



## Case Report

## Tracheal rupture in complicated delivery: A case report and review of the literature

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## ABSTRACT

A case of distal tracheal rupture is described, literature review reveals two previously reported cases of neonatal distal tracheal rupture, as well as 14 cases of anterior subglottic rupture. All patients had shoulder dystocia, and 59% had associated brachial plexus injury. Delayed diagnosis (>3 days) was common in the distal tracheal group (66%), compared to 0% in the anterior subglottic group. The 2 distal tracheal rupture patients were initially managed conservatively, but ultimately required open repair. Distal tracheal rupture is exceedingly rare and more difficult to diagnose and manage than the more common anterior subglottic rupture.

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## 1. Introduction

Rupture of the larynx or the trachea in neonates is a rare but well described condition. It is often attributed to traumatic endotracheal intubation, often in low birthweight, premature infants [1–3]. In most of these cases, there was reported difficulty with the intubation, particularly advancing the endotracheal tube beyond the glottis. There is clearly a second subset of patients who are also at risk for neonatal tracheal rupture, specifically the macrosomic neonates with shoulder dystocia complicating their delivery. In these cases, the trauma of the delivery itself is thought to be the cause of the rupture. These patients also often have other signs of traumatic delivery, usually represented by brachial plexus injury. We describe a case of traumatic distal tracheal rupture, highlighting the difficulties in diagnosis and management of this potentially fatal diagnosis. A literature review reveals 16 previously reported cases including 2 distal ruptures similar to ours. We reviewed the presenting features, management decisions and outcomes in all 17 cases to determine prognostic factors for this rare diagnosis.

## 2. Case report

A 3600 g male was born at 38 weeks gestation following a normal pregnancy. The delivery was complicated by dystocia of the left shoulder, and a vacuum was used to assist in the delivery. The child was also noted to have meconium aspiration, and was placed on Bilevel positive airway pressure (BiPAP) following suctioning of the meconium. The child was then transferred to the NICU where he developed dramatic subcutaneous emphysema (Fig. 1) and respiratory distress, necessitating intubation. Otolaryngology first evaluated him following intubation, and at this time he was requiring the high frequency oscillating ventilator with 100% FiO<sub>2</sub>, and there were active discussions regarding initiation of ECMO if he failed to improve. Urgent rigid bronchoscopy was considered but deferred since his condition stabilized. He steadily improved over the next 3 days with resolved subcutaneous emphysema, and a plan was made to extubate on day of life 4, however he became more difficult to ventilate over the course of the day with increased peak pressures. Fiberoptic bronchoscopy revealed soft tissue at the distal end of the endotracheal tube, so the decision was made to take him to the operating room for a formal airway evaluation.

Bronchoscopy revealed what appeared to be soft tissue collapse at the distal end of the endotracheal tube. We were able to maneuver past this obstruction (Fig. 2) and visualize normal carina. The endotracheal tube was placed distal to the obstruction under direct visualization, and a CT angiogram was ordered for

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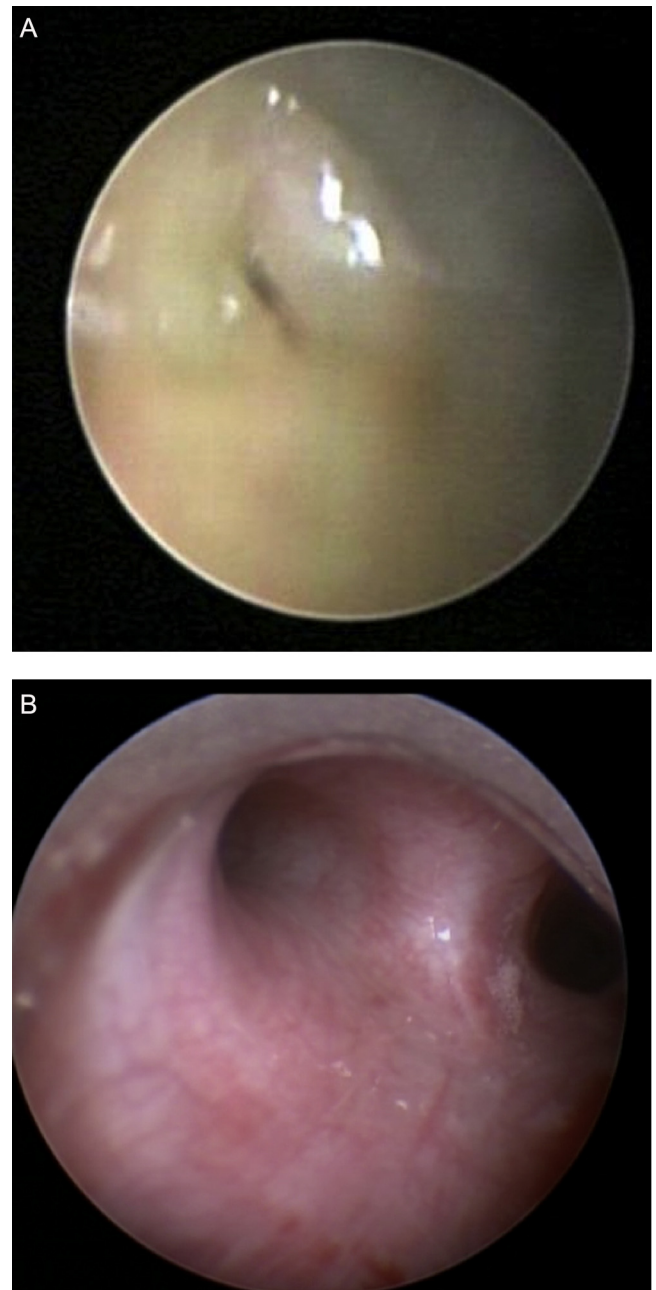
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**Fig. 1.** X-ray showing significant subcutaneous emphysema which developed shortly after birth.

further evaluation of the airway. CT angiogram showed a large distal tracheal rupture (Fig. 3). The patient remained intubated in the PICU, and vancomycin and zosyn were initiated to prevent mediastinitis. In consultation with the pediatric cardiothoracic surgeons, the decision was made to employ a watchful waiting approach initially, with a plan to repair the defect after medical treatment of anticipated acute mediastinal inflammation. However, on day of life (DOL) 9, he became increasingly difficult to ventilate with high peak pressures and air leak around the endotracheal tube, so he was taken to the operating room urgently. Bronchoscopy at this point revealed a near circumferential rupture of the distal trachea as shown in Fig. 4. A sternotomy was performed, and the patient was placed on cardiopulmonary bypass. The tracheal defect could not be primarily reanastomosis because of extensive necrosis and friable tissue adjacent to the rupture. We opted to use a bovine CorMatrix ECM graft to bridge the tracheal defect, and an absorbable miniplate external to the anterior trachea to suspend the repaired anterior wall. He was reintubated with the endotracheal tube bypassing the reconstructed region, 4 chest tubes were placed, and he returned to the PICU where we continued to treat him for mediastinitis.

The patient remained stable, but required pressors, and surveillance bronchoscopy on post-operative day 10 revealed a nicely healing reconstruction which appeared to be intact (Fig. 5).



**Fig. 2.** Bronchoscopy revealed soft tissue collapse in the trachea (A) and a normal carina (B).

Tracheotomy was performed at this time, and a size 3.5 adjustable bivona was placed such that the distal end of the trach sat beyond the repair. On post-tracheotomy day 3, the patient developed an air leak at through the sternotomy incision, and was taken back to the OR for bronchoscopy to evaluate for a tracheal defect. No obvious defect was identified, but the bovine mucosal graft looked less viable. The tracheotomy tube was replaced with an endotracheal tube to minimize friction across the repair. The leak through the presumed tracheocutaneous fistula resolved. He continued to require pressors, and continued to receive vancomycin and zosyn for sepsis. Another bronchoscopy was performed 3 weeks later, and the trachea appeared intact and healthy. Unfortunately his PICU course was complicated by acute kidney injury requiring peritoneal dialysis as well as IJ thrombosis, fungal peritonitis, pleural effusions and sepsis, and he ultimately expired on DOL# 61.

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