

Management of cesarean delivery in a parturient with osteogenesis imperfecta : A case report

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Abstract

Osteogenesis imperfecta (OI) is characterized by bone fragility, defects in the teeth, blue sclera, and deficits in hearing and vision [1,2]. Because of an anticipated difficult airway and back anatomy, there is a high risk of choosing either general or spinal anesthesia, especially in critically ill obstetric patients. It is still controversy about the anesthesia method in patients with OI. Ultrasound-guided transversus abdominis plane block (TAPB) has been used for analgesia after cesarean section, but rarely for anesthesia in this operation [3-5]. We describe a critically parturient with OI who underwent cesarean section under ultrasound guided TAPB with spontaneous breathing general anesthesia. The patient's vital signs have remained stable during the operation, and a live female infant was delivered successfully by cesarean section. The mother and daughter were safe at last. Written informed consent was provided by the patient for publication of this report with photos.created.

Abbreviations: OI: Osteogenesis imperfecta; SBGA: Spontaneous breathing general anesthesia; TAPB: Transversus abdominis plane block

Case Report

A 18-year-old parturient G1P0 with type-III OI [6] was admitted to our obstetric unit at 31 weeks of pregnancy. She was an abandoned baby with severe pelvic and spinal deformities, who was taken to hospital after been found by her adoptive mother. She was diagnosed with OI and multiple fractures throughout the body at that time. And then she was given conservative treatment. Because of dizziness and worsening dyspnea, she was admitted to the hospital at 31 weeks of gestation. She was diagnosed with 31⁺⁴ weeks of gestation, G1P0 and osteogenesis imperfecta. She has never received nerve block anesthesia or general anesthesia ever before.

She was 90 cm in height, and 29 kg in weight (body mass index (BMI): 35.80 kg/m²). Physical examination: T 36.4°C, P 135/min, RR 30/min, BP 104/60mmHg, pelvic and spinal deformities, thick breath sounds in both lungs, wet rales can be heard in the lower right lung, lower limb muscle strength level 0, upper limb muscle strength level 3-4. Auxiliary examination: hemoglobin 91g/L, hematocrit 0.293, platelet count 216*10⁹/L; prothrombin time 13.7s; total protein 55.3 g/L, albumin 30 g/L. Arterial blood gas analysis without oxygen inhalation: PH 7.436, PaCO₂ 29.4mmHg, PO₂ 275mmHg, hematocrit 30.8%. Echocardiography showed a small amount of pericardial effusion

and mild regurgitation of the second and tricuspid valves. Chest radiography showed fusion from the cervical spine to the lumbar spine, ground-glass opacity in the right upper lung and poor visualization of the heart shadow. A chest CT scan (low-dose) showed: scoliosis, thoracic deformity, effusion, and multiple old rib fractures on the left side.

We believed that the woman had poor cardiopulmonary function and lung infection. The American Society of Anesthesiology (ASA) class of the patient was IV. Although Mallampati is grade I, there is a high risk of anesthesia in the perioperative period of cesarean section. As the woman's dyspnea worsened, she finally had a cesarean section at 32 weeks of gestation. The patient fasted for more than 8 hours before surgery. After signing the informed consent for anesthesia, the patient was admitted to the operating room in a supine position with the uterus shifted to the left. And then she was given routine monitoring, venous access and oxygen inhalation with mask at 5 L/min.

Dexmedetomidine was premedicated 2 µg.kg⁻¹.h⁻¹ for 10 minutes, then was adjusted to 0.4µg.kg⁻¹.h⁻¹ and stopped until 40 minutes before the end of the operation.

Ultrasound-guided TAPB was performed using the method described by Hebbard et al. [7] Using ultrasound (GE venue 40, 12MHz linear probe) to locate the transversus abdominis plane (TAP) at one side on the axillary midline between costal margin

and iliac spine, and then guide the puncture to the TAP, inject 15ml 0.5% Ropivacaine into the TAP through the needle. The same method was used on the other side.

Larynx and airway surface anesthesia with 2% lidocaine was applied. The orotracheal tube with an internal diameter of 5.5 mm was intubated with the help of Glidescope (Zhejiang UE Medical Corp, TD-C-IV) and then fixed at 18 cm. Anesthesia was then induced with sufentanil (8 ug), and etomidate (6 mg), and maintained with remifentanyl ($0.05\text{ug}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$) and isoflurane with MAC values between 0.6 and 0.7.

Using simultaneous intermittent mandatory ventilation (SIMV) ventilation mode, the parameter is adjusted according to the patient's tidal volume, respiratory rate and end-tidal carbon dioxide (ETCO₂). A healthy female infant (weight of 1630g and 40cm in length) was delivered, who had a normal appearance, and the Apgar scores were 8, 9, and 9 at 1, 5, and 10min, respectively. Muscle relaxants were not used, and the patient's spontaneous breathing was preserved. At the end of surgery, the patient was quickly awake, the tracheal tube was removed, and she was able to complete the command action. The patient was successfully sent to the intensive care unit for continuous observation. No other analgesic drugs were used postoperatively. Estimated blood loss and urine was respectively 200mL and 100mL, and crystalloid (500mL) were given slowly. Intraoperatively, norepinephrine was intermittently injected to maintain arterial blood pressure between 90-110/45-60 mmHg, HR100 -125 beats/min, oxygen saturation 97-99%, and RR12 -18 /min and end-tidal carbon dioxide (ETCO₂) between 35 and 40mmHg.

No malignant hyperthermia (MH) occurred intraoperative. After the operation, the symptoms of dyspnea disappeared..

Discussion

This Women with severe OI do not usually tolerate pregnancy to term and require early cesarean delivery as the increasing size of the gravid uterus. Pregnancy negatively effects on the already severely altered respiratory function [8]. Preoperative assessment is essential for the preoperative preparation of patients with OI. The initial workup of these patients should always include a thorough cardiac and pulmonary exam. An echocardiogram should be ordered to rule out valvular disease and show the pulmonary artery pressure of the patient with heart failure. An airway exam should detail mandibular and cervical spine function, mouth opening, and arrangement of teeth, and the presence of a difficult airway, to assess the severity of spinal deformity and whether spinal anesthesia can be practiced.

Due to the anticipated difficulty of the neuraxial block, we did not try neuraxial block and chose general anesthesia directly. Given the nature of OI, extreme caution must be taken to prevent fractures. Arterial cannulation for blood pressure measurement has been suggested to avoid repeating trauma from a blood pressure cuff. Positioning can be difficult because of the fracture

risk, aortocaval compression from the gravid uterus, respiratory difficulty caused by pregnancy-induced restrictive lung disease [9], exposure for surgery or block placement [10], and short stature. And it is also concerned that the application of excessive pressure during laryngoscopy may cause fractures or dental damage [11], We attempted a very gentle directly Glidescope to avoid any mandibular trauma and the actual intubation process was uneventful and atraumatic.

Mei et al. reported the combined use of TAP and ilioinguinal-iliohypogastric nerve blocks can provide effective anesthesia for cesarean section [4]. We believed that our patient might benefit from the ultrasound-guided TAPB. We did not use muscle relaxant, because the mechanical control breathing could interfere with the ventilation/perfusion ratio of the patient, and extubation could be performed as early as possible after surgery to reduce the adverse effect of mechanical ventilation on pulmonary function. Coffman [5] reported a case in which the patient strongly desired avoiding general anesthesia as her Harrington rod surgery was complicated by a six-week period of postoperative intubation and mechanical ventilation due to muscle weakness and restrictive lung disease.

The hemodynamic parameters of our patient did not fluctuate significantly during skin incision, indicating that ultrasound-guided TAPB can effectively inhibit the skin incision stimulation of the lower abdominal wall. No complication attributed to the ultrasound-guided TAPB was observed in our patient [4]. Postoperatively, we did not use other analgesic methods and medications, and the patient did not complain of pain. It is also an option for postoperative analgesia, especially after general anesthesia [12]. OI was once thought to increase the risk of MH during general anesthesia [13-15]. No increase in ETCO₂ was found in our patient.

In summary, the use of ultrasound-guided TAPB can effectively inhibit the incisional skin stimulation of the lower abdominal wall and provide good postoperative analgesia. In patients with impaired pulmonary function, spontaneous breathing is preserved during surgery, and extubation can be performed as early as possible after surgery to reduce the adverse effect of mechanical ventilation on pulmonary function. The authors of this case report believe that this method is effective in cesarean section in patients with severe scoliosis or poor cardiopulmonary function.

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