Congenital Cervical Teratoma: Anaesthetic Management  
(The EXIT Procedure)  

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Summary  
Ex utero intrapartum treatment (EXIT) is a procedure performed during caesarean section with preservation of fetal-placental circulation, which allows the safe handling of fetal airways with risk of airways obstruction. This report aimed at describing a case of anaesthesia for EXIT in a fetus with cervical teratoma. A 30-year-old woman, 70 kg, 160 cm, gravida 2, para 1, was followed because of polyhydramnios diagnosis at 24 weeks’ gestation. During a routine ultrasonographic examination at 35 weeks’ gestation, it was noticed that the fetus had a tumoral mass on the anterior neck, the mass had cystic and calcified components and with a size of was 10 x 6 x5 cm. The patient with physical status ASA I, was submitted to caesarean section under general anaesthesia with mechanically controlled ventilation for exutero intrapartum treatment (EXIT). Anaesthesia was induced in rapid sequence with fentanyl, propofol and rocuronium and was maintained with isoflurane in 2.5 at 3 % in O₂ and N₂O (50%). After hysterotomy, fetus was partially released assuring uterus-placental circulation, followed by fetal laryngoscopy and tracheal intubation. The infant was intubated with an uncuffed, size 2.5 endotracheal tube. Excision of the mass was performed under general anaesthesia. After surgical intervention, on the fourth postoperative day, the infant was extubated and the newborn was discharged to the pediatric neonatal unit and on the seventh day postoperatively to home without complications. Major recommendations for EXIT are maternal-fetal safety, uterine relaxation to maintain uterine volume and uterus-placental circulation, and fetal immobility to help airway handling. We report one case of cervical teratoma managed successfully with EXIT procedure.  

Key words  
Cervical teratoma, Mass in the neck, Extrauterine intrapartum treatment procedure.  

Introduction  
Teratomas are germ cell tumour derived from pluripotential cells and consisting elements of different types of tissue from one or more of the three germ cell layers¹,². Commonly found in the sacrococcygeal region, teratomas may originate from any part of the body. In 3-5 % of cases, the teratoma may arise from the head and neck region, with mostly anterior and lateral placement. The mortality of newborn increases in proportion to the size of lesion without treatment. Early diagnosis with imaging methods and planning of delivery are the most important steps of the treatment at the postpartum period¹⁴.  

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procedure allows the fetoplacental circulation to continue during cesarean section and it is also helpful to apply invasive instrumentation to the neonate. Initially, only the infant’s head and shoulders are delivered, thus maintaining uteroplacental blood flow. After the infant’s airway is secured, umbilical cord is clamped and delivery of the infant is completed. Our case was diagnosed with cervical teratoma at the prenatal period and an EXIT procedure was planned because of expected difficulty of airway instrumentation. This procedure was successfully applied and the infant was intubated during oxygenation.

**Case**

A 30-year-old woman, 70 kg, 160 cm, gravida 2, para 1, was followed because of polyhydramnios diagnosed at 24 weeks’ gestation. During a routine ultrasonographic examination at 35 weeks’ gestation, it was noticed that the fetus had a tumoral mass on the anterior neck, which was protruding outwards under the jaw bone, pushing the floor of mouth upward, obliterating the trachea and esophagus and extending to the sternal notch (Fig 1). The mass had cystic and calcified components and with a size of was 10 x 6 x5 cm. The blood supply of the mass was poor. All of the findings were confirmed with CT scan too (Fig 2). The woman and her husband had no significant medical history. The case was diagnosed with cervical teratoma and evaluated with a team including anaesthesiologists, gynecologists, neonatal intensive care specialists and pediatric surgeons because of possibility of respiratory distress due to an airway obstruction at postnatal period. It was decided that the infant would be intubated immediately after the delivery which would be a cesarean section under elective conditions and the cervical mass would be excised if necessary. After information and explanation about the anaesthetic procedure, without anaesthetic background, physical status ASA I, was submitted to cesarean section under general anaesthesia with mechanically controlled ventilation for exutero intrapartum treatment (EXIT).

Patient was placed in the supine position and uterus was displaced to the left with the aid of Crawford’s wedge. Monitoring consisted of cardio-scope at DII lead, noninvasive blood pressure, pulse oximetry, capnography and neuromuscular block evaluation by acceleromyography. Patient was premedicated with intravenous metoclopramide (10 mg) and ranitidine (50 mg), 30 minutes before anaesthesia. Upper limb vein was catheterized in the operating room at room temperature with disposable 14G catheter for volume replacement and drug administration. General anaesthesia was induced in rapid sequence, oxygenation with 100 % oxygen under mask, intravenous fentanyl (250 µg), propofol (140 mg) and rocuronium (50 mg), Sellick maneuver and tracheal intubation. Anaesthesia was maintained with isoflurane in 2.5 % concentration at 3% through gauged vaporizer and administered in mixture of O2 and N2O (50 %). Maternal arterial oxygen saturation remained at 100 % during the entire procedure. Fetal was partially released (head, shoulders and upper limbs) after hysterotomy and, after assuring fetal-placental circulation. The intubation procedure was started while uteroplacental blood flow was still continuing. During intubation, Cormack and Lehane direct laryngoscopy score was evaluated as grade 4 (Fig 3). Oxygen peripheral saturation and fetal pulse frequency were continuously evaluated during the procedure with the aid of pulse oximetry and sterile sensor placed on right hand, which were maintained in approximately 70
SpO2 has increased to 92% and pulse frequency was maintained in approximately 100 bpm. Fetus was then totally released, umbilical cord was clamped and uterus was continuously sutured. Isoflurane concentration was gradually decreased and oxytocin (20 UI) continuous infusion and intravenous methyl-ergonovine (0.2 mg) were administered to reestablish uterine tone. Maternal end-tidal carbon dioxide ranged from 25 to 35 mmHg and maternal systolic blood pressure was over 100 mmHg all through the surgery. The infant was intubated with an uncuffed, size 2.5 endotracheal tube. Fetal airway handling and tracheal intubation were performed in 3 minutes, the cord was clamped after the correct endotracheal tube positioning was confirmed and the airway was secured, with total surgery duration of 80 minutes. The infant was transferred to another operation room where the resection of the cervical mass would be performed by pediatric surgeons. Anaesthesia was maintained with routine procedure. At the operation, the cervical mass was observed to be attached in places to anterior group muscles of the neck and pushed the larynx posterolaterally. Newborn presented Apgar scores of 6 and 9 at 1 and 5 minutes, respectively, being referred to neonatal ICU with spontaneous ventilation. The diagnosis may be achieved antenatally with ultrasonographic examination. Polyhydramnios accompanies in 20-30 % of cases. The rate of stillbirth is reported in 17 % of cases3. The mortality rate is reported to be 9-17 % in the surgically cases treated4.

Jordan and Gauderer5 reviewed 163 congenital cervical teratoma cases and classified them in three groups based on the survey. They are; a- Premature infants b- Newborn with respiratory distress c- Newborn without respiratory distress. The mortality rates of the groups were reported as 100 %, 43.4 % and 2.7 %. Our case was in the newborn with respiratory distress group defined as group b.

Early diagnosis and multidisciplinary management are extremely important. Formation of a multispecialty team and use of the EXIT procedure is essential for survival of the neonate. The EXIT procedure was described first in 1990 by Zerella and Finberg for tra-
cheal obstruction in a neonate and has been refined by Mychaliska et al. In the EXIT procedure, deep inhalational anaesthesia ensures uterine relaxation, which is crucial to preserving uteroplacental gas exchange. The EXIT procedure allows for the management of newborns diagnosed antenatally with extrinsic (lymphangiomas, teratomas) or intrinsic (laryngeal atresia, congenital upper airway obstruction syndrome) obstructive malformations. EXIT, for maintaining uterus-placental circulation and, as a consequence, adequate fetal oxygenation during the time needed for airway handling (laryngoscopy, bronchoscopy, tracheal intubation or tracheotomy), is a common procedure in such situations.

The most important step of the management is providing a safe airway for newborn while the uteroplacental flow is continuing during delivery. Shih et al. reported that they secured the airway via tracheostomy for a case with pretracheal teratoma at the 51 minute of the delivery. Mychaliska et al. proposed that this period may be prolonged to 60 minutes with deep inhalation anesthesia. In our case, we have intubated the newborn at the fifth minute after skin incision. In the literature there are different methods for the maintenance of uteroplacental blood flow. Prophylactic indomethaene may be applied to the pregnant for tocolysis. Nitroglycerin and terbutaline may be used to reduce the uterine tonus perioperatively. High dose inhalation anesthetic agents (2-3 MAC) may be applied to achieve uterine relaxation. In our case, we achieved uterine relaxation via inhalation of 2-3 % sevoflurane deep inhalation anaesthesia and so we gained extra time for securing the airway while the uteroplacental blood flow continued.

Although the uterine tonus is reduced, it is important to maintain sufficient maternal systolic blood pressure for the continuation of placental blood flow. For this purpose, efficient treatment with relaxant agents, vasopressors and fluid resuscitation is necessary. Angiotensin II infusions are useful to maintain the systolic blood pressure over 100 mmHg or the blood pressure may be increased with small boluses of iv ephedrine when blood pressure declines. In our case, the heat loss was minimized, the umbilical cord was not endangered and premature placental separation was prevented with delivery of only head and shoulders.

It is emphasized that placement (occupation) of the fetus at the same level with the placenta may prevent excessive hydrostatic pressure over the fetoplacental circulation and also prevent premature placental separation. We have paid attention to this point in our case. The surgical excision of the mass is the most appropriate treatment for congenital cervical teratomas. In our case, the mass was completely removed from surrounding tissues. Respiratory distress persisted until the fourth day postoperatively, and the newborn was extubated on the fourth day.

Congenital cervical teratomas are one of the rare congenital abnormalities which necessitate early antenatal diagnosis and multidisciplinary approach. Otherwise, the mortality rate will be high. We have already made our crucial preparations for the case who was diagnosed at the antenatal period and planned a cesarean section. In this manner, we have secured the newborn’s airway with suitable interventions. The EXIT procedure allows therapeutic interventions on the neonate while maintaining fetoplacental circulation and thereby maintaining oxygenation. These cases highlight the possible airway scenarios that may confront the anaesthetist in the immediate postpartum, elective surgery and postoperative stages and the variety of techniques that may be employed in order to overcome the potential difficulties encountered.

In conclusion, major recommendations for EXIT are maternal-fetal safety, uterine relaxation to maintain uterine volume and uterus-placental circulation, and fetal immobility to help airway handling. Formation of a multispecialty team and use of the EXIT procedure is essential for survival of the neonate. We report one case of cervical teratoma managed successfully with EXIT procedure.
References

1. Green JS, Dickinson FL, Rickett A, Moir A. MRI in the assessment of a newborn with cervical teratoma. Pediatr Radiol 1998; 28:709-10.

2. Elmasalme F, Giacomantonio M, Clarck KD, Othman E, Matbouli S. Congenital cervical teratoma in neonates. Case report and review. Eur Pediatr Surg 2000;10:252-7.

3. Newstedt JR, Shirkey HC. Teratoma of the thyroid region. Am J Dis Child 1964; 107:88.

4. Gundry SR, Weley JR, Klein MD. Cervical teratomas in the newborn. J Pediatr Surg 1983; 18:382-6.

5. Jordan RB., Gauderer MW. Cervical teratomas: an analysis. Literature review and proposed classification. J Pediatr Surg 1988;23:583-91.

6. Zerella JT, Finberg FJ. Obstruction of the neonatal airway from teratomas. Surg Gynecol Obstet 1990; 170:126-31.

7. Mychaliska GB, Bealer JF, Graf JL, Rosen MA, Adzick, NS, Harrison MR. Operating on placental support: The Ex Utero Intrapartum Treatment procedure. J Pediatr Surg 1997; 32:227-31.

8. Shih GH, Boyd GL, Vincent RD Jr, Long GW, Hauth JC, Georgeson KE. The EXIT procedure facilitates delivery of an infant with a pretracheal teratoma. Anesthesiology 1998;89:1573-5.

9. Dahlgren G, Törnberg DC, Pregner K, Irestedt L. Four cases of the ex utero intrapartum treatment (EXIT) procedure: anesthetic implications. Int J Obstet Anesth 2004;13:178-82.

10. Hasiotou, M Vakaki, G Pitsoulakis, M Zarifi, H Sammouti et al., Congenital cervical teratomas. Int J Pediatr Otorhinolaryngol 2004;68:1133-9.

11. Hirose S, Harrison MR. The ex utero intrapartum treatment (EXIT) procedure. Semin Neonatol 2003;8:207-14.