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Caudal Epidural Anesthesia in an Infant with Epidermolysis Bullosa

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Epidermolysis bullosa (EB) refers to several hereditary diseases characterized by separation of the epidermis and/or dermis following shear forces. Patients with EB present with bullae, blister, and erosion formation following seemingly minor trauma; these may heal completely or may lead to scarring, deformity, malnutrition, chronic infection, and death.¹ Because of the skin contact involved, many aspects of general anesthesia, especially manipulation of the airway, expose patients with EB to severe risk.²⁻⁵

Reviews of the anesthetic management of patients with EB have cautioned against the use of regional anesthesia; concerns were raised that scarring and contractures would obscure landmarks; chronic infections would make block placement unsafe; and injection of local anesthetics might lead to bullae formation.^{2,3,5} Despite these concerns, there have been recent reports of the use of regional anesthesia in patients with EB; these include two reports of the use of brachial plexus block in children undergoing reconstructive hand surgery^{6,7} and two reports of subarachnoid and epidural block in adults having gynecologic, obstetric, and abdominal procedures.^{8,9} There have been no reports of the use of regional techniques in infants with EB; we therefore present this report of an infant with the simplex form of EB who received a caudal epidural anesthetic for circumcision.

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CASE REPORT

A 6-wk-old, 5-kg male infant presented for elective circumcision. He was the 3.8-kg product of an uncomplicated gestation and delivery;

following birth bullae and erosions were noted on his face, scrotum, and extremities. Biopsy of the lesions at an outside hospital was consistent with the simplex form of EB. The patient had required daily dressing changes at home until the lesions healed 1 week prior to admission for circumcision. The infant was receiving no medication and there was no family history of EB.

Physical examination was unremarkable except for well-healed erosions on the extremities and face; the presacral region was free of bullae and erosions. Hemoglobin and hematocrit were within normal limits for age. The anesthetic plan was discussed with both parents and surgeon and all agreed to proceed.

The infant was taken to the operating room where a Nellcor® infant pulse oximeter probe was placed on a toe; the adhesive on the probe was covered with cellophane except over the LED and photodiode. The heart rate was 133 beats/min and the oxygen saturation was 98%. The infant was then held on the shoulder of an assistant in a comfortable burping position and landmarks gently identified. The presacral area was blotted with Betadine solution; our usual skin wheal over the sacral hiatus was omitted because of concern about bullae formation. A single-shot caudal epidural block was performed using a 1-inch 23-G needle and 4 ml of 0.375% bupivacaine with 1:200,000 epinephrine (0.8 ml/kg or 3 mg/kg). The procedure was completed in less than 2 min. Onset of block, determined by loss of withdrawal to gentle toe pressure, occurred in 6 min. Although we elected not to test the level of the block to avoid trauma to the skin, progression of block was followed by observation of muscle tone and activity in lower extremities, abdomen, and chest. The infant was then placed on an Exu-Dry® dressing, and an iv catheter was placed in the right foot and secured with a gauze bandage over Webril®; the infant did not respond to iv placement. Five minutes after block placement arterial blood pressure was 75/60 mmHg using a Dinamapp® cuff over a layer of Webril®. The blood pressure cuff was then removed and further monitoring consisted of the pulse oximeter and continuous observation and interaction with the infant. The infant's arms were gently restrained with gauze bandages over Webril®, and he was given a pacifier containing gauze soaked with 50% dextrose in water.

The infant rested quietly throughout the 20-min procedure and showed no response to surgical stimulation. Oxygen saturations and heart rate remained stable at 98–100% and 130–155 beats/min, respectively. The infant was cared for briefly in the recovery room and then returned to his room; other than cautioning caregivers that the lower extremities would remain numb for several hours, no special postoperative orders were deemed necessary. The duration of anesthesia, again determined by withdrawal to gentle toe pressure, was 156 min.

The postoperative course was unremarkable and the infant was discharged on the afternoon following surgery. Phone consultation with the infant's mother 10 days after the surgery revealed no new formation of bullae on the toes, wrists, or presacral area.

DISCUSSION

The simplex form of EB is the most benign of the mechanobullous diseases. Bullae usually heal without scarring and long-term prognosis is good. Inheritance is by classic Mendelian genetics with an autosomal dominant pattern. Estimated incidence of inherited EB simplex is 1 in 50,000 live births, whereas new sporadic cases occur with a frequency of 1 in 500,000 live births.¹

Because repeated manipulation of the foreskin during daily hygiene may lead to bullae formation, balanitis, and phimosis, male infants with EB may require circumcision.

Leaving aside ethical considerations, performance of circumcision using only forcible restraint could lead to massive bullae formation in an infant with EB. Alternatives to spinal or epidural anesthesia in this infant would include general anesthesia, which would require manipulation of the airway, and dorsal nerve block of the penis,¹⁰ which is contraindicated because the technique involves sc infiltration of local anesthetic. We currently have greater experience with caudal epidural anesthesia than with subarachnoid block in infants and find the caudal technique easier and quicker to perform; otherwise, we see no advantage of caudal over subarachnoid block. The volume, dose, and concentration of bupivacaine given were chosen to provide dense anesthesia and motor blockade to a level sufficient to prevent movement of the hips and legs during surgery. Takasaki *et al.*¹¹ suggested that the dose given should have produced a T8 level. As noted above, we chose not to formally test the level; however, rate and pattern of progression of loss of muscle tone was consistent with epidural block, and no impairment of respiratory effort or movement of hips or legs was noted during surgery.

Caudal epidural block is recognized to be an extremely safe and simple procedure to perform in infants and children.¹² Hypotension following spinal or epidural anesthesia in infants is uncommon¹³; we have not found it necessary to administer a fluid bolus before block placement or to maintain these infants in a supine position postoperatively. The absence of hypotension after epidural block in infants allows iv access to be delayed until vasodilation due to sympathetic block and loss of sensation provide optimal conditions; in addition, attempts to obtain iv access prior to block placement would have exposed this infant to unnecessary skin trauma. Although not necessary in this case, we have found that small doses of ketamine (0.25–0.5 mg/kg) administered iv provide adequate sedation in the older or more vigorous infant having surgery under caudal block.

The management of the patient with EB receiving general anesthesia has been extensively reviewed.^{2–9,14} Clearly, precautions such as padding of pressure points and avoidance of skin traction and adhesives apply equally well to the patient with EB receiving regional anesthesia. In addition, the site of the planned block must be free of lesions, care must be exercised during palpation of landmarks, and the skin must not be wiped during disinfection.^{7–9} The use of caudal or other regional techniques in infants and children requires familiarity with both the technique to be employed and the anatomic, pharmacologic, and psychologic differences between infants, children, and adults.^{12,15}

The principle advantage of the use of regional anesthesia in patients with EB is the avoidance of manipulation of the airway with the attendant risk of bullae formation

on the face and in the mouth, oropharynx, and trachea. In addition, the awake patient may assist in positioning and protect the unanesthetized limbs. Finally, contact with the awake patient may limit the number and intrusiveness of monitors required.

In summary, we have presented a case of an infant with EB simplex undergoing circumcision successfully managed with caudal epidural blockade. When surgical site, procedure, and patient condition permit, a regional technique can provide safe and effective anesthesia in patients with EB.

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Recurrent Respiratory Depression after Alfentanil Administration

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Recurrent respiratory depression after apparent recovery from the administration of fentanyl was first reported in 1976,¹ over 10 yr after its introduction. Subsequent reports have implicated secondary rises in plasma fentanyl concentration as the cause of many (but not all²) of these events.³⁻⁶ Alfentanil is a new opioid analgesic, with a shorter terminal elimination half-time ($t_{1/2\beta}$) than fentanyl. This is due to its smaller steady-state volume of distribution, although its hepatic clearance is also less.⁷ These pharmacokinetic differences may mean that recurrent respiratory depression is less likely after alfentanil,⁸ and that secondary peaks in alfentanil plasma concentration should not occur by the same mechanism

thought to cause secondary peaks in plasma fentanyl concentration: reuptake from tissue stores.⁷ However, we now report an episode of recurrent respiratory depression after alfentanil administration and discuss the possible causes.

CASE REPORT

A 49-yr-old, 60-kg man with squamous cell carcinoma of the posterior wall of the hypopharynx was scheduled for panendoscopy and biopsies for tumor mapping, and fine-needle aspiration of an enlarged left cervical lymph node. The patient complained only of mild dysphagia without hoarseness. Past medical history included 30 yr of heavy smoking and alcohol consumption. The patient was taking no medications. Examination showed a thin man, edentulous, without gross deformities of the upper airway. The trachea was in the midline and the cardiopulmonary examination was normal. Complete blood count, serum electrolytes, chest radiograph, and ECG were normal.

Without premedication, anesthesia was induced with alfentanil 75 $\mu\text{g} \cdot \text{kg}^{-1}$ iv and isoflurane 0.5% along with vecuronium 0.1 $\text{mg} \cdot \text{kg}^{-1}$ iv for muscle relaxation. Direct laryngoscopy was performed and the trachea intubated without difficulty. Alfentanil 1.0 $\mu\text{g} \cdot \text{kg}^{-1} \cdot \text{min}^{-1}$ iv, nitrous oxide 60%, and isoflurane 0.5% maintained anesthesia. During bronchoscopy, the FIO_2 was increased to 1.0, and ventilation controlled via the side arm of the bronchoscope. Forty-eight minutes after induction of anesthesia, thiopental 75 mg and additional vecuronium was given iv in response to movement, and the alfentanil infusion was

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