective secondary cesarean section.

She was previously healthy with no known allergies and no reported personal or family history of dermatologic or autoimmune diseases.
Due to her obstetric history of gestational hypertension in the prior pregnancy, she was maintained throughout the current pregnancy on oral aspirin, 100 mg once daily, which was discontinued at 36 weeks of gestational age.

She presented to the delivery suite on her scheduled day and received the usual preoperative anesthesia assessment and obstetric care. She was consented for spinal anesthesia. In the operating room and under sterile technique, scrubbing and draping of her back was done with 2% chlorhexidine gluconate (CHG) / 70% isopropyl alcohol (IPA) swabs and sterile drapes, respectively. Afterwards, the skin was infiltrated with 4 mL of 1% lidocaine. Then, using a 25-G Whitacre spinal needle, 12.5 mg of hyperbaric bupivacaine, 12.5 mcg of fentanyl, and 0.1 mg of intrathecal morphine were injected intrathecally, after CSF aspiration, at the level of L3-L4 intervertebral space at a needle depth of 5 cm, without inflicted skin trauma. The skin at the puncture site was then covered by a sterile latex-free transparent film dressing (3M Tegaderm™ Film). A spinal level was confirmed at the T6 dermatome. The patient tolerated the cesarean delivery very well. She left the operating room in a stable condition. Following two hours of close postoperative observation in the delivery suite, the patient was safely transferred to the postpartum ward.

On day 1 post-partum and upon general assessment by the nurse in charge, pruritic red fluid-filled clustered bullae, highly resembling those of bullous pemphigoid, were noted at the area around the spinal anesthesia injection site (Fig. 1). Upon further assessment by the medical team, no similar or other skin lesions were noted on the rest of her body. She remained afebrile with stable vital signs. She denied other associated symptoms such as pain, burning sensation, or tenderness upon palpation of her skin lesions. Her serum white blood cell (WBC) count increased from a preoperative value of 10,400/µL to 16,300/µL, consistent with normal post-partum laboratory findings.

On day 2 post-partum, no major changes occurred in her skin lesions except for a mild increase in their size. Upon consultation, the dermatologist initially described bullous pemphigoid-like lesions based on their clinical appearance, and thus requested skin biopsy and serologic tests for a definitive diagnosis. The patient refused to do so due to her fear that a skin biopsy, tissue cultivation, or further needle pricks for serologic tests might cause the eruption of more of her pruritic bullae. An infectious cause of her lesions was also suspected. Herpes Zoster was low on the differential due to the non-painful nature and the non-dermatomal distribution of her lesions. The possibility of a rare skin reaction to lidocaine infiltration was also low on the differential since the patient already had received a lidocaine skin infiltration at the dorsum of her hand during the insertion of her peripheral intravenous catheter, which was covered by the same sterile latex-free transparent film dressing (3M Tegaderm™ Film), and no such skin lesions were noticed. Also, no such skin lesions were noticed 3 years ago at the spinal anesthesia injection site after her first cesarean delivery.

Fusidic acid/betamethasone skin cream was prescribed by the dermatologist for a twice-daily application with close follow-up. The patient was discharged home on day 3 post-partum to continue fusidic
acid/betamethasone skin cream application for a total of 10 days. After one week, follow up examination showed that her bullae decreased in size and became crusted with mild residual pruritus.

Two weeks later, the patient reported sloughing of her bullae with no pruritus. However, a persistent brownish skin discoloration at the bullae site was noticed (Fig. 2). Six weeks post-partum, her skin lesions completely disappeared with no persistent scarring.

**Discussion And Conclusions**

Bullous pemphigoid is an acquired, chronic, blistering, autoimmune, subepidermal bullous disease in which autoantibodies are directed against components of the basement membrane zone of the skin. It is characterized by formation of bullae on the skin and mucous membranes. The pathogenesis involves migration of inflammatory cells into the subepithelial tissues due to activation of complements caused by antigen-antibody reaction. The incidence of bullous pemphigoid is 6–7 cases per million population per year in the western world. It usually involves elderly more than 60 years of age and is rare in children, although childhood bullous pemphigoid has been described. One of the variants of the disease is cicatricial pemphigoid, which commonly involves mucous membranes of the oropharynx, conjunctiva, nasopharynx, larynx, esophagus, genitalia, and anus. Bullous eruptions are usually followed by scarring.

The clinical course of the disease is one of exacerbations and remissions. The diagnosis is based on clinical presentation, histopathological analysis, direct and indirect immunofluorescence microscopy on perilesional skin, analysis of staining patterns, and characterization of circulating autoantibodies. There have been several reports of its association with other autoimmune skin bullous diseases like pemphigus, pemphigoid, epidermolysis bullosa acquisita, dermatitis herpetiformis (Duhring disease), linear immunoglobulin-A disease, and multiple autoimmune syndrome. The relationship between bullous pemphigoid and malignancy is a matter of debate. Despite several published case reports, there is no definite association. Ogawa et al., found a significantly higher incidence of malignancy in bullous pemphigoid patients in Japan. Other authors from other countries in Asia have also reported a higher incidence of malignancy in these patients; however other studies done on Caucasian patients have failed to prove any statistically significant association. Treatment is with corticosteroids, immunosuppressive agents, and occasionally antibiotics if an associated secondary bacterial infection develops.

There are no definite recommendations for either general or neuraxial anesthesia in bullous pemphigoid patients. Use of neuraxial anesthesia in these patients is controversial, the reason being that blister formation can occur at the site of needle insertion. In case of epidural anesthesia, fixation of the epidural catheter on the skin may be difficult. However, neuraxial and regional anesthesia have been described for surgeries in these patients, as it avoids general anesthesia and airway instrumentation. The use of adhesive tape to secure the epidural catheter to an area of skin should be avoided. Spinal anesthesia has been described for cesarean sections while avoiding local anesthetic skin infiltration at the site of spinal
needle insertion because of the risk of bullae formation and skin sloughing.\textsuperscript{12} In case of spinal needle insertion, an area devoid of skin lesions should be chosen. Also, skin infection at the site of local anesthetic injection is possible, which may lead to subsequent sepsis. A choice of an area devoid of skin lesions is thus recommended and considered safe for performing spinal anesthesia.\textsuperscript{12} Several published case reports showed that a single shot spinal anesthetic is safe for cesarean sections, however general anesthesia is not contraindicated if the oral mucosa is not involved with bullae.\textsuperscript{12} Neuraxial opioids, especially intrathecal morphine, are preferably avoided in these patients as they are associated with pruritus.\textsuperscript{13}

Much is yet to be determined in terms of the association between spinal anesthesia, pregnancy, and bullous skin diseases. Our patient did not have any personal or family history of dermatologic or autoimmune diseases. She had previously received spinal anesthesia with lidocaine skin infiltration during her first cesarean section without such unique complication. One limitation to her case is the lack of diagnostic skin biopsy, tissue cultivation, or serologic tests for circulating autoantibodies confirming the diagnosis of bullous pemphigoid. Yet, another explanation for these post spinal anesthetic bullae could have been a rare local skin reaction to lidocaine.

In our opinion, it would be wise for all anesthesiologists to check the site of neuraxial block on the first day postoperatively, before discharging the patient. This can allow the early detection of such unique skin lesions, be it self-limiting or serious.

If a parturient patient presents with similar skin lesions after spinal anesthesia and has no known dermatologic or autoimmune diseases, we suggest expectant management of her skin lesions with close observation of any new onset signs and symptoms.

**Abbreviations**

Th2
T-Helper 2.

**Declarations**

- Ethics approval and consent to participate: The American University of Beirut Medical Center (AUBMC) Institutional Review Board (IRB) does not require its approval for the involvement of human participants in the publication of case reports.
- Consent for publication: Written informed consent was obtained from the patient for the publication of this case report.
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References

Figures

Figure 1

Spinal anesthesia injection site at day 1 post-partum.
Figure 2

Spinal anesthesia injection site at 2 weeks post-partum.