Laryngeal Injury in Neonate and Temporary Extracorporeal Membrane Oxygenation

Nanna Browaldh1*, Yinghua Li2, Lars Falk3, Andreas Ekborn1, Björn Westrup2, and Georgios Papatziamos1
1Department of Otorhinolaryngology, Karolinska University Hospital, Sweden
2Department of Neonatology, Astrid Lindgren’s Children’s Hospital, Karolinska University Hospital, Sweden
3ECMO Unit, Karolinska University Hospital, Sweden

Abstract
Laryngeal or tracheal injury in neonates is a severe and life-threatening condition. Early diagnosis of airway damage is crucial. In the present case, a laryngeal rupture occurred during a difficult vaginal delivery. In this case report, we describe the advantages of ECMO treatment in connection with acute airway surgery and also the need for multidisciplinary management of the troublesome pediatric airway.

ABBREVIATIONS
ECMO: Extracorporeal Membrane Oxygenation; CPR: Cardiopulmonary Resuscitation

INTRODUCTION
Idiopathic laryngeal injury in neonates is rare, but potentially fatal. Difficult delivery by instrumentation increases the risk due to cervical traction; moreover, tracheal lacerations have also been reported after endotracheal intubation. The condition usually presents with respiratory distress associated with subcutaneous emphysema and pneumomediastinum [1]. The injury can be treated conservatively, but some patients require surgical management [2]. This paper describes a case with a life-threatening laryngeal injury in a neonate, managed with surgical repair and treatment with extracorporeal membrane oxygenation (ECMO).

CASE REPORT
A 33-year-old healthy primipara with a normal pregnancy was admitted at 41 wks+0 for delivery. The delivery progressed slowly and a vacuum extraction was performed. Subsequently, there was an imminent shoulder dystocia, which was resolved within one minute. The Apgar score was 3, 7, and 9 at 1, 5, and 10 minutes, respectively. Umbilical blood gases were normal, birth weight 5000 g, birth length 55 cm, and head circumference 38 cm. Due to an absence of spontaneous breathing, the infant was briefly bag-ventilated but recovered promptly with normal respiration at 2 min. In order to further stabilization, a ventilation mask with continued positive airway pressure was used up to 13 minutes of age. At delivery the large head and the swollen face and neck region were already noticed, but were considered to be due to the infant’s apparent large-for-length constitution.

At 30 minutes of age the baby suddenly developed respiratory distress, cyanosis, and bradycardia, and was difficult to ventilate manually. The child received cardiopulmonary resuscitation (CPR) and was intubated at approximately 40 minutes and recovered. Considering the prompt recovery by adequate ventilation in both incidents, the cause was believed to be difficulties in providing clear airways due to the swollen face, and the infant was extubated. However, immediately after extubation the child could not maintain a patent airway. At reintubation, using indirect laryngoscope HEINE, Herrshing, Germany, there seemed to be a resistance just below the vocal cords. The infant stabilized promptly. Since the swelling of the neck had progressed, the infant was transported to the neonatal intensive care level III. On arrival, 3.5 hours after birth, the baby suddenly became bradycardic and cyanotic and did not respond to positive pressure ventilation. Under the suspicion of extubation, reintubation was performed without any difficulties, but with little response in chest movement. An X-ray of the thorax confirmed cervical subcutaneous air and bilateral pneumomediastines, (Figure 1). Even though the large bilateral pneumomediastines were drained, and conventional ventilation was tried, the child did not improve. The blood gas showed a pH of 6.5, pCO2 of 18, and a BE of -28. The saturation was between 50–70% for approximately 40 min during this third event.

Due to the massive air leakage from the airways, ECMO treatment was recommended. FiO2 was maintained at 100% until
At 10 weeks of age, the follow-up at the otolaryngology (ORL) department showed that the patient’s voice was a bit weak and leaking. A flexible nasofiber endoscopy showed scar tissue in the anterior commissure, but with retained abduction and adduction ability of the vocal cords. Subglottic thickening was also seen. The patient was breastfed without difficulties.

At the 10-month follow-up at the ORL department, the infant’s voice had improved. There was no stridor at rest, though still a subglottic airway narrowing that remained at the two-year follow-up.

Currently, the patient is two years old and has been diagnosed with mild cerebral palsy with increased muscle tonus and delayed motor skills. The social abilities are developing quite well.

Full ECMO flow was reached. The patient was cannulated with a veno-arterial approach by direct exposure of the carotic and jugular vessels with a 10 F cannula and a 14 F cannula respectively. ECMO-flow was maintained at 500 ml/min. Anticoagulation was initiated at the time of cannulation with a bolus dose of 50 IU/kg of heparin after which a heparin infusion was started to achieve an Activated Partial Thromboplastin Time of 70 – 80 seconds. At the same time as ECMO was initiated, the otolaryngologist was consulted and a laryngeal injury was confirmed by removing the endotracheal tube and performing a laryngoscopy with an intubation laryngoscope together with a rigid nasal endoscope and a battery light source. A venoarterial approach was chosen due to extreme bradycardia, and the child had also had CPR performed in the minute prior to the start of ECMO. The child was then transferred to the operating room.

During ECMO treatment, the otolaryngologists performed a conventional direct laryngoscopy and rigid bronchoscopy that revealed a laceration in the cricothyroid membrane and also that the right vocal cord was detached from the anterior commissure. Through a transverse collar incision and a laryngofissure, surgical repair was made see Figures (2-4). Under visual monitoring and with a suction catheter as a guide wire, an endotracheal tube was inserted from the nose. Thereafter, the laryngeal cartilage was sutured along the laryngofissure line. The cricothyroid membrane laceration was closed and the collar muscles adapted.

To minimize the risk of air leakage [3], the patient was not ventilated during the next four hours. Hypothermia treatment was induced for 48 hours due to the asphyxia. After five days of ECMO treatment, the patient was decannulated and transferred to the neonatal intensive care unit. The child developed convulsions, but with good response to anticonvulsive treatment, and a renewed electroencephalography indicated recovery. Magnetic resonance imaging at 6 days of age revealed extracerebral bleeding and extracranial and subdural hematomas, as well as minor intracranial bleeding, which was considered to be caused by asphyxia, though complications due to ECMO treatment could not be excluded.

At day 11, the patient underwent a new laryngoscopy and rigid bronchoscopy, which showed mobile vocal cords and normal healing of the anastomosis. Exubation was performed, and the baby was referred to a neonatal care unit.
DISCUSSION

The laryngeal injury in the present case was a very severe condition. The otolaryngologist was consulted late in the process when ECMO was already initiated. ECMO is not the first treatment of choice, but was initiated as the child had a life-threatening uncontrolled condition. It then served as a way to achieve a safe and unrushed repair of the neonate airway.

Tracheal injury in neonates is extremely rare and has an associated mortality of 75% [4]. Kacmarynski et al. reported four cases with spontaneous tracheal and subglottic tears in neonates [5]. There are also case reports that describe the rupture as a complication of endotracheal intubation [4]. Excessive hyperextension of the neck places the most strain on the anterior portion, and may cause overstretched which surpasses the elasticity of the tissues and results in a rupture [6]. The carina is fixed, and a common area of tracheal injury is within 2.5 cm of the carina, which is the least mobile region.

It has been theorized that there must be a weak area predisposed to rupture in the trachea or larynx of these patients, compared with the many deliveries using traction that do not result in airway injury [6,7]. Further, laryngotracheal damage from birth trauma may be aggravated by blind endotracheal intubation [5].

The diagnosis can be confirmed by flexible nasofiberendoscopy or direct laryngoscopy. Conservative treatment is mainly used in small laryngotracheal tears, and a surgical approach is used when the damage is considered to be more extensive [8].

The reason for the tracheal injury in this child is unknown. The first symptoms of respiratory distress usually appear within 30 minutes. Early recognition and treatment is important. As in this case, important signs of laryngotracheal damage are: [1] risk factors for a macrosomic baby, particularly in mothers with small pelvic dimensions, [2] instrumental delivery and shoulder dystocia [3], extended time of traction on the neck and posterior traction on the head that adds bending stress to the axial stress on the anterior portion of the airway [4], complicated intubation with several attempts, and [5] signs of subcutaneous emphysema, pneumothorax, and pneumomediastinum.

In cases with pediatric airway anomalies, the advantage of cooperation between many different specialities is clear [9]. The present case underscores the importance of early consultation with an otorhinolaryngologist for endoscopic investigation. Moreover, the possibility of controlled intubation may limit the damage.

Further, the current case is an example of when ECMO can be used outside the conventional indications. Compared to cardiopulmonary bypass, ECMO requires less anticoagulation therapy and it also avoids sternotomy. The crucial role of ECMO in patients where ventilation is impossible due to the location and severity of the laryngeal-tracheobronchial injury has been described earlier [10,11] Kunisaki et al., describe the advantages of ECMO in neonates, all with tracheal anomalies and where ECMO served as a bridge to lifesaving tracheal reconstruction immediately after birth [12]. Another advantage of ECMO is the possibility of reducing airway pressure and decreasing ventilator pressure [13,14].

There are several causes that explain the need for ECMO in the present case. Firstly, there is the need to save the patient’s life in a critical situation. Secondly, the rupture of the airway can lead to life threatening tension pneumothorax and pneumomediastinum, especially when the patient receives positive pressure ventilation, which can be prevented by ECMO. Thirdly, the need for emergency tracheostomy may be avoided. A tracheostomy might worsen the chances of surgical repair, depending on its placement relative to the location of the injury. Fourthly, after surgical repair it is desirable to have low airway pressures, which may impair the tidal volumes and cause hypoxia. ECMO therapy enables not only reduction of the mean airway pressure, but suspension of ventilation. This will facilitate the tissue-healing process. The disadvantages of ECMO are mainly due to the anticoagulation treatment, which increases the risk of bleeding [15].

The absence of chest movements during ventilation indicates that the tip of the tube was probably dislodged into the ruptured criothyroid membrane. The repeated intubations might have worsened this situation due to the natural tendency the tip has to move anteriorly.

Retrospectively, other treatment options might have been available. However, in this case it was extremely difficult to visualize the entrance to the larynx, and the diagnosis of ruptured airway was made late with an endoscope. If the child had been intubated past the injured area with a cuffed tube, the damaged area would have been relieved and the air leakage would have ceased, likely making the ECMO treatment unnecessary.

CONCLUSION

Pediatric airway injuries require a multidisciplinary approach. In hindsight, an early consultation of an otolaryngologist may have avoided ECMO treatment in this case. Nevertheless, the possibility of ECMO in cases where a secure airway is not achieved can be crucial until a surgical and definitive treatment has been completed.

REFERENCES

1. Newman B, Oh KS. Iatrogenic tracheobronchial perforation in infants.


