Clinical Reports

Anaesthetic management of a neonate with prenatally diagnosed cervical tumour and upper airway obstruction

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We report the anaesthetic management of a mother, and airway management of a neonate with a prenatal diagnosis of cervical cystic hygroma causing upper airway obstruction. The mortality of such neonates due to upper airway obstruction is reported to exceed 20% following deliveries. Elective Caesarean section was performed under general anaesthesia, and in utero tracheal intubation of the neonate was accomplished under uninterrupted maternal-fetal circulation. By utilizing isoflurane and ritodrine, the heart rate of the neonate remained between 120 to 150 bpm for four minutes following uterine incision. We believe that it is important that a multidisciplinary approach be initiated for planning of airway management of the neonate soon after the diagnosis is made. Laryngoscopy blades larger than normal for neonates, and a portable Doppler to monitor the viability of the neonate were found to be useful in the management of the neonate during in utero tracheal intubation. Although estimated blood loss was not increased, nor did uterine atony occur postoperatively despite the use of ritodrine during Caesarean section, the efficacy and safety of ritodrine to delay placental detachment have not been proved.

Key words

AIRWAY: obstruction;

ANAESTHESIA: obstetric, Caesarean section, paediatric, neonate;

COMPLICATIONS: cystic hygroma;

UTERUS: tocolytics, ritodrine.

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Nous décrivons la conduite adoptée chez une mère enceinte et son enfant souffrant d'une obstruction des voies respiratoires supérieures causée par un hygroma kystique cervical diagnostiqué in utero. Le taux de mortalité du nouveau-né présentant ce type d'obstruction dépasse 20% après l'accouchement. Une césarienne sous anesthésie générale a été réalisée et la trachée du nouveau-né intubée in utero sans interruption de la circulation foeto-maternelle. Sous isoflurane et ritodrine, la fréquence cardiaque foetale est demeurée entre 120 et 150 pendant quatre minutes après l'incision utérine. Nous croyons en l'importance d'une approche multidisciplinaire pour planifier le contrôle des voies aériennes néo-natales dès que le diagnostic est posé. Des lames de laryngoscope plus grandes que celles qu'on utilise normalement et un Doppler portable pour monitorer la viabilité du nouveau-né ont été utiles pendant l'intubation endotrachéale in utero. Bien que la perte sanguine n'ait pas été excessive, et qu'il n'y ait pas eu d'atonie utérine malgré l'utilisation de la ritodrine pendant la césarienne, on n'a pas fait la preuve de l'efficacité et de la sécurité de ce médicament pour retarder la séparation placentaire.

Improvements in prenatal imaging technology have enhanced the ability to diagnose congenital anomalies *in utero*. Of these, a cervical tumour causing upper airway obstruction of the fetus is the most urgent medical emergency where immediate airway establishment is required after birth. If the tumour involves the tongue or oropharyngeal space, direct laryngoscopy and subsequent tracheal intubation may be extremely difficult. Also, the cervical location may preclude rapid surgical access to the trachea. Zerella *et al.* reported that 30% of neonates with cervical and 21% with oropharyngeal teratoma died of airway obstruction shortly after birth.¹

Information regarding the "critical interval" during

FIGURE 1 Three-dimensional photograph composed from the helical computed tomography at 37 wk gestation. The cystic hygroma is demonstrated as a simulated tumour (indicated by T) attached to the chin, neck, and anterior thoracic wall of the fetus.

which the placental circulation is maintained after the uterine incision in Caesarean section is limited although it would be invaluable in planning airway strategies for such neonates preoperatively. We report a case of the successful anaesthetic management of a neonate with cervical cystic hygroma, who did not develop fetal distress for at least four minutes after the uterine incision before the cord was clamped.

Case report

A 27-yr-old woman, gravida 2, para 1, presented at 30 wk gestation with a cervical tumour of the fetus which had been diagnosed by ultrasound examination. The course of her pregnancy was unremarkable up to 30 wk.

On admission at 37 wk gestation, her weight was 55 kg and height 150 cm. Physical and laboratory examinations were otherwise normal. The helical computed tomography revealed a large multi-cystic tumour without a bone component compatible with cystic hygroma attached to the chin, bilateral surfaces of the neck, and

FIGURE 2 Ultrasound examination at 37 wk. Note that the tongue protrudes from the mouth, indicating that the tumour occupies the oropharyngeal space. N, nose; L, lips; and T, tongue.

the anterior thoracic wall of the fetus (Figure 1). By the second ultrasound examination, the tongue was noted to protrude from the mouth, suggesting that the tumour occupied the oropharyngeal space (Figure 2). Since polyhydramnios was also documented at this time, obstruction of the upper airway and the gastrointestinal tract was suspected. The placenta was implanted at the posterior wall of the uterus. Estimated fetal weight was 3000 g, and no other fetal abnormalities were found. Due to concern for the upper airway of the neonate, elective Caesarean section was planned, and oral ritodrine was started to prevent premature labour. A multidisciplinary approach was initiated with personnel from obstetrics, neonatology, paediatric surgery, and anaesthesiology, and a difficult airway algorithm was proposed (Figure 3). The tasks of each department were defined preoperatively. The primary role for anaesthesia was three-fold, firstly anaesthetic care of the mother by one anaesthetist; secondly tracheal intubation of the neonate *in utero*, and lastly deciding whether to proceed to tracheostomy following failed intubation by the other scrubbed anaesthetist.

For the Caesarean section at 38 wk gestation, rapid sequence induction of anaesthesia was performed using thiamylal $4 \text{ mg} \cdot \text{kg}^{-1}$ and succinylcholine $1 \text{ mg} \cdot \text{kg}^{-1}$ *iv* with cricoid compression. The patient's trachea was intubated and anaesthesia was maintained with 1% isoflurane in oxygen until delivery.² Intravenous ritodrine at a rate of $400 \mu\text{g} \cdot \text{min}^{-1}$ was started after the induction of anaesthesia until the delivery of the neonate.³

Scrubbed personnel consisted of two obstetricians, two paediatric surgeons, one anaesthetist, and three nurses. Airway equipment and drugs for resuscitation including

FIGURE 3 Difficult airway algorithm used for delivery of the fetus with upper airway obstruction.

epinephrine, atropine and lidocaine in appropriate dilutions were prepared in a sterile manner on a separate Mayo stand. A portable Doppler, a pulse oximeter probe, and ECG electrodes were also sterilized to monitor the fetus. A rigid bronchoscope, a fibrescope, and an emergency tracheostomy set were also prepared.

Seven minutes after induction of anaesthesia, the fetal head was delivered through a low transverse hysterotomy. Suctioning the oral and the nasal cavity failed to provoke respiratory effort or crying. The fetus was further delivered to the upper chest. Then, the portable Doppler was applied over the anterior uterine wall to monitor the fetal heart rate (Figure 4). The first attempt to intubate the trachea failed because the enlarged tongue, due to lymphoedema, prevented the Miller #10 blade from reaching the larynx. Then, using a Seward #1 blade, a 3 mm endotracheal tube was placed. After ascertaining bilateral breath sounds and chest movement, a 2871 g male neonate was delivered and the cord was clamped, four minutes after the uterine incision and 11 min after the induction of anaesthesia. The fetal heart rate remained 120 to 150 bpm during the manipulations. Mechanical ventilation was initiated immediately after the delivery, and

FIGURE 4 The fetus delivered to the shoulders before tracheal intubation. T, tumour; and D, portable Doppler applied over the anterior uterine wall.

arterial blood gas values ten minutes after delivery revealed pHa 7.392, PaO₂ 142.4 mmHg (FiO₂ = 0.3), PaCO₂ 29.9 mmHg, and base excess -5.0 mEq · L⁻¹ without acid-base correction. The Apgar scores were 8 at one minute and 9 at five minutes. The neonate was transported to the intensive care unit for further evaluation of the tumour. Blood glucose values of the neonate during the first 24 hr remained between 50 to 80 mg · dL⁻¹, and there were no clinical signs of hypoglycaemia, such as tremour or seizure. Tracheostomy and partial resection of the cervical portion of the tumour performed two days after Caesarean delivery was uneventful.

The maternal systolic blood pressure and pulse oximeter readings were above 108 mmHg and 98% respectively throughout Caesarean section. Immediately after the ritodrine infusion was started, diastolic blood pressure decreased from 71 to 57 mmHg, and heart rate increased from 88 to 129 bpm, while systolic blood pressure was maintained more than 120 mmHg. Blood loss was estimated to be 800 ml by weighing sponges and the volume

of suction from the surgical field and subtracting the estimated volume of amniotic fluid of 800 ml. Maternal pre- and postoperative haematocrit values were 36.1 and 34.1%, respectively. The anaesthetic and postoperative course of the mother was uneventful.

Discussion

Previous literature reported that *in utero* tracheal intubation or tracheostomy under maternal-fetal circulation could be accomplished successfully.^{1,4,5} Langer *et al.* reported a fetus with a large cervical teratoma diagnosed prenatally by ultrasound examination which was delivered through the hysterotomy to the shoulders and the trachea was intubated. Despite the initial success in the management of the upper airway obstruction, the neonate subsequently died of massive airway bleeding from the tumour.⁴ Catalano *et al.* reported a case in which a fetus with epignathus underwent tracheostomy following Caesarean delivery, but without interrupting the umbilical cord.⁵ However, these reports are lacking in anaesthetic details, uterine incision-to-delivery interval, methods of monitoring the fetus, and plans for airway strategies.

More recently, Schulman *et al.* reported two cases of *in utero* tracheal intubation for cervical tumours. The tracheas of both fetuses were intubated within five minutes of uterine incision, and a pulse oximeter monitored the fetal heart rate in one.⁶ In our case, a sterile portable Doppler applied over the uterine wall was equally effective, and did not interfere with other manipulations in the surgical field. The viability of the fetus was noticed by all the personnel in the operating room. Furthermore, the presence of vernix caseosa, amniotic fluid and blood covering the fetus did not preclude reliable application of the ultrasound in this emergency situation.

The necessity for *in utero* tracheal intubation is clear since Zerella reported 30% and 21% mortality of cervical and oropharyngeal teratoma, respectively, due to upper airway obstruction shortly after birth.¹ Therefore, our anaesthetic management focused on maximizing the "critical interval" from uterine incision to detachment of the placenta, during which the placental circulation and oxygen delivery to the fetus were maintained and thus, manipulation of the airway could be performed safely. Prolongation of uterine incision-to-delivery interval was initially reported to cause progressive neonatal acidosis, lower one-minute Apgar scores and increasing fetal depression.^{7,8} However, Anderson *et al.* studied 204 cases of Caesarean deliveries using stepwise multivariate analysis, and found that uterine-incision to delivery intervals did not correlate with Apgar scores nor umbilical cord blood gases.⁹ Although these clinical investigations gave controversial results, direct measurements of uterine blood flow documented increasing resistance in the utero-

fetal circulation, decreasing uterine blood flow, and thus, oxygen delivery to the fetus due to uterine contractions.^{10,11} To maximize oxygen delivery to the fetus and thereby to prolong the "critical interval," uterine contraction should be inhibited, and placental detachment should be delayed. Firstly, general anaesthesia was chosen to utilize the volatile anaesthetic agent for uterine relaxation which would not have been obtained by regional anaesthesia.¹² General anaesthesia was also preferred because epidural anaesthesia may adversely affect the compensatory response to untreated haemorrhage and worsen maternal hypotension, uterine blood flow, and fetal oxygenation.¹³ Secondly, in contrast to previous reports, more than the maximum recommended dose of β_2 selective agonist was started to delay placental detachment after uterine incision. Alternatively, nitroglycerin could have been employed as a potent relaxant of the uterine smooth muscle. Reportedly, nitroglycerin can be safely used for manual extraction of retained placenta.¹⁴ However, the appropriate rate of infusion for continuous uterine relaxation has not yet been determined. Despite the use of volatile anaesthetic and tocolytic agents, the neonate presented metabolic acidosis immediately after birth. This may be attributed to: firstly hypoxemia may have been present during airway management, and secondly placental circulation may have been mildly altered following uterine-incision, though not severe enough to cause fetal distress.

Although ritodrine is presently used to prevent premature labour,³ its efficacy and safety to delay placental detachment during Caesarean section have not been proved. Fortunately, in our case the uterus responded readily to prophylactic *iv* methergine and intramyometrial prostaglandin $F_{2\alpha}$ as evidenced by normal estimated blood loss. Other oxytocic agents such as oxytocin would have been alternatives to prevent postoperative uterine atony.¹⁵

If oro-tracheal intubation had failed, blind nasal intubation or fiberoptic intubation would have been attempted (Figure 3). However, blind naso-tracheal intubation is difficult without spontaneous respiration. Furthermore, the upper airway anatomy may have been distorted by the tumour occupying the oropharyngeal space. Scheller and Schulman presented a technique of intubating the trachea of a neonate with Pierre-Robin syndrome using a 3.6 mm external diameter fiberoptic bronchoscope.¹⁶ Under direct vision of the vocal cords, they passed a 150 cm soft-tipped wire *via* the suction port into the trachea, removed the bronchoscope over the wire, and then passed a 3.5 mm internal diameter tracheal tube over the wire into the trachea. Using a recently available 2.0 mm external diameter fibroscope, however, may eliminate the step of passing a guide wire and expedite se-

curing of the airway. Our last resort would have been extracorporeal membrane oxygenation (ECMO, Figure 3). According to the use of modified ECMO with persistence of fetal circulation as described by Kelly *et al.*,¹⁷ neonates need to be paralyzed prior to delivery to prevent spontaneous respiration. Umbilical vessels instead of cervical vessels would have been cannulated because of the cervical location of the tumour. However, this low-flow ECMO has not yet been proved effective in humans.

In summary, a neonate with a prenatal diagnosis of cervical cystic hygroma was managed successfully by tracheal intubation *in utero*. A larger than normal laryngoscopy blade for a neonate (Seward #1) and a portable Doppler were indispensable for airway management. By utilizing volatile anaesthesia and *iv* ritodrine, the fetus did not become distressed for four minutes under maternal-fetal circulation. Although intraoperative blood loss was normal and postoperative uterine atony did not occur, the efficacy and safety of ritodrine to delay placental detachment have not been proved.

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