Anesthetic management of a parturient with VACTERL association undergoing Cesarean delivery

Prise en charge anesthésique d'une parturiente atteinte du syndrome de VACTERL subissant un accouchement par césarienne

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Abstract

Purpose We present the anesthetic management of a parturient with VACTERL association undergoing combined regional and general anesthesia for Cesarean delivery. Defined as a syndrome, VACTERL association comprises at least three of the following abnormalities: vertebral, anal atresia, cardiac, tracheoesophageal, renal, and limb.

Clinical features The patient's anatomic abnormalities and comorbidities comprised severe cervicothoracic scoliosis, kyphoscoliosis, congenitally fused ribs, and severe restrictive lung disease. She had a Mallampati class 3 airway, a right laterally flexed neck, and reduced mandibular protrusion. We performed a lumbar spine ultrasound for epidural placement which was used to provide peri- and postoperative analgesia. Due to the anticipated difficult tracheal intubation, the patient underwent an awake fiberoptic intubation and subsequently received general anesthesia. The patient's trachea was extubated on the first postoperative day, and she received adequate post-Cesarean epidural analgesia.

Conclusion This case highlights the challenges that anesthesiologists face when managing parturients at extremely high risk for perioperative anesthetic morbidity

due to the presence of severe pre-existing disease, anticipated difficult airway, and major spinal abnormalities complicating neuraxial anesthesia.

We used a combined general and epidural anesthetic approach to control ventilation, provide effective postoperative analgesia, and reduce the risk of anesthetic-related perioperative morbidity. An individualized approach should be considered for the anesthetic management of high-risk pregnant patients with complex and multiple medical and surgical morbidities undergoing labour and delivery.

Résumé

Objectif Nous présentons la prise en charge anesthésique d'une parturiente souffrant du syndrome de VACTERL et subissant un accouchement par césarienne sous anesthésie régionale et générale combinée. Le syndrome de VACTERL, ou association VACTERL, comprend au moins trois des anomalies suivantes : anomalie vertébrale, imperforation de l'anus, anomalie cardiaque, trachéo-œsophagienne, rénale, ou des membres.

Éléments cliniques Les anomalies anatomiques et les comorbidités de la patiente comprenaient une grave scoliose cervico-thoracique, une cypho-scoliose, des côtes fusionnées de façon congénitale, et une maladie pulmonaire restrictive grave. L'évaluation de ses voies aériennes a révélé un score de Mallampati de 3, son cou présentait une flexion latérale droite et elle présentait une protrusion mandibulaire réduite. Nous avons réalisé une échographie de la colonne lombaire pour positionner la péridurale qui a été utilisée pour l'analgésie péri- et postopératoire. En prévision d'une intubation trachéale

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Fig. 1 Photograph showing the patient with VACTERL association (vertebral defects, anal atresia, cardiac defects, tracheoesophageal abnormalities, renal abnormalities, and limb abnormalities) highlighting the gross anatomical abnormalities with emphasis on the head and neck to torso position

difficile, la patiente a subi une intubation fibroscopique éveillée et l'anesthésie générale a été réalisée subséquemment. La trachée de la patiente a été extubée le premier jour postopératoire, et elle a reçu une analgésie péridurale post-césarienne adaptée.

Conclusion *Ce cas souligne les défis auxquels font face les anesthésiologistes lorsqu'ils prennent en charge des parturientes courant un risque extrêmement élevé de morbidité anesthésique périopératoire en raison de la présence de maladies graves préexistantes, de voies aériennes anticipées comme difficiles, et d'anomalies majeures de la colonne, compliquant l'anesthésie neuraxiale.*

Nous avons utilisé une approche anesthésique générale et péridurale combinée afin de contrôler la ventilation, de fournir une analgésie postopératoire efficace, et de réduire le risque de morbidité périopératoire liée à l'anesthésie. Une approche personnalisée doit être envisagée pour la prise en charge anesthésique des patientes enceintes à risque élevé présentant des morbidités médico-chirurgicales à la fois complexes et nombreuses et devant accoucher.

The acronym, VACTERL, is used to denote the following abnormalities associated with the disorder: vertebral defects, anal atresia, cardiac defects, tracheoesophageal abnormalities, renal abnormalities, and limb abnormalities. This multisystem disorder is rare, as the estimated incidence of VACTERL association varies from 1/10,000 - 1/40,000

live-born infants.¹ The diagnosis of VACTERL association requires the presence of at least three of these abnormalities without other major congenital abnormalities.² The exact cause of this disorder is unknown, as no clear genetic or environmental etiologic factors have been previously identified; thus, a high degree of clinical and causal heterogeneity is suspected.¹

Patients with VACTERL association pose important challenges for anesthesiologists. Neuraxial block placement and/or airway management for endotracheal intubation may prove to be difficult in patients with major skeletal and/or spinal deformities. In addition, these patients may have severe restrictive lung disease secondary to scoliosis, which may negatively impact the adequacy of oxygenation and ventilation during neuraxial or general anesthesia. The physiological effects of pregnancy may increase the severity and complexity of these anatomic and physiologic comorbidities.

We describe the successful anesthetic management of a parturient with VACTERL association who underwent a combined neuraxial and general anesthesia technique for a scheduled Cesarean delivery. The patient provided written informed consent for publication of this case report.

Case report

A 20-yr-old parturient G1P0 (height = 1.3 m; weight = 36 kg) with a singleton pregnancy at 39 weeks gestation was scheduled for primary Cesarean delivery. The patient had severe skeletal abnormalities related to VACTERL association, namely, cervical-thoracolumbar scoliosis, kyphoscoliosis, congenitally fused ribs with hemi vertebrae, an absent right upper limb, and an absent right scapula. Gross anatomic abnormalities affecting the head and neck are displayed in Fig. 1. The patient's preoperative chest x-ray showed the severity of the scoliotic deformities affecting the cervical-thoracic vertebral column, with negligible aeration in the right hemithorax. Due to the severity of the scoliosis, the patient had evidence of severe restrictive lung disease without hypoxemia and no active obstruction. Her pulmonary function tests at 28 weeks gestation showed forced vital capacity (FVC) = 0.58 L (26% of predicted), forced expiratory volume in one second (FEV₁) = 0.47 L (22% of predicted), FEV₁/FVC = 80% (91% of predicted), forced expiratory flow (FEF) 25-75% = 0.4 L·sec⁻¹ (13% of predicted), and maximum FEF = 0.87 L·sec⁻¹ (17% of predicted). An echocardiogram performed at 36 weeks gestation revealed mild pulmonary arterial hypertension, an elevated right ventricular systolic pressure (approximately 46 mmHg), a non-impairing ventricular septal defect, mild tricuspid regurgitation, and normal left and right ventricular size and

function. She denied syncope, chest pain, tachypnea, or dyspnea prior to and during pregnancy. She also reported a history of mild asthma but rarely used an inhaler. The patient had undergone a prior cleft palate repair and a Nissen fundoplication; she did not recall any anesthetic complications. The relevant findings on her preoperative airway examination were a Mallampati class 3 airway, fixed neck position in a right lateral flexed position (coronal axis), and mildly reduced mandibular protrusion.

The patient was reviewed in her third trimester by respiratory and cardiology teams at Stanford University Medical Center. As her cardiorespiratory function had been unchanged during her pregnancy, no additional pre-delivery tests or investigations were deemed necessary. A primary Cesarean delivery was scheduled due to maternal request and medical concern that her respiratory reserve was too limited for a trial of labour.

With regard to the anesthetic plan, we had considerable concern about using regional anesthesia as our primary and only anesthetic technique for Cesarean delivery. We speculated that surgical anesthesia up to a T5 level would severely compromise ventilation and oxygenation in the supine position due to our patient's restrictive pulmonary function and small stature. As a result, we determined that general anesthesia was a preferential approach in order to ensure adequate oxygenation and optimal ventilatory control, including careful monitoring of airway pressures. After discussion with the patient, our anesthetic plan was to perform preoperative lumbar epidural catheter placement in order to provide perioperative and postoperative analgesia, followed by an awake fiberoptic intubation (AFOI) with subsequent general anesthesia.

In the preoperative period, her vital signs were as follows: blood pressure = 121/50 mmHg, heart rate = 103 beats·min⁻¹, oxygen saturation = 97% on room air, and respiratory rate = 22 breaths·min⁻¹. Prior to attempting regional and general anesthesia, intravenous access was secured with an 18G intravenous cannula in her left arm, and a 20G arterial line was placed in her left radial artery. Metoclopramide 10 mg *iv* and ranitidine 50 mg *iv* were administered. Due to the severe scoliosis and kyphoscoliosis, we performed an ultrasound examination of the lumbar spine using a SonoSite M-Turbo® ultrasound system with a curvilinear probe (SonoSite, Inc., Bothell, WA, USA) to identify the L2-L3 interspace. After marking the L2-3 interspace in the longitudinal and transverse planes and approximating the spinal torsion, we performed epidural catheter placement using a 17G Tuohy needle at the first attempt at the L2-3 interspace. The epidural space was identified using a loss-of-resistance technique with saline at a depth of 4 cm, and a 19G polyamide epidural catheter was inserted and taped to the patient's skin at 8 cm. An epidural test dose using 2% lidocaine 2 mL with 1:200,000 epinephrine was negative.

Prior to the AFOI, the patient was premedicated with midazolam 1 mg *iv* and glycopyrrolate 100 µg *iv*, and nebulized 4% lidocaine was administered prior to commencing the AFOI. A remifentanyl infusion was used (0.15 µg·kg⁻¹·min⁻¹) to provide sedation and ensure patient compliance during the AFOI, and the patient received oxygen (10 L·min⁻¹) using a modified facemask. While receiving the remifentanyl infusion, the patient remained conscious, breathed spontaneously, and obeyed commands. Before commencing the AFOI, the patient's abdomen was disinfected and draped, and a scrubbed obstetric team remained on standby. The AFOI was successfully performed using a pediatric (3.7 mm outer diameter) Karl Storz intubation fibroscope (KARL STORZ GmbH & Co. KG, Tuttlingen, Germany) via the oral route. The patient's vocal cords were visualized, and 4% lidocaine was administered via the suction port to supplement topical anesthesia of the vocal cords prior to tracheal intubation. A size 7.0 tracheal tube was railroaded over the fiberoptic scope into the trachea without difficulty and end-tidal carbon dioxide was detected. General anesthesia was initiated with propofol 50 mg *iv* and maintained using 0.7–0.8% sevoflurane in a 60:40 ratio of oxygen and air. The remifentanyl infusion was continued at 0.1 µg·kg⁻¹·min⁻¹ until completion of surgery. The patient also received rocuronium 15 mg *iv* in order to optimize ventilation. Prophylactic antibiotics (cefazolin 1 g *iv*) were administered prior to skin incision. She was hemodynamically stable throughout the surgery and had adequate oxygenation and ventilatory parameters. Synchronized intermittent mandatory ventilation mode was used (tidal volume 280 mL, respiratory rate 16–18 breaths·min⁻¹). Intraoperatively, peak inspiratory pressure did not exceed 28 cm H₂O and her oxygen saturation was maintained above 95%.

Delivery of a healthy female infant (weighing 3,350 g) in the vertex position via a Pfannenstiel incision was uneventful. There were no anomalies detected at birth and Apgar scores were 8 and 9 at one and five minutes, respectively. After delivery, the patient received an oxytocin bolus (1 U) *iv* and an oxytocin infusion (7.5 U·hr⁻¹); the uterine tone assessed by the obstetrician was adequate. The total estimated blood loss was 640 mL (approximating 18% of her total circulating volume). Bupivacaine 7.5 mg was administered via the epidural catheter after delivery of the infant, and morphine 2 mg was administered via the epidural catheter at the end of surgery.

The patient's trachea remained intubated electively at the end of surgery and she was transferred to the intensive care unit (ICU). Her lungs were ventilated overnight (arterial blood gas analysis within normal limits) and her cardiorespiratory status remained stable. A postoperative chest *x-ray* taken during the period of ventilation in the ICU is shown in Fig. 2; evidence of the severity of the

Fig. 2 Photograph (left) showing the back of the patient. Postoperative chest *x-ray* (right) highlighting severe cervical-thoracolumbar kyphoscoliosis, abnormal head and neck position, and evidence of restrictive lung volume (the *x-ray* is reversed to compliment the photograph)

cervical kyphoscoliosis can be seen in the film. The patient was sedated with a propofol infusion, and post-Cesarean analgesia was maintained with an epidural hydromorphone infusion ($0.1 \text{ mg}\cdot\text{hr}^{-1}$) that did not require supplementation. On the first postoperative day, her hemoglobin was $11 \text{ g}\cdot\text{dL}^{-1}$ and her coagulation and biochemical indices were within normal limits. After ensuring that experienced anesthesia personnel and airway equipment were available, she underwent an uncomplicated trial of tracheal extubation on postoperative day one. The airway was topicalized with 4% lidocaine prior to extubation, and her trachea was extubated over a Cook Airway Exchange Catheter® (Cook Critical Care, Bloomington, IN, USA). This allowed us to assess the adequacy of spontaneous ventilation for 40 min before removal of the catheter. She was transferred to the postpartum unit later on postoperative day one. The remainder of her postpartum admission was unremarkable, and she was discharged home on postoperative day five.

Discussion

This case report highlights the major challenges that face obstetric anesthesiologists when managing pregnant patients at extremely high risk for perioperative anesthetic morbidity due to the presence of severe pre-existing disease, an anticipated difficult airway, and major spinal abnormalities complicating neuraxial anesthesia. Our patient with VACTERL association had severe restrictive lung disease and major anatomic abnormalities, notably, severe cervical-thoracolumbar scoliosis and an anticipated difficult airway. After gaining specialist multidisciplinary input, we chose to use general anesthesia for Cesarean delivery in combination with an epidural-based analgesic technique for providing peri- and postoperative analgesia.

Although our patient underwent planned Cesarean delivery, an identical delivery plan (Cesarean delivery with combined general and regional anesthesia) would have been adopted if the patient had presented in active labour before the scheduled date of surgery.

Due to the rarity of VACTERL association, the significant male preponderance (69.8%), and the high incidence of infertility problems,² we were only able to identify one previous article in the literature describing the anesthetic management of a patient with VACTERL association for scheduled Cesarean delivery.³ In this report, the patient had similar anatomic and medical morbidities to our patient (major scoliosis affecting the lumbar spine, poor respiratory function, and an unfavourable airway examination). In contrast to our case, an epidural technique was the only mode of anesthesia used to provide anesthesia for Cesarean delivery for this patient. Nonetheless, there was no specific information in the case report describing the adequacy of ventilation, oxygenation, or the peri- and postoperative quality of anesthesia and analgesia.

We were concerned about using a neuraxial block as the primary anesthetic technique for Cesarean delivery for a number of reasons. Previous research suggests that 25% of patients who receive spinal anesthesia for Cesarean delivery may develop sensory blocks to the cervical dermatomal region.⁴ As our patient had a very short stature (height = 1.3 m; weight = 36 kg) and severe restrictive lung disease, a level of spinal anesthetic up to or higher than the T5 dermatome may have severely compromised her functional residual capacity, especially with her lying in a supine position during surgery. To counter the risk of a high sensory block, a reduced dose of intrathecal bupivacaine may have been a viable option; however, findings from a recent meta-analysis indicate that a reduced dose of spinal bupivacaine is associated with an increased risk of

intraoperative breakthrough pain.⁵ As a result, we were extremely guarded about using a neuraxial block alone to provide surgical anesthesia because of the risk of intraoperative anesthetic morbidity, such as intraoperative breakthrough pain, respiratory insufficiency, or a high sensory block. These intraoperative complications may have warranted expeditious conversion to general anesthesia, which may have been particularly challenging as our patient was at high risk for a difficult tracheal intubation and major respiratory compromise. Nevertheless, patients with skeletal deformity and restricted lung volumes have the potential for high airway pressures during mechanical ventilation, and high intrathoracic pressure can negatively impact venous return to the heart and decrease the cardiac output. We were careful in choosing a low tidal volume to reduce peak airway (and intrathoracic) pressures in order to reduce the risk of barotrauma and secondary maternal hemodynamic compromise.

Tracheal reintubation after a failed extubation is not uncommon (0.4–25%) amongst ICU patients⁶; therefore, the timing of tracheal extubation for these high-risk patients needs careful consideration. Since the surgery was performed in our patient during the late afternoon, we determined that the safest approach was overnight lung ventilation with planned tracheal extubation on postoperative day one. We were reassured that she required minimal ventilatory support during the period of intubation in the ICU. We adopted a conservative approach to planned extubation using an airway exchange catheter with anesthesia personnel experienced in difficult airway management on standby. This is one approach that has been recommended for patients with difficult airways who are deemed to be at high risk for failed tracheal extubation.⁶

We acknowledge that a carefully titrated regional anesthetic using a catheter-based technique (epidural *de novo*, continuous spinal, low-dose combined spinal-epidural) may also have been a viable approach. The use of a titrated approach using a catheter-based technique has been described for high-risk obstetric cases with cardiac disease⁷ and for patients with other severe types of respiratory disease.⁸ With a carefully titrated dosing technique, the upper level of the dermatomal block and the secondary effect on the adequacy of spontaneous ventilation can be assessed closely. This approach may have potentially obviated the need for tracheal intubation and general anesthesia.

Previous studies have indicated that patients with a Mallampati score > 1 are at increased risk of difficult tracheal intubation⁹ and failed intubation.¹⁰ In addition to a Mallampati score of 3, our patient had other unique features associated with a difficult airway, notably, a fixed position of her neck in a hyperextended right lateral position. These airway features suggested that a standard tracheal intubation technique for our patient with a rapid sequence induction would prove challenging. A previous report indicated that the timing of airway complications in obstetric patients primarily occurs during tracheal intubation for emergency Cesarean delivery,¹¹ presumably with a rapid sequence induction. Therefore, we considered the safest option for our patient to be general anesthesia using an AFOI. AFOI is an important technique used for managing patients with anticipated difficult airways; however, there are surprisingly few reports in the literature describing the use of AFOI in patients with anticipated difficult airways requiring Cesarean delivery.^{12–15} Summary details for the indications for AFOI and anesthetic management from

Table 1 Summary of articles describing awake fiberoptic intubations in parturients

	Ghaly <i>et al.</i> ¹²	Neumann <i>et al.</i> ¹³	Arendt <i>et al.</i> ¹⁴	Trevisan ¹⁵
Age	34	35	42	29
Gravida parity	G2 P1	G4 P0	G2 P1	G1 P0
Gestational age	38 weeks	35 weeks	37 weeks	37 weeks
Indication for AFOI	Type 1 ACM + cervical kyphosis + extensive syringomyelia	Spinal muscular atrophy	Ankylosing spondylitis	Temporomandibular joint ankylosis (unknown etiology)
Anticipated difficult airway	Yes	Yes	Yes	Yes
Urgency of CD	Scheduled	Urgent	Scheduled	Scheduled
Sedation	Butorphanol 1.5 mg <i>iv</i>	Dexmedetomidine 1 µg·kg ⁻¹ (over 10 min) + 1 µg·kg ⁻¹ ·hr ⁻¹ (total dose 1.84 µg·kg ⁻¹ over 38 min)	Midazolam 2 mg <i>iv</i> + fentanyl 100 µg <i>iv</i>	None documented

AFOI = awake fiberoptic intubation; CD = Cesarean delivery; ACM = Arnold-Chiari malformation

these case reports are summarized in the Table 1. Successful AFOI is dependent on careful planning and preparation prior to the procedure.^{16,17} Key components for ensuring optimal conditions for AFOI include patient comfort and compliance, absence of secretions, excellent topical anesthesia of the airway, and maintenance of the patent airway with spontaneous ventilation. In our case, we decided to use a remifentanyl infusion as well to provide periprocedural sedation and to optimize patient compliance during the AFOI. Remifentanyl has a number of advantages as a sedative for AFOI due to its short onset of action, titratability, antitussive effects, and short elimination half-life.¹⁸ Several case reports have described using remifentanyl to provide adequate sedation for AFOI in patients with anticipated difficult airways.^{19,20} Remifentanyl also appears to compare favourably with dexmedetomidine as a sedative for AFOI,^{21,22} which may be related to attenuating the stimulation of the endoscope and endotracheal tube in the trachea.

For our patient, epidural analgesia was effective in managing post-Cesarean analgesia. In addition to providing analgesia, we determined that effective epidural analgesia would reduce the need for systemic opioid administration, which may have negatively impacted our patient's respiratory recovery and increased the risk of postoperative respiratory morbidity, such as atelectasis and pneumonitis. Although the use of epidural opioids are associated with respiratory depression, the incidence of respiratory depression is known to be very low (0–2.8%).²³

We were concerned that the severity of our patient's scoliosis would make epidural block placement technically challenging. Patients with vertebral column abnormalities are known to have a higher incidence of difficult neuraxial block placement and failure compared with patients without spinal abnormalities.²⁴ A previous literature review of neuraxial techniques in parturients with scoliosis found that a high percentage of patients with uncorrected and corrected scoliosis (79% and 69%, respectively) have successfully received neuraxial analgesia/anesthesia.²⁵ Patients with scoliosis have a slightly higher incidence of unintentional dural puncture that may be related to the higher risk for multiple passes of the epidural needle. Nevertheless, there is limited information on the mode of neuraxial block – spinal, epidural, combined spinal-epidural, or continuous spinal – that offers the best analgesic/anesthetic outcome. In one report, a continuous spinal technique provided good analgesia or anesthesia in 63% of patients with severe scoliosis, cardiorespiratory compromise, or previous spinal surgery.²⁶ Also, data on the anesthetic quality of epidural-based techniques in patients with scoliosis are less well known. The main complications consist of patchy or unilateral blocks as well as an

increased requirement for local anesthetic.²⁵ Fortunately, our patient received effective epidural analgesia, and she did not develop a postdural puncture headache or respiratory depression during her postoperative hospital stay.

Use of ultrasound in our patient made it possible to identify an appropriate lumbar spine interspace with subsequent epidural catheter placement on the first attempt. Recent advances in ultrasound technology have greatly assisted anesthesiologists to identify mid-lumbar interspaces with accuracy, especially in patients with previous spine pathology or surgical interventions.²⁴ Since the torsion of the spine in scoliosis makes it challenging to choose the correct angle of approach, the use of ultrasound to determine this angle was extremely helpful.

The successful obstetric and anesthetic outcomes for our patient were possible as a result of the specialist multidisciplinary review and clinical care that she received during the antenatal, peripartum, and postpartum periods. This approach has been endorsed in the most recent maternal mortality triennial report from the United Kingdom. The report advocates that women with potentially serious medical conditions should receive immediate and appropriate multidisciplinary specialist care.²⁷ As exemplified in this case report, patients with complex medical disorders who have morbidities that may complicate both general and neuraxial anesthesia modalities pose the greatest challenges to obstetric anesthesiologists.

In this case report, we report the successful anesthetic management of a patient with VACTERL association who had an anticipated difficult airway, severe restrictive lung disease, and major cervical-thoracolumbar scoliosis. We used a combined general and epidural anesthetic approach to control ventilation, provide effective postoperative analgesia, and reduce the risk of anesthetic-related perioperative morbidity. An individualized approach should be considered for the anesthetic management of high-risk pregnant patients with complex and multiple medical and surgical morbidities undergoing labour and delivery.

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Competing interests None declared.

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