Case Report Anesthetic management of a neonate receiving prenatal repair of gastroschisis

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Abstract: Gastroschisis requires surgical repair, which is generally performed after birth. We report a case in which a fetus with gastroschisis underwent the abdominal wall defect repair before birth. To ensure reliable operating conditions for the repair (to prevent fetal movement and crying), the fetus received deep anesthesia via placental transfer of maternally administered anesthetics. Meanwhile, the ex utero intrapartum treatment procedure was performed to ensure fetal oxygen supply, which was likely to be compromised by the deep fetal anesthesia. The procedure last for 23 minutes and the gastroschisis was successfully repaired before the neonate was delivered. Maternal hemodynamics was kept stable during this surgical procedure. The prenatal repair of abdominal wall defect is safe for the mother and the fetus, which could potentially improve the neonatal outcomes.

Keywords: Gastroschisis, fetal therapy, general anesthesia, EXIT procedure

Introduction

The diagnosis of gastroschisis is made antenatally in most cases, which, at least theoretically, provides a potential window for early repair of this congenital abdominal wall defect [1]. In this case report, we described the anesthetic considerations of a successful prenantal repair of gastroschisis. Briefly, this neonate received deep anesthesia to ensure immobility during surgical manipulation, while the oxygen supply was maintained by the *Ex Utero* Intrapartum Therapy (EXIT) procedure *via* uninterrupted utero-placental circulation [2, 3]. The maternal vital signs were well maintained during this procedure.

Case report

Patient consent was obtained for publication of this case report, which was in accordance with our institutional guidelines.

A 23-year old, gravid 1, woman at 39⁺⁴ weeks' gestation was referred to our hospital, after routine prenatal ultrasound imaging suggested the presence of fetal abdominal malformation. Detailed fetal ultrasound inspection revealed

that there was a defect in fetal abdominal wall, and extra-abdominal bowel loops. The development of the fetus was otherwise normal. A diagnosis of gastroschisis was concluded.

A multidisciplinary team was soon organized, including anesthesiologists (specialized in obstetric and pediatric anesthesia), obstetricians, pediatric surgeons, neonatologists, and nurses. All the members agreed to repair the defect before the fetus was delivered. Considering the gestational age of more than 39 weeks, an EXIT procedure was planned to provide fetal anesthesia and oxygen supply, to facilitate the repair of the abdominal wall defect immediately before the neonate was delivered.

The consent form was obtained from the patient. On arrival at the operation room, the patient was placed in the supine, left uterine displacement position and received standard anesthetic monitoring. Fetal heart rate was monitored continuously by Doppler ultrasonog-raphy. Two large-born (16G) intravenous lines were inserted in forearm, and radial artery was cannulated under local anesthesia for invasive blood pressure monitoring. Then, 500 mL Ringer's solution and 500 mL colloid were



Figure 1. Newborn infant with gastroschisis delivered via the ex utero intrapartum technique procedure. A. With the maintained utero-placental circulation (red arrow showed the unclamped umbilical cord), the pediatric surgeons return the extra-abdominal intestines (black arrow) to the abdominal cavity. B. After returning the intestines, the umbilical cord was clamped and the abdominal wall defect lied to the right of the umbilical cord.

given, to restore volume deficit due to overnight fasting and prevent hypotension during EXIT procedure. The patient was anaesthetized by rapid sequence induction with propofol 120 mg and succinylcholine 100 mg. After confirmation of proper placement of the tracheal tube, sufentanyl 15 µg, midazolam 3 mg and vecuronium 4 mg were supplemented. To produce profound uterine relaxation, deep inhaled anesthesia was applied with sevoflurane, and end-tidal concentration was set to 4%. Ringer's solution infusion was continued at a rate of 10 ml/kg/h till EXIT procedure completed. Nitroglycerin and phenylephrine infusion were available to provide extra uterine relaxation and maintain maternal blood pressure, when indicated. To ensure the fetal well-being during deepening of maternal anesthesia, the fetal heart rate was monitored continuously till surgery started, which was maintained within the normal range of 110-160 bpm.

Before skin incision, another sufentanyl 15 μ g was given. A vertical skin incision was made and uterine was exposed. The uterus was well relaxed, as evaluated by obstetrician with manual pulsation. Therefore, no extra nitroglycerin was infused. After incising the uterus, the newborn was exteriorized. The uterine incision was clamped to reduce maternal blood loss and pre-warmed normal saline was infused into the uterus to maintain the uterine volume. The newborn was immediately intubated and received midazolam 1 mg and vercuronium 1 mg via i.m. injection into the deltoid muscle. At the same time, the pediatric surgeons returned

the extra-abdominal bowel loops to the abdominal cavity after careful exploration (Figure 1), and covered the abdominal defect with gauze. The fetus was well anaesthetized and the pediatric procedure went on smoothly. No fetal movement or crying occurred. During pediatric manipulation, the obstetrician monitored the fetal heart rate via pulsation of the cord (which varied between 120 to 140 bpm) and the uterus wall tension by manual pulsation. Delivery was completed 27 minutes after skin incision and the EXIT last for 23 minutes. After pediatric procedure completed, the obstetrician clamped the cord and the neonate was transferred to a pre-warmed incubator. The neonate was then connected to a ventilator. A pulse oximetry probe was applied to the neonate and SpO was maintained at higher than 90%. Intravenous line and gastric tube were inserted respectively. Then the newborn was transferred to neonatal ICU. During the EXIT procedure, maternal vital signs were well maintained. After clamping the cord, the uterine relax was effectively reversed with 10 IU of oxytocin infusion and end-tidal sevoflurane was decreased to 2%. After surgery, the patient recovered from anesthesia uneventfully and discharged on postoperative day 4. The estimated maternal blood loss was 700 mL during this procedure.

The neonate was stable for the first 24 hrs and was then extubated. But 48 hrs later the status deteriorated and the pulmonary hypertension was noted by echocardiograph. Tragically, the newborn died 3 days later due to severe hypoxia. As evaluated by an academic panel, the prenatal closured of gastroschisis was not considered to be involved with the death.

Discussion

Gastroschisis is a birth defect in abdominal wall through which an infant's intestines stick out of the body on one side of the umbilical cord [1]. There is no doubt that surgical repair is the only treatment option for gastroschisis. However, the timing for surgical repair is controversial. Current evidences are not able to distinguish any difference in survival rate between neonates receiving immediate closure and infants receiving delayed closure [4], but some data did show that neonates receiving immediate closure had a shorter time to reach full enteral feeding [5] and lower incidence of wound infection [6]. It is therefore rationale for us to believe that the neonates could benefit from even earlier, i.e. prenatal, repair of the defect.

Considering the gestational age of more than 39 weeks, a decision was made to repair the defect just before the newborn was delivered then the newborn would be allowed to develop spontaneous respiration with a normal abdominal anatomy. Therefore, an EXIT procedure was planned, which could 1) provide deep fetal anesthesia to prevent body movement or crying during the surgical manipulation, which would increase intra-abdominal pressure and deteriorate the evisceration of the intestines; and 2) *via* maintained utero-placental circulation, ensure oxygen supply to the deeply anaesthetized fetus who is very likely to develop respiration depression or apena.

Traditionally in operations on placental support (such as EXIT procedure and fetal surgery), volatile anesthetics were applied at high concentrations (e.g. 2 MAC) for profound uterine relaxation, which would provide adequate maternal analgesia and spare the use of additional opioids [7]. In the meanwhile, the neonates also received anesthesia via placental transfer of maternally administrated anesthetics. It is not clear yet how effective the "transferred anesthesia" could be in blocking neonatal responses. Based on the fact that opioids and muscle relaxants were always administrated directly to the newborn during EXIT procedure in published case reports [8] and the results of pilot study on placental transferring of volatile anes-

thetics [9], it is rational for us to believe that volatile anesthetic alone, even at high concentrations, cannot ensure fetal immobility. As mentioned above, prevention of body movement and crying was of vital importance for the success of this procedure. Therefore, a relatively large dose of sufentanyl (30 µg vs 15-20 µg for routine caesarean section under general anesthesia) plus 4% sevoflurane were coadministrated. The transferred sevoflurane, acting synergically with transferred sufentanyl of high fetal-maternal ratio [10] should produce desired anesthesia in the neonate, as demonstrated in this case. Another concern for avoiding using volatile anesthetic alone was that high dose of volatile anesthetic has been reported to cause fetal bradycardia at high concentration [11]. To prevent unexpected interruption of utero-placental circulation, the newborn also received intubation and intramuscular injection of anesthetics as precaution measures. It was surprised to us that maternal hypotension was not observed in this case, and we think the stable hemodynamics may result from the relatively liberal fluid management strategy, in addition to relatively lower concentration of sevoflurane.

This case also raised several important questions. First, guideline is lacking on how should the newborn be monitored and how could we correct abnormal fetal vital signs, such as bradycardia, with very limited vascular access? Second, how would the anesthetics affect the long-term neurologic development of the fetus and what is the best anesthetic regimen for this procedure? Finally, but most importantly, whether this prenatal closure of abdominal wall defect could translate to better fetal outcomes? To answer these questions, multiple-centre, randomized prospective trials were warranted!

In summary, prenatal repair of gastroschisis is safe for both mother and fetus. The success of this novel procedure relies on close multidisciplinary cooperation. Further study should focus on the long-term benefit of this novel procedure.

Disclosure of conflict of interest

None.

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