CASE REPORT

Anesthetic management for right upper extremity amputation due to recidivous cutaneous carcinoma and acute postoperative pain control in patients affected by epidermolysis bullosa

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ABSTRACT

A 22-year-old male who was affected by epidermolysis bullosa (EB) and xeroderma pigmentosa (with structural and pathological changes that preclude orotracheal intubation) underwent right upper extremity amputation and ipsilateral axillary lymphadenectomy. The patient was operated without intubation, thereby assuring an optimal state of acute postoperative pain control by regional anesthesia. Intravenous administration of ketamine and remifentanil plus low-dose sevoflurane resulted in anesthesia with spontaneous breathing by the patient. Moreover, the intraoperative brachial plexus nerve block before amputation followed by positioning of an epidural catheter to deliver continuous infusion of local anesthetics close to the cut nerves during surgery obtained a very good level of acute postoperative pain control. *(Minerva Anestesiol 2010;76:144-7)*

Key words: Epidermolysis bullosa simplex - Ketamine - Postoperative care - Catheterization.

Epidermolysis bullosa (EB) is a rare genetic disease characterized by the presence of extremely fragile skin and recurrent blisters from mechanical friction or trauma.[1, 2]

This case report describes an anesthetic management based on intravenous administration of ketamine, remifentanil and low-dose sevoflurane in a spontaneously breathing EB patient who underwent amputation of the right upper extremity for recidivous cutaneous carcinoma. Moreover, the case also assesses the level of acute postoperative pain control by continuous infusion of local anesthetics through epidural catheters that were positioned close to the brachial plexus.

Case report

The patient was a 22-year-old male (weight: 60 kg and height: 165 cm) who was affected by EB and xeroderma pigmentosa. The patient had already undergone multiple operations for hand and finger unbridling and for removal of recidivous cutaneous spinocellular carcinoma from the upper extremities.

The patient was affected by pseudosyndactyly in the hands, which had previously been treated with microsurgery. All operations were performed using the following anesthetic techniques: brachial plexus anesthesia, deep sedation with fentanyl and ketamine, or with propofol plus local anesthetic infiltration.[2]

The patient presented to us for amputation of the right upper extremity at the humerus neck level (the constricted part of the proximal humerus below the greater tuberosity and the lesser tuberosity of humerus) as well as ipsilateral axillary lymphadenectomy due to recidivous spinocellular carcinoma with axillary lymphonodal metastases.

The mouth opening-inter-incisor distance was <1.5 cm, and the tongue showed a partial fixity in the oral cavity (Mallampati score=4). All attempts at orotracheal intubation had failed. Reduced buccal cavity opening would have prevented the use of the laryngeal mask airway (LMA). Furthermore, using the laryngoscope blade and fiberbronchoscope proba-

bly would have caused the formation of intra-oral bulla from any attempt to intubate. [3, 4] Moreover, intubation in this patient carried a high risk of causing arytenoideae edema. In order to consider the possibility of performing orotracheal intubation on this patient, fiberoptic laryngoscopy was performed immediately before surgery. The imaging studies showed a bifid uvula, chronic pharyngitis and arytenoideae edema, which may be secondary to gastroesophageal reflux.

Therefore, orotracheal intubation was excluded from this study, and all other surgical procedures used to establish medication delivery (removal of the bandage, surgical toilette, hemostasis, and wound dressing) were executed through inhaled anesthesia by the use of a facial mask and sevoflurane.

After venous cannulation and infusion of Ringer's lactate solution (5.5 mL/kg/h), the patient was premedicated with 50 mg ranitidine, 10 mg metoclopramide and 2 mg midazolam.

All hemodynamic and respiratory parameters (non-invasive blood pressure, heart rate, CO_2 levels, and pulse-oximetry levels) were continuously monitored and remained within the normal ranges. Adhesive EKG pellicle pads were cut, and the special silicone dressing described below was applied over them. Cotton was interposed between the blood pressure cuff and the skin of the patient. A pulse oximeter is a forcep probe that does not injure the skin of patients affected by EB.

Anesthesia induction was achieved using a bolus administration method of 2 mg/kg ketamine followed by a continuous infusion rate of 0.5-1 mg/kg/h. Alternatively, we also used intravenous remifentanil at 0.2-0.3 μg/kg/min.

Anesthesia was maintained by continuous infusion of ketamine and remifentanil plus administration of 1% sevoflurane in a mixture of O_2/air (FiO_2=0.3) by facial mask. As for previous cases that required a face mask to administer anesthesia, a special adhesive dressing (Mepilex® Heel with Safetac® Technology, Mölnlycke Health Care AB, Göteborg, Sweden) [2, 4, 5] was interposed between the facial mask and the patient's face to avoid exposing cutaneous traumatic wounds.

The dressing consisted of:

— a soft silicone wound contact layer (Safetac®);

— a flexible absorbent pad of polyurethane foam;

— an outer film that was permeable to vapor but was waterproof.

The medication is usually designed for exuding wounds, but its structural characteristics suggest new applications. The medication can be applied and shaped on the face of the patient according to the shape of the patient's mouth and to the dimensions of the facial mask. The adhesive side is turned toward the facial mask while the foamy side is applied on the face of the patient.[2]

To assure an adequate level of anesthesia and pain control, boluses of ketamine were administered during more painful surgical procedures. One mg/kg of ketamine and 0.5 mg/kg were given after the first and second hour of the operation, respectively. In addition, 2 mg of e.v. midazolam was administered.

The patient was breathing normally (with a respiratory rate of 10-12 breaths per min) throughout the operation; thus, no further assistance or controlled ventilation was necessary.

The brachial plexus cords were isolated by the surgeon during the amputation procedure (medial, lateral and posterior regions of the brachial plexus cords). Local anesthetic was injected in each cord (1% mepivacaine, 0.5 mL).[6]

Subsequently, an epidural multi-orifice catheter was placed with the tip near the cut plexus brachialis cords to provide a bolus of 0.5% levobupivacaine (10 mL), followed by a continuous infusion of 0.5% levobupivacaine (2 mL/h) for postoperative pain control. The epidural catheter was placed and stitched in by the surgeon.

Intravenous infusions of ketamine and remifentanil were stopped 15 min before the end of operation. The patient was immediately awakened at the end of the procedure and responded to simple orders with full cooperation without any sign of excitation.

Postoperative pain was evaluated using the Visual Analogue Scale (VAS), a 10-cm line corresponding to the amount of pain experienced. The mean VAS at 6, 12, 24 and 72 hours was <4 cm, with an optimal level of acute postoperative pain.

On the third postoperative day, an accidental kinking of the catheter during medication delivery prevented continuous infusion of local anesthetic. As a result, the VAS value was higher (VAS=8 cm). A bolus of 0.75% levobupivacaine (10 mL) was necessary to re-establish analgesia.

The epidural catheter was maintained for five days without further complication. No supplemental analgesia was necessary.

Discussion

EB is group of dermatologic and hereditary disorders characterized by the presence of extremely fragile skin and recurrent blisters subsequent to minor mechanical friction or traumas. Normal evolution of these blisters consists of scar formations that, in the oral cavity, are responsible for the structural and pathological alterations that sometimes prevent orotracheal intubation.[2, 3, 7]

The pathology of EB manifests in different clinical presentations:[1, 2]

— simplex EB (blisters within the epidermis);

— junctional EB (blisters at the level of lamina lucida but within the basement membrane zone);

— dystrophic EB (blister lesions are located at the level of lamina densa, with an important scarring component).

The most serious forms can be deadly for patients of intrauterine or neonatal age. The first signs are manifested during neonatal ages or in the first few months of life.

EB involves the skin of the whole body and the great part of the mucosal surfaces, including the

oral cavity, esophagus, stomach, intestines, lungs, bladder, genital region and eyes.

Patients generally die in the second to third decade of life, and the causes of death are usually related to the general compromised state of the patient as a result of multiple organ failures.[1, 2]

In this case report, we refer to a patient affected by dystrophic EB.

Due to a previous axillary lymphadenectomy performed on the patient, the difficult intubation process and the inadequate approach of any regional anesthetic technique to cover the axillary area, we decided to use the anesthetic management described in this paper.

The reduced buccal cavity opening would have prevented the use of the LMA. Furthermore, both the laryngoscope blade and the fiberbronchoscope would have caused the formation of intra-oral bulla during any attempt at intubation, [3, 4] with the added risk of causing arytenoideae edema. A nerve block of the brachial interscalene plexus could have been used concurrently with limb amputation, [8] but it would not have assured sufficient infiltration of the local anesthetic and subsequent analgesia to the armpit due to the presence of a large lymphonodal package.

For these reasons, the management of choice was to opt for an anesthetic management strategy that assured both good analgesia and spontaneous breathing. This was done by continuous administration of ketamine and remifentanil. At the same time, sevoflurane was given to the patient to obtain a deeper level of anesthesia.

Because a block in the brachial interscalene was previously performed, even with the presence of a large lymphonodal package, we proceeded with the amputation of the upper extremity from the section of the plexus brachial nerves after injecting the region with mepivacaine. Intraneural injection was not followed by nerve injury, thus confirming what was already demonstrated previously.[9, 10] Postoperative analgesia was confirmed by placing an epidural catheter through the axilla with the tip turned towards the cut nerves during surgery. Through the epidural catheter, we administrated a bolus of levobupivacaine as well as provided a continuous levobupivacaine infusion. Levobupivacaine was used because it is less toxic to the central nervous system than bupivacaine; its reduced toxicity suggests appropriate usage in clinical situations in which the risk of systemic toxicity due to either drug overdose or unintended intravascular injection is high, , such as peripheral nerve block).[11, 12]

Ketamine was used only for intraoperative anesthetic management.

Postoperative analgesia was provided by a continuous infusion of local anesthetic into the wound. However, the ketamine that was administered intraoperatively may have also contributed to the postoperative analgesia.[13, 14]

We did not assess phantom limb pain in this case report. Phantom limb pain is the pain felt in the area where a limb had been amputated. Almost everyone with an amputated limb experiences the sensation of phantom limb. These patients have the vivid impression that the limb is present. Moreover, 60% to 70% of these amputees suffer from phantom limb pain.[15]

Studies on lower extremity amputation have found that continuous intra- and postoperative epidural analgesia decreases stump pain but not phantom limb pain. Therefore, although an excellent postoperative analgesia was achieved in our case, it was not possible to know whether this also decreased the risk of phantom limb pain.[16]

In summary, continuous infusion of local anesthetics provided excellent postoperative analgesia without major side effects or complications.

Conclusions

In conclusion, it is possible to use ketamine and remifentanil *via* continuous IV infusion along with continuous administration of local anesthetics through the axilla *via* an epidural catheter. We found that this technique achieved adequate pain control for EB patients who are undergoing major orthopedic surgery, especially those who are characterized by difficulties in orotracheal intubation.

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