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CASE REPORTS / CASE SERIES

Congenital adult tracheoesophageal fistula repair with transthoracic ventilation: a case report

Réparation d'une fistule trachéo-oesophagienne congénitale chez l'adulte avec ventilation transthoracique : une présentation de cas

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Abstract

Purpose *To describe our experience using transthoracic ventilation to facilitate oral endotracheal tube (ETT) exchange after accidental ETT cuff rupture during a case of congenital tracheoesophageal fistula (TEF) repair.*

Clinical features *A 53-yr-old male underwent a congenital H-type TEF repair via right-sided thoracotomy with a single-lumen ETT and a bronchial blocker. A large air leak developed after ETT cuff rupture during fistula closure. Transthoracic intubation via tracheotomy was performed to continue ventilation during an oral ETT exchange in the lateral position. No hypoxia or hemodynamic compromise occurred.*

Conclusions *Airway device choice for TEF repair must be carefully considered in conjunction with the surgical team. In the present case of accidental ETT cuff rupture, rescue transthoracic ventilation safely facilitated oral ETT exchange.*

Résumé

Objectif *Décrire notre expérience avec la ventilation transthoracique pour faciliter l'échange de sonde endotrachéale (SET) orale après la rupture accidentelle du ballonnet de la SET lors d'un cas de réparation d'une fistule trachéo-oesophagienne (FTO) congénitale.*

Caractéristiques cliniques *Un homme de 53 ans a bénéficié d'une réparation de FTO congénitale de type H via une thoracotomie du côté droit avec une SET à simple lumière et un bloqueur bronchique. Une importante fuite d'air est apparue après la rupture du ballonnet de la SET lors de la fermeture de la fistule. Une intubation transthoracique par trachéotomie a été réalisée pour poursuivre la ventilation pendant un échange de SET orale en position latérale. Aucune hypoxie ou trouble hémodynamique ne s'est produit.*

Conclusion *Le choix du dispositif pour voies aériennes pour une réparation de FTO doit être soigneusement examiné en collaboration avec l'équipe chirurgicale. Dans le cas présent d'une rupture accidentelle du ballonnet de la SET, la ventilation transthoracique de secours a facilité un échange de SET orale en toute sécurité.*

Keywords Adult · congenital tracheoesophageal fistula · intraoperative airway management · transthoracic ventilation

Tracheoesophageal fistula (TEF) is a rare congenital malformation typically associated with esophageal atresia, and is usually diagnosed in infancy. H-type TEFs, representing 4–5% of cases, exist without esophageal atresia, and may remain undiscovered until adulthood. H-type TEFs can result in chronic aspiration and recurrent pulmonary infections.[1,2] Important considerations for TEF repair are strategies for lung isolation and aspiration prevention. We present the case of a 53-yr-old male with a congenital H-type TEF repaired via right-sided thoracotomy, complicated by endotracheal balloon puncture during repair.

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Case description

Consent was obtained from the patient for publication of this case report. The patient presented with a history of chronic cough and recurrent pneumonias. His past medical history was significant for reflux and 60 pack-year of cigarette smoking. He had had no prior thoracic surgeries. An H-type TEF was incidentally found on a computed tomography (CT) scan (Fig. 1A) and confirmed via bronchoscopy to be 5 cm above the carina (Fig. 1B). The CT showed early evidence of chronic lung damage. Preoperative pulmonary function testing (PFT) revealed moderate chronic obstructive pulmonary disease (COPD) (ratio of forced expiratory volume in one second [FEV_1] over forced vital capacity, 65%; FEV_1, 73% predicted) and a moderately reduced diffusion capacity of the lung for carbon monoxide (59% predicted). After considering the patient's age and comorbidities in the context of lung parenchymal changes related to chronic gastric soiling, the decision was made to proceed with repair.

In the operating room, the patient's preinduction room air peripheral oxygen saturation (SpO_2) was 94%. An arterial line was placed, and general anesthesia was induced with a rapid sequence induction using propofol, sufentanil, and rocuronium. Despite preoxygenation, the SpO_2 reached 90% during apnea, though this recovered to 100% after ventilation. Using a size four Macintosh blade, a Cormack–Lehane grade 1 view was obtained and the patient was intubated with an 8.5-mm single-lumen endotracheal tube (ETT). A bronchial blocker was positioned in the right mainstem bronchus under bronchoscopic guidance.

Anesthesia was maintained with sevoflurane, hydromorphone, and rocuronium. A right internal jugular central line was placed. The patient was positioned in left lateral decubitus and one-lung ventilation (OLV) was initiated. Oxygen saturation was maintained above 95% during OLV. A low-dose phenylephrine infusion (10–20 $\mu g \cdot min^{-1}$) maintained a mean arterial pressure above 65 mm Hg.

A right posterolateral thoracotomy was made at the level of the fourth intercostal space, sparing the serratus. The esophagus, which was dilated, was dissected from the esophageal bed, and encircled with Penrose drains inferiorly and superiorly to the fistula. The fistula itself was encircled with a Vesseloop (Fig. 2) and divided, leaving a cuff on the trachea. The esophagotomy was repaired in two layers.

To repair the trachea, residual esophageal mucosa was resected from the membranous airway. The ETT cuff was visualized through the fistula and displaced with forceps before placing sutures to reapproximate the trachea. Despite this, the cuff was punctured, and a large air leak developed. Though hemodynamically stable, the SpO_2 dropped to 91% after cuff rupture, and required an increase to 100% O_2 and packing around the defect with wet gauze to achieve a SpO_2 above 97%. The large air leak and inability to safely provide OLV necessitated ETT exchange. To safely accomplish this in the lateral position, a temporary transthoracic intubation was performed.

A sterile 11-French airway exchange catheter (AEC) was inserted through the tracheal defect, over which a sterile armoured 6-mm ETT was railroaded (Fig. 3A). This took approximately 12 min. A sterile ventilator circuit was connected to the transthoracic ETT (Fig. 3B), capnography was confirmed, and 100% SpO_2 was achieved. While ventilation was maintained transthoracically, the punctured oral ETT was then replaced with a 7.5-mm oral endobronchial ETT using a videolaryngoscope and AEC, placed distal to the fistula. A ventilator circuit was connected to the oral ETT, capnography was confirmed, and the transthoracic ETT removed. During the brief apneic period used to connect the circuit to the oral ETT and confirm capnography, the SpO_2 did drop to 88%. This recovered to 100% with recruitment. The oral endobronchial ETT was then directed into the left mainstem bronchus via bronchoscopy, and repair of the tracheal defect completed.

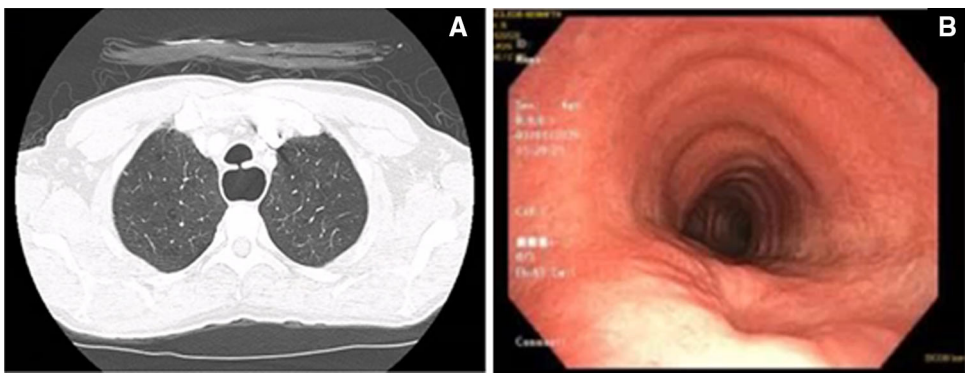

Fig. 1 Preoperative investigations showing the tracheoesophageal fistula. (A) Computed tomography. (B) Bronchoscopy

Leak tests were performed by instilling normal saline into the right hemithorax while ventilating the trachea and inflating the esophagus with esophagogastroduodenoscopy. Both repairs were airtight. Under endoscopic guidance, nasojejunal and nasogastric tubes were placed for feeds and gastric decompression, respectively. To occupy space and provide additional blood supply to the surgical repairs, a right serratus muscle flap was mobilized, delivered through the second intercostal space, and secured between the esophagus and trachea.

A paravertebral catheter was placed percutaneously by the surgeon under direct vision using a 17G Tuohy needle. Two 20-French chest tubes were placed in the right chest, and a Jackson–Pratt drain was placed in the bed of the serratus muscle. The ribs and latissimus were reapproximated and the chest closed. A planned percutaneous tracheostomy was performed because need for prolonged ventilator wean and pulmonary toilet was anticipated preoperatively in the setting of his pre-existing chronic lung injury.

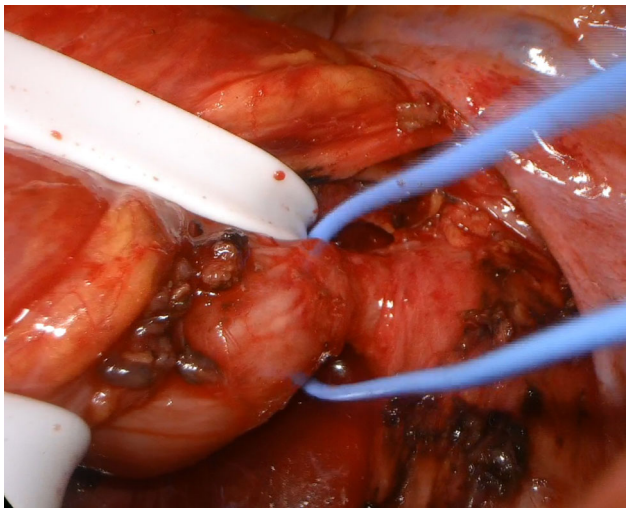

Fig. 2 Isolated tracheoesophageal fistula, encircled by the Vesseloop. The esophagus is encircled by Penrose drains superiorly and inferiorly to the fistula

Postoperatively, the patient did reasonably well. He was weaned from the ventilator on postoperative day (POD) 4 and declassed from the intensive care unit on POD 5. Bronchoscopy was performed daily for the first four days for pulmonary toileting. An increasing white blood cell count on POD 10 raised concern for a leak; however, bronchoscopy confirmed that there was no disruption of the repair. He developed pleural collections and was managed with broad-spectrum antibiotics and pleural drains. He was discharged home on POD 19.

After discharge, the patient complained of dysphagia. Motility studies showed almost complete absence of esophageal peristaltic activity. He was able to manage his dysphagia with dietary changes. Interestingly, he reported a cold sensation in his right chest after consuming cold liquids. He returned to work as a labourer six months postoperatively.

Comment

While rare, congenital H-type TEF diagnosed in adulthood has a typical presentation of chronic cough and recurrent pneumonia secondary to ongoing aspiration.[1,2] Additionally, TEF is associated with abnormal esophageal myenteric plexus development, resulting in disordered peristalsis and impaired function of the lower esophageal sphincter.[3] Our patient exhibited these clinical characteristics and was found to have a large and dilated proximal esophagus intraoperatively. Postoperative high-resolution manometry showed lack of peristaltic activity in his esophagus.

Repair is required to prevent persistent contamination of the airways with gastric contents, which can progress to fibrotic lung parenchyma and recurrent pneumonia. Unfortunately, among adult patients, parenchyma damage is usually present and will not be reversed by fistula repair. Postoperative complications such as multidrug-resistant pneumonia and prolonged ventilator dependency must be anticipated and included in the consent. Tracheostomy

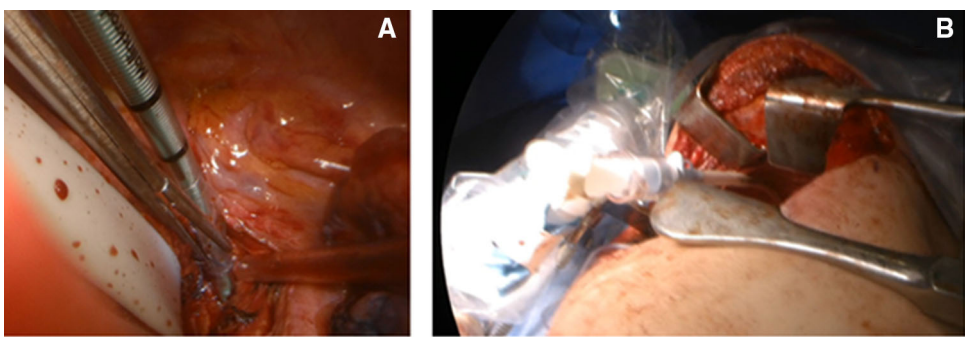

Fig. 3 (A) Insertion of endotracheal tube into the tracheal defect over an airway exchange catheter. (B) Transthoracic ventilation set-up with a sterile ventilator circuit connected to the endotracheal tube

placed during the surgery and postoperative bronchoscopy were planned to assist in clearing secretions.

The decision to approach the repair with a right thoracotomy was based on fistula location on preoperative imaging. This approach also permitted the use of a serratus flap to protect the repair.

Anesthetic and airway management for TEFs can be challenging. Patients may tolerate apnea poorly or be difficult to oxygenate and ventilate secondary to parenchymal lung damage. Without lung isolation distal to the fistula, they may not tolerate positive pressure ventilation as it may result in poor ventilation, abdominal distension, and increased aspiration risk because of gastric insufflation via the fistula. The goals of airway management are to quickly secure a cuffed ETT distal to the fistula and to provide lung isolation for a clear surgical field. Different techniques have been described, including double-lumen ETTs; longer endobronchial single-lumen ETTs; double-cuff single-lumen ETTs, which have a tracheal cuff and a bronchial cuff that can be selectively deflated; or tracheal single-lumen ETTs with bronchial blockers.[4–6]

In addition to considerations of airway device choice, some authors advocate deflation of the ETT cuff during surgical repair of the tracheal defect to reduce risk of cuff rupture.[6] Despite visualizing and displacing the ETT cuff during repair, a rupture still occurred. Therefore, we also advocate ETT cuff deflation during repair.

In our case, several factors influenced our decision to use a single-lumen ETT with a bronchial blocker. Following discussion with the surgical team, it was determined the large outer diameter of a double-lumen ETT would interfere with the repair. The planned percutaneous tracheostomy required a large single-lumen ETT to facilitate the adult size bronchoscope. Also, the patient's PFTs and CT showed COPD and parenchymal damage, so it was felt the larger diameter ETT may help reduce peak airway pressures for lung-protective ventilation and that the bronchial blocker would easily permit intermittent two-lung ventilation if required. At our institution, the largest single-lumen endobronchial tube is 7.5 mm, which was felt to be too small, so by using a larger single-lumen ETT we avoided a tube exchange prior to the tracheostomy. We also felt that the location of the TEF, 5 cm above the carina, was favourable for placement of a tracheal ETT cuff distal to the fistula.

Retrospectively, we feel that placing the largest available single-lumen endobronchial ETT would have been the optimal airway management for this procedure. Using fibreoptic guidance, it can be placed in the trachea or the bronchus depending on current requirements (e.g., intermittent two-lung ventilation), and had the tracheostomy required a larger ETT, this could have been exchanged after the TEF repair.

In the present case, ETT cuff rupture resulted in significant air leak requiring ETT exchange to preserve lung isolation and maintain ventilation. Patient positioning and poor apnea tolerance made this technically challenging. Transthoracic intubation via tracheotomy enabled effective ventilation during the oral ETT exchange. To our knowledge, transthoracic intubation has been reported for blunt trauma thorocotomy,[7] and for complex cancer resections.[8] Nevertheless, this is the first report of elective performance for ETT exchange.

Author contributions *Olga Bednarek*, *Brendan Morgan*, and *Daniel French* contributed to all aspects of this manuscript; acquisition, analysis, and interpretation of data; and drafting the article. *Andrei Khorovets* contributed to the acquisition, analysis, and interpretation of data. *Madelaine Plourde* contributed to the acquisition of data. All authors reviewed and approved the final manuscript.

Disclosures None.

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