

## CLINICAL INFORMATION

# The multidisciplinary challenge of anesthesia for ex utero intrapartum treatment: a case report



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### PALAVRAS-CHAVE

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**Abstract** The Ex Utero Intrapartum Treatment (EXIT) is a surgical procedure performed in cases of expected postpartum fetal airway obstruction, allowing the establishment of patent airway while maintaining placental circulation. Anesthesia for EXIT procedure has several specific features such as adequate uterine relaxation, maintenance of maternal blood pressure fetal anesthesia and fetal airway establishment. The anesthesiologist should be aware of these particularities in order to contribute to a favorable outcome. This is a case report of an EXIT procedure performed on a fetus with a cervical lymphangioma with prenatal evidence of partial obstruction of the trachea and risk of post-delivery airway compromise.

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### O desafio multidisciplinar da anestesia para procedimento intraparto extra-uterino: relato de caso

**Resumo** O procedimento Intraparto Extra-Uterino (EXIT) é procedimento cirúrgico realizado em casos de previsão de obstrução de via aérea fetal no pós-parto, que permite estabelecer via aérea patente enquanto a circulação placentária é mantida. A anestesia para o procedimento EXIT apresenta várias características específicas, tais como relaxamento uterino adequado, manutenção da pressão arterial materna, anestesia fetal e estabelecimento da via aérea fetal. O anestesiologista deve estar ciente dessas especificidades para contribuir para desfecho favorável. Trata-se de relato de caso de procedimento EXIT realizado em feto com linfangioma cervical e evidência pré-natal de obstrução parcial de traqueia e risco de comprometimento de via aérea pós-parto.

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**Figure 1** Fetal MRI revealing a cervical mass arising from the right region of the fetal neck (arrows).

## Introduction

The ex utero intrapartum treatment procedure was initially developed for the fetus with severe congenital diaphragmatic hernia. This technique allows uteroplacental circulation to be maintained during the removal of the tracheal clips immediately followed by caesarean delivery.<sup>1</sup> Since then, other indications to EXIT have arisen not only relate to airway compromise, but also congenital malformations that can result in perinatal death.

The procedure is usually scheduled after 35 weeks of gestation and it is combined with an elective cesarean section. Prematurity is not considered a contraindication.<sup>1</sup>

The fetus is partially delivered from the uterine cavity with the maintenance of uteroplacental circulation to preserve fetal gas exchanges, allowing therapeutic procedures to the fetus.

The anesthetic management includes deep volatile anesthesia with uterine relaxation, preservation of uteroplacental circulation and fetal anesthesia. This implies rigorous strategic planning with the involvement of a multidisciplinary team.

This article describes a case in which the EXIT procedure was successfully performed in a fetus with a prenatal diagnosis of cervical lymphangioma and discussed the anesthetic management in the light of current clinical practice.

## Case report

A 37 year-old woman, pregnant 2, para 0, was scheduled for an elective EXIT procedure at 38 weeks of gestation due to prenatal ultrasound diagnosis of fetal cervical lymphangioma with partial obstruction of the trachea and risk of post-delivery airway compromise. A Magnetic Resonance Imaging (MRI) test was performed at 31 weeks and confirmed a right cervical cystic formation with  $60 \times 38 \times 52$  mm, associated with tongue protrusion (Fig. 1, arrows).

The preoperative planning involved several meetings with a multidisciplinary team comprising anesthesiologists, obstetricians, neonatologists, pediatric surgeons, otorhinolaryngologists and pulmonologists. Every role and positioning in the operatory room were clearly defined. Anesthesia material, room temperature, blood grouping, hemoderiva-



**Figure 2** Endotracheal intubation performed on the partially delivered fetus.

tives availability and both neonatology and post-anesthetic care unit vacancies were all preoperatively confirmed.

In the operating room, the maternal monitoring included pulse oximetry, ECG and invasive blood pressure. The parturient was positioned in supine and left lateral tilt position with manual uterine displacement to the left. Two peripheral intravenous lines (18G and 16G) were placed and urinary catheterization was performed. Additional pharmacological preparation included drugs for supplementary intramuscular fetal anesthesia, namely, fentanyl, rocuronium and atropine.

Balanced general anesthesia was initiated with a remifentanil perfusion 2 minutes before induction. Rapid sequence was performed with propofol  $2 \text{ mg} \cdot \text{kg}^{-1}$  and rocuronium  $1.2 \text{ mg} \cdot \text{kg}^{-1}$ , followed by endotracheal intubation with a 7.5 cuffed tube and mechanical ventilation in a volume-controlled mode. Anesthesia was maintained with sevoflurane (2.5%) and remifentanil. For the maintenance of maternal hemodynamic stability, it was necessary to administer ephedrine 10 mg and goal-directed fluid therapy with crystalloids.

After hysterotomy, the head, trunk and upper limbs of the fetus were externalized and orotracheal intubation by direct laryngoscopy was performed with a 3.5 uncuffed tube. The intubation was achieved on the second attempt, by a neonatologist, 4 minutes after hysterotomy (Fig. 2). The correct positioning of the tracheal tube was confirmed, and the umbilical cord was clamped and cut. After full extraction, the newborn was stabilized and transported in a neonatal incubator under mechanical ventilation to the neonatology unit.

During the procedure, uterine relaxation was reasonably obtained with sevoflurane, and there was no need for additional tocolytic drugs. Uterine hypotonicity was effectively reversed with oxytocin and volatile concentration reduction. Remifentanil infusion was discontinued, and anesthesia emergence progressed uneventfully. Intravenous analgesia was performed with paracetamol, ketorolac and tramadol. Nausea and vomiting were prevented with droperidol and ondansetron.

The total volume of infused fluids was 1.000 mL of crystalloid and 500 mL of colloids. The estimated blood loss was

800 mL, and there was no need for transfusion of blood components.

## Discussion

EXIT is a surgical procedure performed when there is a prenatal diagnosis of a congenital malformation associated with high perinatal mortality. The objective is to maintain the fetoplacental circulation while optimizing the fetal condition in order to improve the transition from fetal to neonatal life.<sup>1,2</sup>

A success key factor of this procedure is to have a multidisciplinary team and a thoroughly designed plan. Of the six medical teams involved in this case, anesthesiology, obstetrics and neonatology were directly involved in the procedure. Participation of pulmonology, otorhinolaryngology and pediatric surgery was justified in the event of a difficult direct laryngoscopic approach, to execute a bronchoscopic intubation, establish a surgical airway or to perform a partial mass resection, respectively.

To provide and maintain anesthesia for this procedure is a clinical challenge since there are always two patients to consider, mother and fetus. General anesthesia is the preferred technique despite being associated with higher rates of mortality and morbidity among the obstetric population. Aside from its contribution to an adequate uterine relaxation, it allows simultaneous induction of both mother and fetus through placental penetration of anesthetic agents. In cases where general anesthesia is contraindicated a locoregional technique should be considered with an accompanying administration of tocolytic drugs to provide uterine relaxation.<sup>2</sup> The disadvantages of regional anesthesia are the risk of hemodynamic instability if massive haemorrhage occurs and the need of additional fetal anesthesia.

The main goals of this procedure are deep uterine relaxation for the prevention of placental separation and fetoplacental circulation preservation that ensures fetal oxygenation.<sup>2-4</sup> Simultaneously, the transplacental passage of the anesthetics is essential to provide fetal anesthesia.

Several authors have recommended inhalation anesthetic at concentrations of at least two Minimum Anesthetic Concentration (MAC), which in addition of allowing uterine relaxation, also provides deep anesthesia for mother and fetus.<sup>1,2</sup> However it has been reported that the use of 0,5-1 MAC combined with a tocolytic drug (nitroglycerin) might offer an effective strategy for uterine tone control with cardiovascular stability. Nitroglycerine is the best drug for this procedure due to its rapid and short-term onset, but it may have adverse effects like hypotension.<sup>5</sup> Other tocolytics drugs like terbutaline and magnesium sulphate can also be used to enhance uterine relaxation.<sup>4</sup>

It's crucial to prevent maternal hypotension and myometrial tone, in order to preserve fetoplacental gas exchange for fetal oxygenation.<sup>3</sup> Maintenance of maternal blood pressure within 10%-20% of baseline is therefore critical for adequate fetal oxygenation.<sup>3</sup> Fluid resuscitation and administration of vasopressors and inotropic drugs may be crucial to maintain maternal hemodynamic stability and fetoplacental circulation.<sup>2,4</sup> Invasive blood pressure monitoring

in addition to routine monitors is highly recommended for adequate control of hemodynamic stability.<sup>1-3</sup> One of the major intraoperative and postoperative complications is blood loss, which is usually correlated with uterine atony due to the tocolytic drugs or procedure duration.<sup>1-4</sup> The deterioration of fetoplacental blood flow is a considerable risk and may lead to fetal bradycardia and acidosis.

In our case, the depth of anesthesia obtained with 2.5% sevoflurane was satisfactory and provided a stable hemodynamic profile. The uterine relaxation was sufficient, and additional administration of tocolytic drugs was not necessary. The fetus did not react to the manoeuvres involving laryngoscopy and tracheal intubation, therefore, it was not necessary to use supplemental fetal anesthesia. After securing the fetal airway, full externalization with umbilical cord clamping was realized. The period of time between hysterotomy and umbilical cord clamping was approximately 4 minutes and 20 seconds. This is a shorter time concerning other cases in literature and seems to be a factor for fetal well-being.<sup>5</sup> Uterine hypotonicity was reversed with oxytocin and the discontinuation of the high inhalational anesthetic concentrations.

No reports of hemorrhagic incidents were issued following the transference to the post-anesthetic care unit.

## Conclusion

The EXIT procedure is an essential tool in the management of prenatally diagnosed congenital malformations because a life-threatening emergency at birth can be converted into an elective procedure. The intervention's success was fundamentally due to the planning of the procedure by a multidisciplinary team. Considering the current literature, anesthesia can be performed in various ways, as long as uterine relaxation and fetoplacental circulation are maintained until the fetal airway is secured.

The anesthesiologist should be aware of the particularities of this procedure and coordinated with the surgeons during each phase of the intervention, from uterine incision to the umbilical cord clamping.

## Authorship

All authors participated in the procedure described in the report. Data collection was performed by all. The report was drafted by A.C. and revised by all authors.

## Conflicts of interest

The authors declare no conflicts of interest.

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