Case report

Post-intubation tracheobronchomalacia in a young adult: A rare case report

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A B S T R A C T

Tracheobronchomalacia (TBM) is characterized by weakness of cartilaginous supporting structures of tracheal and bronchial walls, resulting in central airway obstruction. It is a rare condition that can occur after prolonged intubation. Here, we report a 26-year-old, alcoholic male who had TBM, mild subglottic, and severe tracheal stenosis following 2 weeks of intubation.

Subglottic and tracheal stenosis can occur after extubation but TBM is uncommon. The exact mechanism is still unknown but post-intubation TBM is a life-threatening condition with high morbidity and mortality if left untreated. Early detection and timely management can improve the outcome of patients.

Tracheobronchomalacia (TBM) is characterized by weakness of cartilaginous supporting structures of tracheal and bronchial walls, resulting in central airway obstruction [1]. Here, we report a patient of 26-year-old, morbidly obese man presents with worsening upper airway obstruction (UAO) for two weeks. Previously, he was admitted to ICU with metabolic encephalopathy and multiple organ dysfunction syndromes. Initially, he had a brief episode of generalized tonic-clonic seizure due to alcohol withdrawal syndrome, and was intubated with a styletted 8.5 mm cuffed endotracheal tube (ETT) by an anesthesiologist with one attempt. At ICU, his blood culture was positive for Staphylococus Aureus. His hospital course was complicated by severe ARDS (PaO_2/FiO_2 40.1, Oxygen index 32.4), severe alcoholic hepatitis (requiring prolonged course of prednisone), fulminant hepatic failure (Albumin 2.9 g/dL, PT 18.5 secs, and INR 1.66, Hepatic encephalopathy grade 4), and acute kidney injury stage 1 (urine output 870 ml/24 hours, serum creatinine 1.5 mg/dL). He was on pressure-regulated volume controlled with lung protective strategies. His peak pressure was in the range of 23–37 cmH_2O. Then, he was extubated successfully on ICU day 15 without any signs of upper airway obstruction. A 14-day course of antibiotics was completed and he was discharged on day 23 of hospitalization.

Two weeks later, he was re-admitted because of UAO. Biphasic stridor, hoarseness, productive cough and shortness of breath were appreciated. Physical exam showed stable hemodynamics but hypoxemia (SpO_2 86%, on room air) and increase work of breathing. Chest X-ray showed no infiltration. Contrast-enhanced spiral CT of neck reveals mild subglottic wall thickening with patent airway. ENT performed direct laryngoscopy. It showed proximal tracheal stenosis of 6 cm, involving tissue just below cricoid. This stenosis is inclusive of the segment with dynamic airway collapse. Proximal trachea and subglottic area was subsequently dilated up to 8 French of endotracheal tube size. He was discharged with beclomethasone, proton pump inhibitor and ciprofloxacin.

The second bronchoscopy was performed at day 8 after discharge because of persistent shortness of breath. Tracheal stenosis was found with a diameter of 5 mm. Subsequently, narrowed airway was dilated by balloon, cauterized by Argon plasma coagulation and injected with mitomycin C.

The third bronchoscopy was repeated at day 14 after discharge because of recurrent UAO. It showed subglottic stenosis of approximately 1.5 cm below the vocal cords. Trachea was stenotic (90% occlusion) from 1.5 cm below the vocal cords to roughly 6 cm above the carina with near total collapse during expiration. Balloon dilation was performed in the trachea using a 6 cm (length) and 15 mm (diameter) CRE balloon with 50% dilatation of lumen size for management of tracheal stenosis. Cryotherapy was performed in the trachea for destruction of abnormal tissue. Mitomycin C was then injected into the granulation tissue. He was discharged with extended period of steroid taper and amoxicillin/clavulanic acid. The plan is to follow as outpatient for evaluation for possible stent placement and/or tracheostomy.
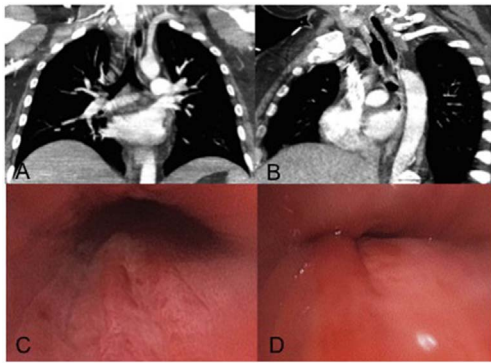
A and B: CT chest showing severe tracheal stenosis.

C and D: Bronchoscopic view of tracheomalacia with total collapse during expiration.

Discussion

TBM in a young man after extubation is very rare. We ruled out secondary cause such as external compression from vascular ring, goiter, and esophageal disorders. Prolonged intubation and gastro-esophageal reflux (GERD) are risk factors in this patient [2]. We proposed a mechanism of prolonged internal compression that can predispose degeneration of normal cartilage. For GERD, studies in the past proposed repeated exposure of tracheal tissue to an acid milieu and digestive enzymes from GERD might alter the matrix protein structure, resulting in the reduced tensile strength of tracheal tissue, leading to TBM [3,4]. Primary TBM is less likely in this case. It is much more common in children which is explained by impaired maturation in cartilaginous rings and/or decreased tone of the trachealis muscle [5]. TBM has been shown to be associated with obesity, asthma, and COPD [6]. However, the exact mechanism of TBM is still poorly understood. Diagnosis can be made with either dynamic flexible bronchoscopy (DFB) or dynamic expiratory CT scan, but DFB remained the gold standard for diagnosis of TBM [6]. Pulmonary Functional Test (PFT) is helpful during the initial workup in case of fixed airway obstruction, with a classic flow-volume loop shape. There has been no universally accepted classification system. Treatment for TBM is dependent on a degree of expiratory central airway collapse as well as response to initial airway stabilization techniques. Severe TBM is treated with stent placement or tracheobronchoplasty. Silicone stent has been used, as a bridge to surgery but there is a high rate of relapse after removal. Furthermore, there is no consensus on the indication for placement as well as optimal timing for removal. Non-invasive positive pressure ventilation (NIPPV) can also be used as adjuvant therapy. NIPPV results in increase in intra-luminal pressure, lung volume and rigidity of airway, preventing UAO during expiration.

Post-intubation tracheal stenosis (PITS) is a well-established complication of intubation. It commonly occurs around cuff site due to disruption of local blood supply, with fibrotic replacement of ischemic tissue [7]. The invention of high-volume, low-pressure endotracheal cuff has dramatically resulted in reduction of PITS incidence [7]. In this case, the patient's predisposing factors were prolonged intubation, obstructive sleep apnea, and severe hypoxemic respiratory failure. Bronchoscopic evaluation remains the diagnostic modality of choice as it allows determination of nature, length and severity of PITS [8]. CT scan can be utilized to locate lesion but it tends to overestimate severity of this condition [8]. Pulmonary function test is neither helpful in

establishing diagnosis nor provide risk stratification. Treatment of PITS is complex and requires multidisciplinary collaboration between thoracic surgeons, interventional pulmonologists, and otolaryngologists. Surgical resection of trachea with end-to-end anastomosis is usually recommended for a patient without significant co-morbidity. The advancement of non-invasive intervention through endoscopy has expanded treatment options for PITS. Commonly performed procedures extend from balloon dilatation, electrocautery, cryotherapy, mitomycin C application, to stent placement [7]. Balloon dilatation requires multiple attempts to obtain satisfactory result and study has shown high relapse rate [9]. Likewise, stent placement also has high relapse rate after removal. In comparison, cryotherapy has been demonstrated to be an effective and safe treatment method with lower relapse rate [13].

Novel therapies, including bilateral bronchoplasty, bio-resorbable three-dimensional airway stents, and tracheal cartilage regeneration, have been reported but the efficacy and safety have not been well studied [10–12]. Regardless of treatment options, patients with post-intubation tracheal stenosis and tracheobronchomalacia will require long-term follow-up and outcome after interventions.

Conclusion

Prolong intubation tracheobronchomalacia is a rare and life-threatening condition with high morbidity and mortality. Early detection and timely management can improve the outcome of patients.

Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.rmcr.2017.12.007.

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