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CASE REPORT

Ex Utero Intrapartum Treatment (EXIT) for a Large Fetal Neck Mass

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Introduction

A 23-year-old primigravida was referred to our hospital at 36 + 5 weeks gestation with scan showing gross polyhydramnios and fetus having a large neck mass. She had no antenatal risk factors. An anomaly scan done at 22-week gestation was told to be normal.

Repeat scan done here showed gross polyhydramnios with the amniotic fluid index (AFI) being 44 cm. The fetus was growth restricted with an estimated fetal weight of 1.8 kg. There was a 8 × 6 cm left anterolateral mass on the fetal neck, which was solid, well defined with minimal vascularity (Fig. 1). There seemed to be no retrosternal extension of the mass. There was no other structural abnormality to account for the fetal growth restriction (FGR) with polyhydramnios. Her glucose tolerance test at fifth month was normal.

Based on the above scan findings, a probable diagnosis of cervical teratoma or lymphangioma was made.

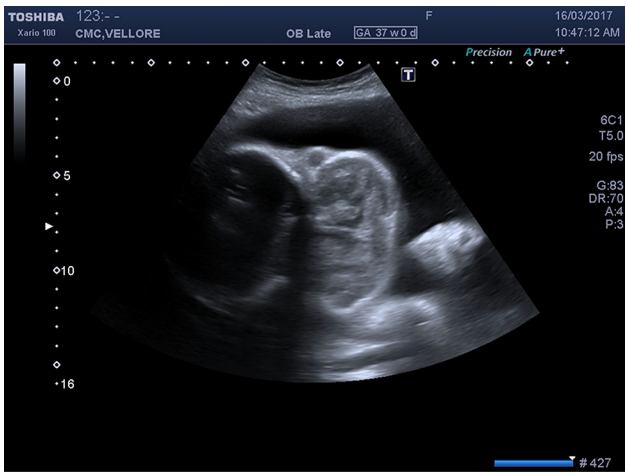

Fig. 1 Prenatal USG of 37-week fetus showing 8 × 6 cm cervical mass, lying next to fetal head. The mass appeared solid, with heterogenous appearance and minimal vascularity on *Color* Doppler

Compression of the esophagus due to the neck mass was thought to be the cause of gross polyhydramnios. Fine needle aspiration biopsy of mass was not carried out since, being an invasive procedure, it might initiate labor. Magnetic resonance imaging (MRI) was not performed since the neck mass appeared benign on scan. The presence of polyhydramnios resulted in excessive fetal movements which would have made MRI technically challenging.

Since there was a high probability of hypoxia of the newborn after birth due to huge mass obliterating the airway, the decision to deliver the fetus using EXIT procedure was made.

A multidisciplinary team, comprising of senior obstetricians, neonatologists, pediatric anesthetists and pediatric otolaryngologists, was set up. Following several meetings, a detailed algorithm of steps to secure the fetal airway was formulated. Plan was made to attempt endotracheal intubation followed by rigid bronchoscopy, and tracheostomy was kept as a rescue plan. The time limit for each procedure was set at 5, 10 and 15 min after the partial delivery of the fetus.

A detailed consent was obtained from the parents regarding the risks involved, including risks of perinatal death (in case airway could not be secured) and/or fetal brain damage. The maternal complications such as postpartum hemorrhage, need for massive blood transfusion and ICU (intensive care unit) care were also explained. Four units of whole blood were arranged preoperatively. A mock drill was carried out by the team a day prior to surgery, during which details such as position of the personnel and equipments, patient's position and movement of doctors according to planned sequence were discussed. Severe fetal bradycardia (fetal heart rate $\leq$80 beats per minute) and/or evidence of placental separation were accepted as thresholds for abandoning the procedure.

General anesthesia (GA) was planned for the mother along with invasive arterial blood pressure monitoring and adequate intravenous access. Uterus was kept relaxed with two minimal anesthetic concentration (MAC) of isoflurane. Additionally, terbutaline (250 mcg subcutaneously) and magnesium sulfate were required to maintain uterine relaxation.

Laparotomy was done using Pfannenstiel incision. Once the desired relaxation was achieved, the uterus was opened by low transverse incision, well away from the placenta. The uterus was opened cautiously such that the bag of membranes was left intact. Controlled artificial rupture of membranes (ARM) was done using spinal needle to avoid sudden decompression of the uterus which would accelerate placental separation.

The fetal head and the upper torso of the fetus, including the right upper arm, were carefully delivered by the obstetricians. The lower torso, cord and the placenta were left inside the uterine cavity to keep the uteroplacental circulation going. Amnioinfusion, using 1 l of warm ringer lactate, was carried out to maintain the uterine volume. A pulse oximeter was connected to the right hand of the fetus to check the oxygen saturation levels. A neonatologist monitored the fetal heart by continuous auscultation during the entire procedure.

The fetal airway was secured by the coordinated efforts of the pediatric anesthetist, otolaryngologists and neonatologists. Since the mass impinged on the field, the mass was lifted laterally by one of the assistants, after which the airway could be visualized (Fig. 2). An endotracheal tube size 3 mm was negotiated through the airway, successfully in the second attempt by direct laryngoscopy. The position of the ET tube was checked using rigid bronchoscope and measuring the ET CO_2 (end-tidal carbon dioxide levels). It took 7 min to secure the fetal airway.

Once the airway was secured, the rest of the fetal body was delivered. Cord was clamped and cut, and baby was handed over to the neonatologists. The isoflurane was immediately stopped, and intravenous oxytocin infusion was started, using 20 units of oxytocin in 500 ml of normal saline. Placenta was allowed to separate spontaneously, and uterotonics were given prophylactically to help contract the uterus. Fortunately, our patient did not have much blood loss. The total blood loss during the procedure was 700 ml.

The girl baby, weighing 1.8 kg at birth was immediately shifted to level 3 nursery. The cord pH was 7.25 with a base deficit of -8.7. Baby had an MRI which confirmed the prenatal findings, and a differential diagnosis of soft tissue sarcoma, undifferentiated teratoma or neuroblastoma was suggested. Biopsy, however, showed malignant rhabdoid tumor. Due to poor prognosis associated with the

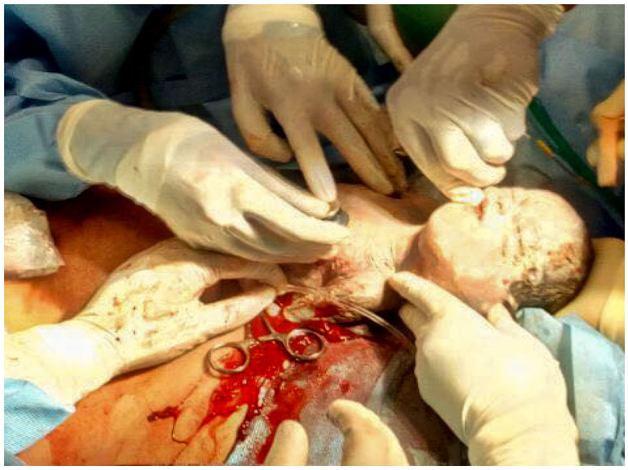

Fig. 2 Having delivered the head and the upper torso of the baby, the airway is being secured by the team. The neck mass lifted up to secure the airway by direct laryngoscopy. The fetal heart rate was continuously being monitored by the neonatologist using a sterile stethoscope

condition, parents opted for palliative care. Baby died on Day 5 of life.

Discussion

The EXIT procedure is a life-saving procedure for delivery of fetuses with airway obstruction. When such fetuses are delivered by traditional lower segment Caesarean section (LSCS) perinatal death and/or hypoxia are likely to occur, due to lack of airway. In the EXIT procedure, the partially delivered fetus is maintained on placental circulation till its airway is established [1]. The benefits of the EXIT procedure to the fetus have to be balanced against the maternal risks involved [2].

EXIT procedure, in contrast to the conventional LSCS, requires the use of general anesthesia for uterine relaxation. This is of paramount importance in preserving the uteroplacental gas exchange, while attempts are being made to secure the fetal airway. This, in turn, may lead to uterine atony and may lead to massive obstetrical hemorrhage [3]. Despite these theoretical concerns, our patient had blood loss of only 700 ml. A review of the EXIT procedure has also found the risks of PPH to be more theoretical than real [1].

The neonatal outcome following EXIT procedure depends mainly on size, location and underlying etiology of neck mass [4]. In some cases, early neonatal death has been reported in spite of successful establishment of the airway due to associated pulmonary hypoplasia [5]. In our case, although the EXIT procedure was successful in securing the complicated airway, we could not salvage the baby due the presence of malignant neck tumor.

Conclusion

The EXIT procedure is a safe and effective method of securing airway in fetuses with airway obstruction due to large neck masses. This advanced perinatal resuscitation technique requires careful planning and execution by a skillful multidisciplinary team. Despite a successful EXIT procedure, the neonatal outcome depends on the nature of the neck mass.

Compliance with Ethical Standards

Conflict of interest Manisha Beck, Ekta Rai, Reeta Vijayaselvi, Mary John, Naina Picardo, Sridhar Santhanam, Maneesh Kumar and Bejamin Ross declare they have no conflict of interest.

Ethical Statements All procedures performed on our patient were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendment or comparable ethical standards.

Informed Consent Informed consent was obtained from the patient whose details are included in the case report. Additional consent was obtained for publication of scan images and the photograph of the baby.

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