Case Report/*Olgu Sunumu*

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Spinal Anesthesia to Avoid Airway Manipulation in Epidermolysis Bullosa: A Case Report

Epidermolizis Büllozada Hava Yolu Manipülasyonundan Kaçınmak İçin Spinal Anestezi: Olgu Sunumu

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ABSTRACT

Epidermolysis bullosa (EB) is a rare group of inherited disorders characterized by mucocutaneous blistering after minimal trauma; in dystrophic subtypes, squamous cell carcinoma (SCC) arising from chronic wounds is a serious complication. We report spinal anesthesia for wound biopsy in a 22-year-old woman with EB and SCC receiving chemotherapy, in whom a previously reported laryngeal mask airway-related airway event shaped the anesthetic plan. As the patient reported prior airway swelling and declined general anesthesia, airway instrumentation was avoided. Spinal anesthesia was performed in the sitting position at L4-5 with a 25 G Quincke needle and intrathecal 10 mg hyperbaric bupivacaine plus 10 µg fentanyl; no sedation was given, and the patient breathed room air spontaneously. Skin-preserving modifications were applied: dabbing antisepsis without friction, omission of electrodes to avoid epidermal stripping, and placement of the blood pressure cuff over petrolatum-impregnated padding, while pulse oximetry and intermittent noninvasive blood pressure were maintained. With a T8 sensory level, back and foot biopsies were completed within two hours without vasopressors; hemodynamics remained stable, and no new blisters were noted at 24 hours. In selected adult EB patients with a complicated airway history, spinal anesthesia may be an alternative to general anesthesia.

Keywords: Epidermolysis bullosa, neuraxial anesthesia, perioperative management, spinal anesthesia, squamous cell carcinoma

ÖZ

Epidermolizis bülloza (EB), minimal travma sonrası mukokutanöz bül oluşumuyla karakterize, nadir görülen kalıtsal bir hastalık grubudur; distrofik alt tiplerde kronik yaralardan köken alan skuamöz hücreli karsinom (SHK) ciddi bir komplikasyondur. Kemoterapi alan, EB ve SHK tanılı 22 yaşında bir kadın hastada yara biyopsisi için uygulanan spinal anesteziyi sunuyoruz; bu hastada daha önce bildirilen laringeal maske ilişkili bir hava yolu olayı anestezi planını şekillendirdi. Hasta önceki hava yolu şişliğini ifade ettiği ve genel anesteziyi reddettiği için hava yolu girişiminden kaçınıldı. Spinal anestezi oturur pozisyonda L4-5 aralığından 25 G Quincke iğnesi ile, intratekal 10 mg hiperbarik bupivakain ve 10 µg fentanil verilerek uygulandı; sedasyon uygulanmadı ve hasta oda havasında spontan soludu. Cilt koruyucu modifikasyonlar uygulandı: sürtme olmadan dokundurma tekniğiyle antisepsi, epidermal soyulmadan kaçınmak için elektrokardiyografi elektrotlarının takılmaması ve kan basıncı manşonunun petrolatum emdirilmiş ped üzerine yerleştirilmesi; bu sırada pulse oksimetre ve aralıklı noninvaziv kan basıncı izlemi sürdürüldü. T8 duyusal blok düzeyiyle sırt ve ayak biyopsileri vazopresör gerekmeksizin iki saat içinde tamamlandı; hemodinami stabil seyretti ve 24. saatte yeni bül izlenmedi. Komplike hava yolu öyküsü olan seçilmiş erişkin EB hastalarında spinal anestezi, genel anesteziye alternatif olabilir.

Anahtar sözcükler: Epidermolizis bülloza, nöroaksiyel anestezi, perioperatif yönetim, skuamöz hücreli karsinom, spinal anestezi

INTRODUCTION

Epidermolysis bullosa (EB) comprises a rare group of genetic disorders caused by loss-of-function mutations in structural proteins of the dermal-epidermal junction, producing extreme skin fragility after minimal trauma. In severe subtypes such as recessive dystrophic EB, esophageal stricture and squamous cell carcinoma (SCC) arising from chronic wounds are leading causes of morbidity and mortality (1). Anesthetic management is challenging because of mucosal vulnerability to airway devices, the need to adapt monitoring and positioning, and impaired thermoregulation (2). Although neuraxial techniques are reported to be safe in adults, most published experience concerns the pediatric population (2,3).

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We describe spinal anesthesia for wound biopsy in an adult patient with EB-associated SCC in whom a self-reported prior laryngeal mask airway (LMA)-related airway event guided the decision to avoid all airway instrumentation. This case highlights the importance of individualized anesthetic planning, informed by previous airway complications and directed at minimizing skin and mucosal trauma.

CASE PRESENTATION

A 22-year-old woman had been followed for EB for many years; however, the subtype had never been confirmed by molecular or immunohistochemical testing. A large proportion of her body surface area was covered with active lesions and dressings. Her history included repeated esophageal stricture dilations, foot wound biopsy and resection procedures, and surgical separation procedures for pseudosyndactyly of the hands. Following a diagnosis of SCC, she was receiving chemotherapy via an implanted port. She had no other systemic disease, and preoperative laboratory values were within normal limits. Airway assessment revealed adequate mouth opening and a Mallampati class 2 score. According to the patient's own account, a previous general anesthetic with an LMA had been followed by tongue and pharyngeal swelling and a severe sore throat, which she recalled as a distressing and potentially serious event. This episode was not witnessed by our team, and its exact severity, duration, and management could not be independently verified. Nonetheless, this self-reported history, together with the patient's own refusal of general anesthesia, was the principal reason for avoiding airway instrumentation.

Given this history, an airway device was avoided and spinal anesthesia was planned. The operating room temperature was set to 23°C, and all contact surfaces, including the area beneath the blood pressure cuff, were covered with petrolatum-impregnated padding. Peripheral oxygen saturation was monitored continuously with a pulse oximeter. Electrocardiography electrodes were omitted to avoid epidermal stripping on removal of adhesive surfaces; the plethysmography waveform from the pulse oximeter, together with intermittent noninvasive blood pressure measurement, provided continuous surveillance of heart rate and rhythm. No sedation was administered. The patient breathed room air spontaneously throughout the procedure; no supplemental oxygen or accessory airway device, such as a nasal cannula, was used, consistent with the aim of avoiding unnecessary adhesive or device contact. The positioning decision was made jointly with the patient: because lateral positioning was expected to cause friction over the lesions on her back, the sitting position was chosen for less pressure and greater comfort (Figure 1).

Povidone-iodine antiseptic was applied using a dabbing technique without circular or rubbing motion. Spinal puncture was performed at the L4-5 interspace using a 25 G sharp-tipped (Quincke) needle (Egemen International, İzmir, Türkiye), and 10 mg hyperbaric bupivacaine plus 10 µg fentanyl were administered intrathecally. The puncture site was covered with non-adherent sterile gauze without tape. A sensory block level of T8 was confirmed.

Simultaneous wound biopsies of the back and foot took approximately two hours. Normothermia was maintained using an active warming device, and hemodynamics remained stable throughout; no vasopressors or additional systemic analgesics were required. The patient was discharged uneventfully, and no new blisters or skin breakdown were observed at the 24-hour follow-up. Skin was not systematically re-examined beyond 24 hours; although no new lesions were reported subsequently, delayed blistering after this window cannot be entirely excluded.



Figure 1. Intraoperative photograph of the patient in the sitting position during spinal anesthesia. Extensive erosions and denuded skin are visible over the lower back; the trunk and arms are covered with non-adherent dressings, and the noninvasive blood pressure cuff is applied over petrolatum-impregnated padding.

DISCUSSION

The central decision in this case was to avoid airway instrumentation altogether. Although less invasive than direct laryngoscopy, an LMA can still cause bullous injury of the pharyngeal mucosa through pressure and friction (2,3). Our patient reported a prior episode of tongue and pharyngeal edema with severe sore throat after general anesthesia with an LMA. Despite a reassuring airway examination (Mallampati 2, adequate mouth opening), this reported event was treated as decisive: in EB, a favorable static airway assessment does not exclude device-induced mucosal injury, and a prior complication should outweigh reassuring examination findings. Inoue et al. applied a comparable rationale in an adult EB patient with SCC undergoing lower-leg amputation, selecting peripheral nerve blocks specifically to avoid airway instrumentation in the setting of restricted mouth opening and anticipated postoperative airway compromise (4).

The risks of orotracheal intubation itself warrant comment. In EB, airway instruments that generate shear on the mucosa (including direct and video laryngoscopes and supraglottic devices) are regarded as capable of provoking mucosal bullae and are generally avoided; when general anesthesia is unavoidable, atraumatic fiberoptic intubation with a smaller, well-lubricated cuffed tube and low cuff pressure is preferred (2,3). Even gentle laryngoscopy therefore carries a real, if not inevitable, risk of mucosal injury, which further supported our preference for a technique that avoided airway manipulation altogether.

Spinal anesthesia was delivered with a bundle of simple, reproducible skin-preserving modifications. Antiseptic was applied by dabbing without friction; a fine 25 G sharp-tipped (Quincke) needle minimized tissue handling; the puncture site was secured with non-adherent gauze without tape; and the noninvasive blood pressure cuff was placed over petrolatum-impregnated padding. Electrocardiography electrodes were omitted to avoid epidermal stripping on removal of adhesives, while continuous pulse oximetry (including the plethysmography waveform) and intermittent noninvasive blood pressure provided adequate surveillance of heart rate and rhythm during a two-hour, hemodynamically stable anesthetic. Choosing the sitting position jointly with the patient, to minimize friction over the back lesions, illustrates how patient-reported comfort can be incorporated into technical decision-making in EB. Noninvasive blood pressure monitoring can likewise transmit shear to fragile skin; the accepted mitigation, used here, is a padded cuff combined with a clip-type oximeter probe rather than adhesive sensors (2). Invasive arterial monitoring is not recommended merely to avoid cuff-related trauma: it adds an arterial puncture and, because adhesive fixation is contraindicated in EB, would require non-adhesive or sutured securing, so it is reserved for a definite hemodynamic indication. For this short, superficial, and hemodynamically stable procedure, a padded noninvasive cuff was judged proportionate and lower-risk.

The EB subtype had never been confirmed molecularly or immunohistochemically. Nonetheless, recurrent esophageal stricture dilations, SCC arising from chronic wounds, and prior surgical release of hand pseudosyndactyly together indicate a phenotype consistent with dystrophic EB; pseudosyndactyly in particular is a robust marker of the dystrophic scarring tendency (1). Management was therefore guided by clinical phenotype and history rather than genotype. Mittal et al. similarly emphasize that general, neuraxial, and regional techniques can all be used safely in adults, provided planning is individualized to subtype and clinical findings (2).

This procedure formed part of the oncologic surveillance of an adult EB patient receiving chemotherapy for SCC, underscoring that anesthetic care is repeatedly required across the EB lifespan; this adult, oncologic context remains underrepresented relative to pediatric reports (3). In conclusion, in selected adult EB patients, particularly those with a complicated airway history, spinal anesthesia may be a suitable alternative to general anesthesia. Skin integrity can be preserved through simple modifications such as shared positioning decisions, non-adhesive materials, dabbing antisepsis, and selective monitoring, and an individualized, phenotype-based approach is appropriate when the EB subtype is unconfirmed (5).

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AUTHOR CONTRIBUTIONS

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Data collection: OKG, OOO
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The authors (OKG, OOO, ZB) reviewed the results and approved the final version of the manuscript.

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