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Natal and Postnatal Approach in a Fetus with Intrauterine Cystic Hygroma: Case Report

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Abstract: Cystic hygroma (CH) is a neonatal abnormality that may cause respiratory problems especially due to pressure of huge masses located in the neck and thorax on the vital organs. A mother already known to have an intrauterine baby with CH underwent operation under general anesthesia. The newborn, who had cyanotic and respiratory problems, was immediately intubated and operated the day following delivery. This case report is intended to elaborate and discuss anesthesia in a newborn with CH and the mother.

Key Words: Cystic hygroma, EXIT procedure

Kistik Higromali Bebeğe Doğum ve Doğum Sonrası Yaklaşım: Olgu Sunumu

Özet: Kistik higroma (KH), özellikle infantlarda boyun ve toraks girişine yerleşerek, siyanoz, stridor, apne ve disfajiye neden olan lenfatik sistemin spesifik tümörünü tarif etmek için kullanılmaktadır. KH olarak doğacağı tespit edilen bebeklerin solunum yolu basısı olacağı düşünülerek normal doğum yapılması önerilmemektedir. KH olgularında büyük kitleler hayati organlara bası yaparak solunum problemlerine neden olduğu için entübasyon ve trakeostomi seti hazır bulunmalıdır. Fetüse kord klemplenmeden entübasyon yapılması ideal olanıdır. Entübe edilemezse lezyonun parsiyel rezeksiyonu veya trakeostomi yapılabilir.

Anahtar Sözcükler: KH, EXIT Prosedür

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Introduction

Cystic hygroma (CH) is a term used to define a specific tumor of the lymphatic system mostly seen in children that is typically located around the neck. It is observed in 1/12,000 births. This abnormality is detected in 50-65% of the cases at birth and in 80-90% of the cases before two years of age (1). CH presents as painless soft tissue masses ranging from a few millimeters to enormous volumes located in the neck, axilla, anterior wall of the abdomen, or inguinal region. Most of the cases are diagnosed at birth and cysts continue to expand slowly. Pressure on the nerves and some vital organs can lead to numerous symptoms depending on localization (2). Cyanosis, stridor, apnea and dysphagia may be observed particularly in infants if the cysts are localized in the thoracic inlet. Obstruction of the airway is the most frightening complication (3). We present a case report of the anesthetic management of a parturient who had a fetus with CH *in utero*-term and of the newborn.

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Case Report

An emergency cesarean section was performed for a 23-year-old multigravida (3 live births out of 4 pregnancies) at 36 weeks of gestation due to fetal abnormality. Preoperative vital signs were blood pressure 100/70 mmHg and pulse 92 beat/min, and laboratory examinations revealed hemoglobin 10.1 g/dl, WBC 10300/mm³, platelet 432000/mm³, and blood glucose 82 mg/dl. On admission, she had recently eaten and preoperative antiemetic 10 mg of intravenous (i.v.) metoclopramide HCL (primperan, Biofarma) and H2 receptor blocker of 50 mg i.v. ranitidine (ulcuran, Mefar) were administered. The patient was positioned on her left side to achieve uterine

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displacement. Standard monitoring was performed (electrocardiogram, pulse oximetry, arterial blood pressure) and external fetal heart rate monitor was applied. The patient was intubated after induction by 5 mg kg⁻¹ thiopentone sodium (pentothal, İ. E. ULAGAY) and 0.6 mg kg⁻¹ rocuronium bromide (esmeron, Organon), and anesthesia was maintained with 3% sevoflurane (sevorane, Abbott), 50 nitrous oxide and oxygen, rocuronium 7 mg and fentanyl 200 mcg. After clamping the fetal umbilical cord, fentanyl citrate (fentanyl, USP Abbott) was infused at a rate of 1.5 µg kg⁻¹ h⁻¹. A nitroglycerin infusion at 25 mcg/min was initiated to ensure uterine relaxation and maternal blood pressure was maintained with 6 mcg kg⁻¹min⁻¹ of dopamine (sterile dopamine, Fresenius Kabi) and lactated Ringer's solution. During the delivery of the fetus, tracheostomy set was prepared together with an intubation set due to the risk of difficult intubation. The fetus was cyanotic at birth and had a soft mass measuring 15 × 30 cm extending from the inferior margin of the orbita down to occupy the whole neck and showed ecchymotic areas on its surface (Figure 1). The fetus was monitored by a pulse oximeter probe, which was attached to the cleaned hand. The newborn was bradycardic (58 beat/min) and tachypneic (60 beat/min); intubation was achieved on the third attempt. Atropine 0.2 mg kg⁻¹ was administered to treat bradycardia and fentanyl 20 µg kg⁻¹ intramuscular (i.m.) was used to provide analgesia. After delivery, nitroglycerin and dopamine infusions were discontinued, and sevoflurane was reduced to 1.5%.



Figure 1. A cystic hygroma case was intubated after birth.

Intravenous oxytocin infusion was given to enhance uterine contraction. On physical examination, blood pressure of the newborn was 76/34 mmHg⁻¹, pulse was 134 beat/min, axillary temperature was 36.4 °C, and eyes were edematous. EtCO₂ was between 29-30 mmHg, and oxygen saturation between 90-92 was noted. His echocardiography showed normal cardiac chambers, an aortic valve with three cusps and 13 mm gradient shunt on the pulmonary artery that was consistent with patent ductus arteriosus (PDA). Ejection fraction (EF) was 83% and a 2/6 systolic murmur was detected at the left inferior margin of the sternum. The newborn was connected to a mechanical ventilator in the neonatal intensive care unit. The decision was then made to attempt to ventilate the fetus with oxygen. Dextrose 10%, 80 cc kg⁻¹ and dopamine 7 µg kg⁻¹ min⁻¹ infusion was used to correct low blood pressure and 3 µg kg⁻¹ h⁻¹ fentanyl citrate infusion was started. Intravenous fluid infusion rate was increased to 120 ml kg⁻¹ considering hemorrhage within the mass and volume loss. Laboratory examinations revealed: hemoglobin 15.7 mg/dl, hematocrit 44.1%, platelets 250,000/mm³, WBC 17,900/mm³, glucose 61 mg/dl, K 3.3 mmol/L, SGOT 49 U/L, SGPT 5 U/L, Na 131 mmol/L, Ca 9.7 mg/dl, Mg 1.8 mg/dl, Cre 0.6 mg/L, TBil 2.6 mg/dl, DBil 0.4 mg/dl, albumin 3.3 mg/dl, CK 391, LDH 844 U/L, PT 16.1%, and PTT 45.4. Fresh frozen plasma (20 ml kg⁻¹) was also administered preoperatively.

The newborn was operated on the day following delivery. Pulse, blood pressure, oxygen saturation and end-tidal carbon dioxide levels were monitored. General anesthesia was maintained with 1.5-2% sevoflurane, 50% oxygen and 3 µg kg⁻¹ h⁻¹ fentanyl citrate infusion. The newborn was manually ventilated throughout the procedure. Blood pressure remained low during the operation and dopamine was administered as 7 µg kg⁻¹ min⁻¹ infusion. Dextrose 10% was used for maintenance. Inhalation agents were discontinued at the end of the surgical procedure (Figure 2). After the operation, the newborn was kept intubated in the neonatal intensive care unit and 3 µg kg⁻¹ h⁻¹ fentanyl citrate infusion was continued. Controlled ventilation was done but pneumonia and bilateral pneumothorax developed on the postoperative fifth day in the Pediatrics Department despite an in situ chest tube. The infant remained hospitalized for several weeks.



Figure 2. A cystic hygroma case after operation.

Discussion

Cystic hygroma is a neonatal abnormality that may cause respiratory problems due to mechanical pressure of huge masses located in the neck and thorax on the vital organs (1). They are believed to be caused by incomplete development of lymphatic jugular sacs or their relation to the venous system and insufficient drainage to the venous system. These lymphatic lakes may enlarge to infiltrate the surrounding tissues over time (4). Spontaneous regression is very rare in CH (5).

Injection of sclerosing agents into the cyst has been shown to be effective in uniloculated cysts, but has limited success in multiloculated cysts. Recently, OK-432 injection and bleomycin was used, and was reported to be successful in treatment of CH (6). Many authors recommend delaying intervention until the child is 6 months old if the cysts are not of considerable size (2). However, early surgical treatment is justified if vital organs are compromised by direct pressure. It is imperative to excise the mass completely. Recurrence is usually observed within several weeks after the surgery, but occasionally it may take several months. In our case, a small uniloculated cyst presented three weeks later at the same location and was aspirated.

Two anesthesia methods are used for these patients. Regional anesthesia is the method of choice, as it does not cause fetal depression and has no deleterious effect on uteroplacental blood flow or relaxation of the uterus (7). The second method is balanced anesthesia with or

without epidural anesthesia. General anesthesia is preferred over a regional technique. Prevention of uterine contractions and placental separation and preservation of the uterine and placental blood flow following hysterotomy and partial delivery of the fetus are the hallmarks of the *ex utero* intrapartum treatment (EXIT) procedure (8). Profound uterine relaxation is mandatory to prevent uterine contractions and placental separation. Several authors have recommended inhalation anesthetic at concentrations of at least 2 MAC, which in addition to causing uterine relaxation also provides deep anesthesia for both the mother and fetus (9,10). Prior case reports describe intramuscular paralysis of the fetus (8), but Chang et al. (9) did not find this necessary. We used the balanced anesthesia technique in our case. Sevoflurane (2 MAC) was preferred during clamping of the umbilical cord, after which we started the fentanyl infusion. High concentration inhalation anesthetic agents with tocolytics can lead to uterine atony. We thus did not prefer using the inhalation agent above 2 MAC. Rapid induction and orotracheal intubation are necessary to avoid supine hypotension syndrome and the risk of aspiration of the mother.

Pregnant women may have mucosal edema, which can cause difficult intubation; thus, small sized tubes such as 6-6.5 should be used to reduce the potential airway complications in the postoperative period. There was no intubation difficulty in our case. Induction of cesarean section was achieved by 5 mg kg⁻¹ thiopentone sodium, 2 mg kg⁻¹ succinylcholine and 1-2 µg kg⁻¹ fentanyl. Post-delivery oxytocin and carboprost in conjunction with uterine massage were needed to facilitate uterine contraction. Nitroglycerin is especially potent and has the added benefit of being easily titratable and short-acting (9,10). We used nitroglycerin infusion for uterine relaxation before delivery. Fentanyl 20 µg kg⁻¹ should be administered via i.m. route to provide immediate and postoperative analgesia for the fetus. The fetus should be intubated before the cord is clamped. If fetus cannot be intubated, partial resection of the obstructive lesion or tracheostomy can be performed (7,11). After safety of airway is established, the cord is clamped and the fetus is delivered. We were able to intubate our case on the third attempt before clamping the cord. It is important to reverse the uterine relaxation after delivery of the baby (10). The volatile agent should be reduced after the cord

is clamped, and opioid analgesics and local anesthetics should be administered through the epidural catheter. In the present case, we started i.v. fentanyl infusion because we could not place the epidural catheter. Intubation and tracheostomy set should be kept readily available. Emergency surgical intervention should be performed if necessary. If surgery is not justified, the newborn should be monitored in the neonatal intensive care unit (11).

In conclusion, the *ex utero* intrapartum treatment procedure is used in case of fetal lesions with the potential to cause life-threatening airway obstruction immediately after delivery, such as in CH. Deep anesthesia is often employed. Before or after clamping the cord, the baby is intubated. If the fetus can not be intubated, partial resection of the obstructive lesion or tracheostomy can be performed.

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