Successful Management of Ex–Utero Intrapartum Treatment Procedure for Cystic Teratoma Under Sole Spinal Anesthesia: an Anesthetic Challenge Case Report

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Abstract:

The Ex–Utero Intrapartum Treatment (EXIT) procedure is a technique that establishes the fetus's airway, while uteroplacental fetal circulation is still maintained. Anesthesia for this specific procedure requires special anesthetic considerations; such as, uterine relaxation, maternal hemodynamic parameters, the establishment of a fetal airway and maintenance of postpartum uterine contraction. In most cases, general anesthesia is used as the preferred method, due to its effect on uterine relaxation. This presented case demonstrated that a sole spinal anesthetic technique can be used safely for an EXIT procedure.

Keywords: case report, congenital teratoma, EXIT procedure, neuraxial anesthesia

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Introduction

The Ex Utero Intrapartum Treatment (EXIT) procedure is a technique that establishes the fetus's airway, while uteroplacental fetal circulation is still maintained. Anesthesia for this specific procedure requires special anesthetic considerations; such as, uterine relaxation, maternal hemodynamic parameters, the establishment of a fetal airway, and maintenance of postpartum uterine contraction. General anesthesia is the preferred technique, due to providing proper uterine relaxation, maintaining uteroplacental circulation, hemodynamics, and fetal sedation.¹⁻⁴ Multidisciplinary team planning improves survival and reduces neonatal morbidity and mortality.⁵ There are few case reports showing the safety and success of a regional anesthetic technique for the EXIT procedure. In this present case report, the single-shot spinal anesthetic technique was used safely; in terms of maternal hemodynamics and uterine relaxation for the EXIT procedure.

Case Report

A 32 year old, gravida 2 para 1, 38 weeks of pregnancy, and ASA II; presenting with a fetus, having a cervical mass diagnosed by prenatal ultrasound at 32 weeks of pregnancy. At 36 weeks of gestation, magnetic resonance imaging (MRI) showed multiloculated cystic lesions measuring 5.9 x 5.3 x 6.3 cm in origin from the bilateral submental space, and extending to the pharyngeal, parapharyngeal, left paraglottic, anterior glottic, carotid space; resulting in focal, severe narrowed oropharynx. Karyotype testing showed 46,XX. The perinatologist team were concerned that the fetus would have probable, immediate airway difficulties after delivery.

Therefore, elective lower cesarean section was scheduled for the EXIT procedure to secure the fetus's airway. In most cases, general anesthesia is used as the preferred method, due to its effect on uterine relaxation.¹⁻⁴

This case is the first case from three cases of EXIT procedures that were undertaken at the institution. The patient was informed of the risks associated with this EXIT procedure. A counseling session was conducted to form the management plans, which included the patient's backup plan. The chosen anesthetic technique for cesarean section was single shot-spinal anesthesia, and nitroglycerin preparing for inadequate uterine relaxation.

On the day of surgery, a multidisciplinary team; involving pediatric anesthesiologists, obstetricians, otorhinolaryngologists, neonatologists, a pediatric surgeon, and nurses were available in the operating room. Regarding the plan after childbirth, immediate intubation had been planned, due to complicated airway situation concerns (i.e., a severely narrowed oropharynx, and both size and location of the mass that extended to the anterior glottis part).

In the operating room, baseline monitoring was applied with a pulse oximetry, ECG, and noninvasive blood pressure. As per standard anesthetic care, the vasopressor medications were prepared in case of hypotension occurring. Anesthetic medications and airway equipment for general anesthesia was also arranged for any emergency situation. An intravenous line, 18G, was established, and the patient was preloaded with Ringers Lactate at 1 liter. The patient was positioned in the left lateral position, and spinal anesthesia was performed with a single attempt. After administering 11 mg of 0.5% hyperbaric bupivacaine combined with intrathecal morphine 0.2 mg, the patient was repositioned back into a supine position; with the uterus displaced to the left to prevent aortocaval compression. The level of sensory block was T4. The uterine tone was adequately relaxing without need of nitroglycerin infusion. Upon delivery of the fetal head and shoulders, fetal tracheal intubation was successfully achieved; under video-assisted laryngoscopy (a blade, No. 1), via a 3.5 plain endotracheal tube anchored at 9 cm. The position of the tracheal tube was confirmed with auscultation and capnograph (Figure

1). After confirmation of the tracheal tube placement, we proceeded with clamping and cutting of the umbilical cord: time from uterine incision to cord clamp was 5 minutes. Syntocinon[®] (Oxytocin) infusion was used to promote uterine tone and rhythmic contraction of the uterus. The total volume of fluids given was 2,100 milliliters of Ringers Lactate. Estimated blood loss was 600 milliliters, and the patient did not require any blood transfusions. The total time elapsed between starting the anesthetic procedure and finishing the operation was 125 minutes. The cost of treatment along this admission was 17,013 Thai Baht, for which the government fully supported following the Universal Coverage; (UC) policy.

The neonate was transferred to the Neonatal Intensive Care Unit (NICU) for further mechanical ventilation and sedation. Subsequently, on Day 8 of Life, the neonate underwent complete resection of the cervical teratoma, and was successfully extubated at Day 12 of Life.

Discussion

Prenatal diagnosis of airway malformations is essential for the operating team to plan and define the best approach to be undertaken during delivery, as prudential planning improves survival, and reduces neonatal morbidity and mortality.⁵ The EXIT procedure allows the operating team to secure the fetal airway, without compromising fetal oxygenation, as uteroplacental circulation is maintained. In most cases, general anesthesia is the preferred technique of anesthesia for the EXIT procedure, due to it providing adequate uterine relaxation, maintaining the maternal hemodynamics and uteroplacental blood flow.¹⁻⁴ On the other hand, volatile anesthetic agents and intravenous induction/maintenance medications can be passed through the placenta into fetal circulation, due to their high lipid solubility properties. Thus, this can provide fetal sedation in various degrees; depending on the dosage.⁶



Figure 1 Successful intubation during the EXIT procedure

3

Currently, there are only a limited number of cases reports that advocate using neuraxial central blockade as a mode of anesthesia in the EXIT procedure. George et al., have published 3 case series regarding successful usage of combined spinal anesthesia with intravenous nitroglycerin in the EXIT procedure.⁷ Benonis et al. has also reported the successful use of continuous spinal anesthesia and intravenous nitroglycerin for an EXIT procedure in a patient with arthrogryposis multiplex congenita.⁸ Moreover, Fink et al. have 3 case reports regarding the use of combined spinal–epidural with intravenous remifentanil infusion for fetal immobilization and analgesia for an EXIT procedure.⁹

Central neuraxial blockade is currently the anesthetic of choice for a cesarean section, as it is associated with lower morbidity and mortality compared with adverse effects of general anesthesia from failed intubation, hypoxia, and risk of aspiration. Local anesthetics; such as Bupivacaine, have high protein binding properties and low degrees of ionization at physiological pH. Hence, at the spinal anesthetic dose, it produces less risk of going across the placenta into fetal circulation.⁶ Neuraxial anesthesia also has benefits in terms of better patient satisfaction, and good intra and postoperative analgesia. Additionally, it also offers an advantage in patients that are contraindicated for general anesthesia. Hypotension and inadequate uterine relaxation can happen during spinal anesthesia with use of the EXIT procedure.⁴ Preload fluids can reduce the incidence of hypotension from spinal anesthesia;¹⁰ furthermore, Nitroglycerine (NTG) has also been used for uterine relaxation during cesarean section.¹¹ In this present case, NTG was pre-prepared in the event that uterine relaxation was not sufficient. The patient had neither hypotension, nor inadequate uterine relaxation under the sole spinal anesthesia technique.

The recent systematic review by Kumar et al. reported that most of the EXIT procedures have been performed under general anesthesia.¹² To our knowledge, there have

been only 10 cases that have used the regional anesthesia technique combined with NTG infusion, or combined spinalepidural anesthesia for maternal anesthesia for the EXIT procedure. This study's results showed that there were no maternal or fetal complications correlated to the single-shot spinal anesthetic technique.

Conclusion

In this present case report, we want to emphasize that the sole spinal anesthesia technique can be used safely for the EXIT procedure.

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