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Intrapartum endotracheal intubation in fetuses with airway obstruction. Two first cases of EXIT surgery at Hospital Rebagliati - EsSalud

Intubación endotraqueal intraparto en fetos con obstrucción de la vía aérea. Dos primeros casos de cirugía EXIT en el Hospital Rebagliati - EsSalud

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ABSTRACT

Two cases of pregnant women in the third trimester of gestation referred for management of fetal cervical tumor with airway obstruction are presented. The cases were scheduled for EXIT (*ex-utero intrapartum treatment*), fetal surgery which allowed controlled intrapartum orotracheal intubation avoiding the period of hypoxia and potential asphyxia that would occur with the conventional neonatal procedure. The neonates were subsequently operated on by head and neck surgeons for reduction of the cervical mass. It is suggested to consider this procedure in cases where severe airway obstruction is suspected and to refer them to centers that have a fetal surgery unit with a multidisciplinary medical team.

Key words: Lymphangioma, cervical, Teratoma, cervical, Surgery, fetal, EXIT surgical procedure.

RESUMEN

Se presenta dos casos de gestantes en el tercer trimestre de gestación referidas para manejo de tumor cervical fetal con obstrucción de la vía respiratoria. Los casos fueron programados para cirugía fetal EXIT (*ex-utero intrapartum treatment*) que permitió la intubación orotraqueal intraparto controlada evitando el período de hipoxia y potencial asfixia que ocurrirían con el procedimiento neonatal convencional. Los recién nacidos fueron operados posteriormente por los cirujanos de cirugía de cabeza y cuello para la reducción de la masa cervical. Se sugiere plantear este procedimiento en los casos en que se sospeche obstrucción severa de la vía aérea y referirlos a centros que cuenten con una unidad de cirugía fetal con equipo médico multidisciplinario.

Palabras clave: Linfangioma cervical, Teratoma cervical, Cirugía fetal, Procedimiento quirúrgico EXIT.

INTRODUCTION

Lymphangioma and cervical teratoma are the two most frequent fetal cervical tumors that cause severe airway obstruction and are associated with high neonatal mortality and morbidity⁽¹⁾. The difficulty occurs at birth, as it is impossible to perform endotracheal intubation due to the obstructed airway. It is a real clinical challenge and many cases of neonatal death by asphyxia have been reported due to failure of orotracheal intubation^(2,3).

The EXIT technique (*ex-utero intrapartum treatment*) was designed in the 90's with the purpose of securing the neonatal airway in complicated cases⁽⁴⁾. The surgical and anesthetic technique allows oxygenation through the umbilical cord and placental circulation with the fetus exposed, to allow more time for endotracheal intubation⁽⁵⁾. To be able to perform it, an exact balance of a multidisciplinary team is needed.



CASE REPORTS

CASE 1

A 28-year-old primipara was referred from a private center at 36 weeks of gestation for presenting a fetal cervical tumor and airway obstruction, with significant growth in the last weeks. Ultrasound showed a multicystic cervical tumor measuring 169 x 118 mm, with severe airway obstruction and concomitant polyhydramnios (Figure 1).

Following a medical round with fetal medicine, obstetrics-gynecology, neonatology, head and neck surgery and anesthesiology, the EXIT surgery was scheduled urgently, as the patient went into labor. Anesthesia consisted in general inhalation anesthesia with dexamethasone premedication and intravenous induction with fentanyl, propofol and rocuronium. During the EXIT procedure, a maximum sevoflurane minimum alveolar concentration (MAC) of 2 was achieved to help relax the uterus and achieve adequate uteroplacental circulation. Pfannenstiel incision and Kerr hysterotomy were performed to exteriorize only the upper half of the fetus. Continuous irrigation of the uterine cavity with tempered 0.9% NaCl and clinical monitoring of the fetal heart rate was performed. The cystic tumor involved the entire anterior and lateral compartment of the fetal neck. Neonatology attempted to access the airway by endotracheal intubation, without success due to the large tumor mass, so the head and neck surgery team performed decompression of the tumor by aspiration of the cystic content, after which endotracheal intubation and cord ligation were performed (Figure 2). The total time of the utero-placental bypass was 21 minutes and the fetal heartbeat remained constant throughout the procedure.

FIGURE 1. CASE 1: ULTRASOUND FINDINGS OF THE FETAL CERVICAL TUMOR (MULTILOCLAR CYSTIC).

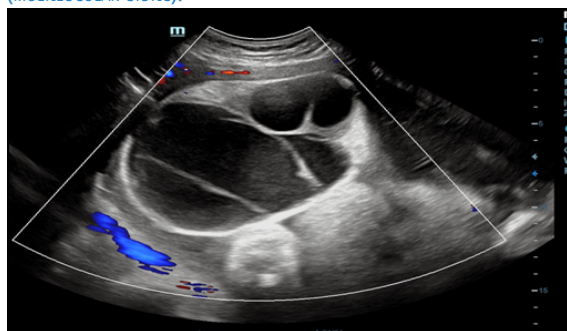


FIGURE 2. CASE 1: NEWBORN INTUBATED DURING THE EXIT PROCEDURE.



After removal of the newborn, a safe airway was considered pertinent and a tracheostomy was performed following placement of an umbilical venous catheter, to provide analgesia with fentanyl and midazolam. The newborn weighted 3710 gram and was transferred to the neonatal ICU.

After six days he underwent excisional surgery by head and neck surgery. The pathology result was a lymphangioma. The evolution was unfortunately torpid, with recurrence of lymph filling and involvement of deep planes with subsequent sepsis and neonatal death.

CASE 2

27-year-old pregnant woman referred from a private center at 37 weeks of gestation for presenting a fetal cervical lump and suspected airway obstruction. Ultrasound showed a cystic/solid cervical tumor measuring 12 x 9 cm with extension to the upper third of the thorax. The ultrasound and MRI findings are shown in Figure 3. The EXIT protocol was initiated by convening the multidisciplinary team according to case 1 described. The presence of an anterior and right lateral cervical tumor was noted. Endotracheal intubation was successful on the third attempt after partial decompression of the mass by aspiration by the head and neck surgery team (Figure 4).

The total time of placental uterine bypass was 20 minutes. The fetal heartbeat, verified by ultrasound, remained constant during the procedure. The male newborn weighed 3260 grams and was transferred to the neonatal intensive



FIGURE 3. CASE 2: ULTRASOUND FINDINGS OF FETAL CERVICAL TUMOR (CYSTIC-SOLID) AND FETAL RESONANCE HIGHLIGHTING DEEP PLANE INVOLVEMENT.



FIGURE 4. CASE 2: INTUBATED NEWBORN DURING EXIT PROCEDURE.



FIGURE 5. CASE 2: NEONATE IN THE POSTOPERATIVE PERIOD FOR CERVICAL TERATOMA.



care unit after the described airway approach. After twenty days he underwent excisional surgery by head and neck surgery, verifying infiltration from the right parotid region and floor of the mouth to the superior mediastinum and involving large vessels and nerve plexuses. The pathology result was a grade 2 immature teratoma, for which he was referred to pediatric onc-hematology and is currently receiving satisfactory chemotherapy management (Figure 5).

DISCUSSION

Fetal cervical tumors represent a rare pathology that can cause severe airway obstruction with fatal consequences for an otherwise healthy newborn. Once diagnosed, it is necessary to determine the extent of the mass and the level of airway compromise by ultrasound in expert

hands and, if feasible, to complement with fetal MRI⁽⁶⁾. Although EXIT has several indications, the main one is when airway obstruction is suspected (Table 1).

The next step is to convene a multidisciplinary team composed of physicians specialized in fetal medicine, anesthesiologists, neonatologists, specialists in fetal airway management (head and neck surgeons, otolaryngologists, or pulmonologists) and nurses specialized in surgical center and neonatology⁽⁵⁻⁷⁾. Planning is fundamental and receiving this case in a tertiary center allowed us to have the necessary human and logistic tools. Mean gestational age considered in systematic reviews for delivery is 35 weeks, due to the high incidence of premature rupture of membranes or preterm delivery that occurs due to secondary polyhydramnios, as occurred in case 1, which limited the MRI study. Emergency surgeries should be avoided, due to the need of adequate implementation of the procedure⁽⁸⁻¹⁰⁾.



TABLE 1. INDICATIONS FOR PLANNING EXIT SURGERY.

Extrinsic airway obstruction	Intrinsic airway obstruction
<ul style="list-style-type: none"> • Cervical tumors: teratoma, lymphangioma, hemangioma, goiter, neuroblastoma • Orofacial tumors: epulis, epignathus • Micrognathia, severe / retrognathia 	<ul style="list-style-type: none"> • CHAOS Congenital upper airway obstruction syndrome • Reversing artificial tracheal occlusion performed in fetuses with diaphragmatic hernia
Cases in which neonatal resuscitation is a challenge	
<ul style="list-style-type: none"> • Fetal pulmonary masses • Pulmonary hypoplasia / agenesis - secondary to congenital diaphragmatic hernia, among others • Mediastinal tumors 	

Once human resources and logistics have been secured, an intervention plan for the management of a difficult airway must be in place and known by each member of the team. Cruz and collaborators mention that such management should be planned from the least to the most invasive procedures, such as the need for tracheostomy, which according to a recent systematic review reaches 20%⁽¹⁰⁾ and was necessary in case 1.

The EXIT procedure is a surgery based on the maintenance of maternal-fetal circulation (utero-placental bypass) during cesarean delivery, which allows the necessary time to achieve a safe airway through procedures ranging from orotracheal intubation to excision of the obstructive lesion itself. In both cases the participation of head and neck surgeons was required for decompression by tumor aspiration, due to the difficulty in accessing the airway.

The anesthetic technique is aimed at avoiding uterine contractions and placental abruption and maintaining fetal respiration. General anesthesia is indispensable. In both cases, for the inhibition of fetal respiration we proceeded to induction with general anesthesia reinforced with fentanyl, propofol and rocuronium. Once the fetal extraction was performed only up to half of the dorsum, keeping the rest of the body in utero, the inhibition of contractions was achieved with an inhalation anesthetic (sevoflurane). Currently, it has been proven that the administration of halogenated anesthetics at analgesic doses of 0.5 to 1 CAM with or without N2O does not have a major impact on the neonate or on uterine contraction⁽²⁾. Regarding fetal cardiac monitoring, ultrasound

and clinical monitoring have been described. In case 1 clinical monitoring was chosen due to the urgency of the case, while for case 2 the registry was under ultrasound guidance. To avoid intrauterine compression of the umbilical cord, the umbilical cord was irrigated with tempered NaCl 0.9/00 by means of a Nélaton probe. During the EXIT procedure, one of the main complications is hysterotomy bleeding, for which it is necessary to perform clamping of the angles. The use of surgical clamps has been described⁽¹¹⁾; in our case, foester clamps were used in the angles.

Regarding access to the airway, in most cases endotracheal intubation is difficult due to tumor obstruction^(12,13). In both cases, it could not be achieved in the first attempt due to the large tumor mass, so head and neck surgery decompressed the tumor by aspiration of its liquid content. Cases of instability of the cannula have been described making it necessary to secure the airway; in case 1, tracheostomy was performed due to a drop in saturation despite intubation.

In the management of a congenital tumor that compromises access to the airway, it is very important to have a multidisciplinary team and the necessary materials to avoid the morbidity and mortality associated with this scenario^(3,6). Although both patients were referred late, the intervention was possible thanks to the optimal and satisfactory organization of the entire surgical team. Referral of these cases to a tertiary center with resolution capacity should be made as soon as the diagnosis is made, in order to implement the case protocol, which includes a complete fetal morphological study, mapping of the extension and invasion of the lesion by ultrasound and magnetic resonance imaging, if necessary, and convene the multidisciplinary team for the performance of a scheduled procedure. We believe it is convenient to add this case to those already reported in the national literature^(2,3,14), as well as to disseminate that this specialized procedure is currently performed by the fetal surgery unit of our hospital in EsSalud. It is necessary to promote and strengthen timely diagnosis and referral to ensure timely and comprehensive care of both the mother and the newborn.



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