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Complex airway reconstruction in children with tracheobronchial injuries: a case series

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Abstract

Paediatric airway surgery in the setting of complex tracheobronchial defects is challenging. This report describes the surgical management and outcomes of pericardial flap repair in three children. The first patient was a 4-month-old boy with a history of tracheoesophageal fistula repair who presented after out-of-hospital cardiac arrest. He was treated by re-do tracheobronchial reconstruction of the carina using a pedicled pericardial flap. The second patient was an 11-month-old boy who presented following aspiration of a button battery. Bronchoscopy showed erosion of the battery through both main bronchi and the oesophagus. The patient underwent emergency reconstruction of the extensive tracheobronchial defect with pedicled right and left pericardial patches. The third patient was a 5-year-old girl who fell from a swing, resulting in avulsion of the right main bronchus. Pedicled pericardium was used to reconstruct the damaged posterior tracheal wall and the right and left main bronchi. All three patients underwent successful repair of complex tracheobronchial defects with good outcomes in terms of survival and quality of life during 6 to 21 months of follow-up. Pedicled pericardial flap repair may be a viable option for achieving improved results in children with severe tracheobronchial defects.

Keywords

Tracheobronchial defect, tracheobronchial injury, pedicled pericardial flap, pericardial flap repair, case report, paediatric surgery, paediatric thoracic surgery

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Introduction

Tracheobronchial injuries in children arise from heterogenous causes and have varied presentations necessitating individualised surgical approaches. Common mechanisms of such injuries include blunt or penetrating chest trauma, iatrogenic injuries during endotracheal intubation or endobronchial dilatation, complications of congenital tracheoesophageal fistula repair, and foreign body-induced erosive trauma.[1–7] Prompt diagnosis is aided by a high index of suspicion, especially in patients presenting with stridor, respiratory distress, haemoptysis, subcutaneous emphysema, pneumothorax, pneumomediastinum, or a persistent air leak associated with failure of lung re-expansion following chest tube insertion.[2–4,8] Computed tomography of the chest offers the tracheal team a detailed view of the affected anatomy and associated injuries.[4,9] Bronchoscopy remains the gold standard diagnostic modality, although this may not be feasible in patients with severe tracheobronchial injury.[2–4,6,9] Rigid bronchoscopy and flexible bronchoscopy are used to confirm the diagnosis, determine the extent of the injury, and define its anatomy. One surgical option for repair is primary repair by end-to-end anastomosis in patients with complete tracheal or bronchial transection.[4,6,7,10,11] This may involve passing an endotracheal tube across the injured tracheal segment and completing the repair over the tube. Other authors have also described the use of extrathoracic muscle flaps to bridge defects or reinforce tracheobronchial repairs.[4,12] The increased complexity of management in paediatric patients with extensive devitalised tissue or large defects secondary to these heterogenous injuries may necessitate the use of autologous pedicled pericardial repair under cardiopulmonary bypass.[13–15] In the present report, we describe our experience with this approach for several different clinical presentations among children recently treated at the Great Ormond Street Hospital for Children, London, UK.

Patients and methods

This short case series describes three patients who presented with complex tracheobronchial injuries. The parents/guardians of all three patients consented to treatment, and the patients' details are de-identified in this paper. The institutional ethics committee waived the need for consent for publication based on the nature of the report (case review) and its retrospective design. The reporting of this study conforms to the CARE guidelines.[16]

Case presentation 1

A 4-month-old boy developed out-of-hospital cardiac arrest following a choking episode and required resuscitation by his parents 22 months previously. The child had a history of antenatal diagnosis of tracheoesophageal fistula and oesophageal atresia, which were repaired on day 2 of life at another tertiary hospital. Upon arrival at the referral hospital, the child was apnoeic and required bagging and cardiopulmonary resuscitation. Microlaryngoscopy, bronchoscopy, and a bronchogram showed significant distal tracheal stenosis (Figure 1). Non-contrast computed tomography revealed a tract arising from the posterior trachea, indicating a persistent fistula. The child was then transferred to us for ongoing management.

The initial surgery was performed via midline sternotomy under cardiopulmonary bypass (CPB) with a run time of 77 minutes and the patient cooled to 32°C. Intraoperative findings included severe stenosis of the distal trachea (2-mm lumen) 0.5 cm proximal to the carina and extending to the origin of the right main bronchus.

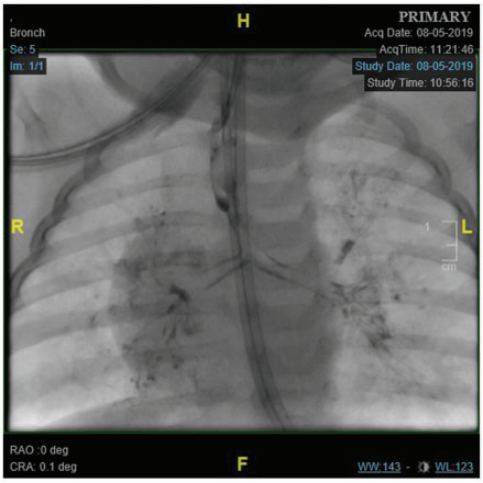

Figure 1. Preoperative image showing the distal tracheal stenosed segment with minimal extension into the right main bronchus.

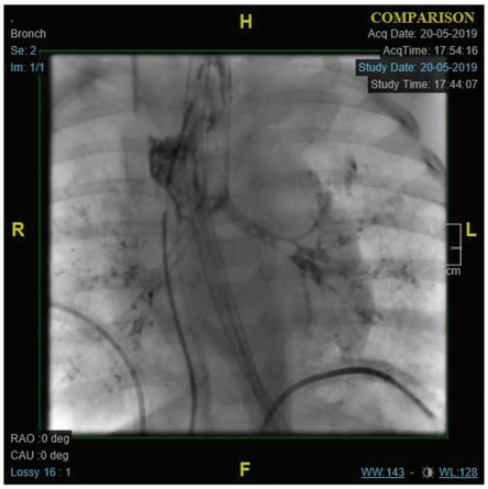

Figure 2. Bronchogram bronchoscopy image showing evidence of initial direct anastomosis repair dehiscence with spillage of contrast into the mediastinum.

Failed formation of the right upper lobe bronchus was also observed. Both the left and right main bronchi were patent with malacia of the right lobar bronchus. The oesophagus was intact, and a patent ductus arteriosus was present, which was ligated.

The trachea and both mainstem bronchi were mobilised. The trachea was incised immediately proximal to the stenosed region, and the stenosed segment of the distal trachea was resected. The anterior wall of the trachea was laid open, and the incision was extended across the carina and into the right main bronchus. Direct anastomosis of the tracheal segment was performing using running 5-0 polydioxanone for the posterior wall and interrupted 5-0 polydioxanone for the anterior wall. The endotracheal tube was repositioned under direct vision. Fibrin sealant and a fibrillar scaffold were placed around the tracheal suture line.

The patient was initially stable during intensive care but later developed respiratory deterioration. He was cannulated for veno-venous extracorporeal membrane oxygenation (ECMO) 9 days following tracheal repair. Bronchography confirmed dehiscence of the tracheal repair and mediastinitis (Figure 2). A re-sternotomy was performed, and the findings included dehiscence of the tracheal anastomosis at the carina and right main bronchus. Tracheobronchial repair was performed with reconstruction of the carina using a pedicled pericardial flap under CPB with a run time of 51 minutes. The right main bronchus was also stented (Figure 3). A plication suture was placed on top of the patch and fixed upward to the sternum.

Postoperative ventilation was difficult, and the child was therefore supported by central veno-arterial ECMO. This was converted back to veno-venous ECMO 5 days later, and the child then achieved delayed sternal closure 15 days following re-do tracheal repair. He was weaned off ECMO and decannulated 20 days after the second surgery. He was maintained on prolonged ventilation and eventually weaned off 2 months postoperatively onto nasal continuous positive airway pressure.

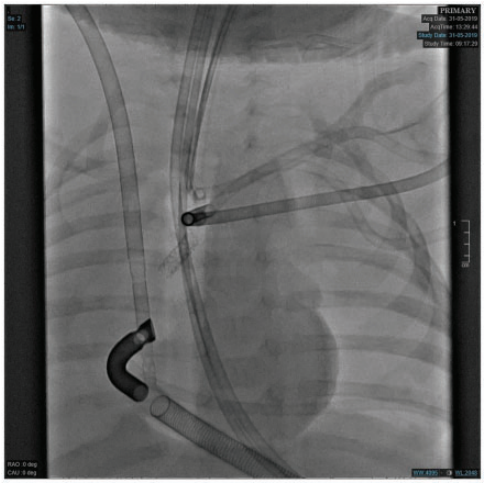

Figure 3. Bronchoscopy bronchogram image after pedicled pericardial repair showing stent insertion in the right main bronchus.

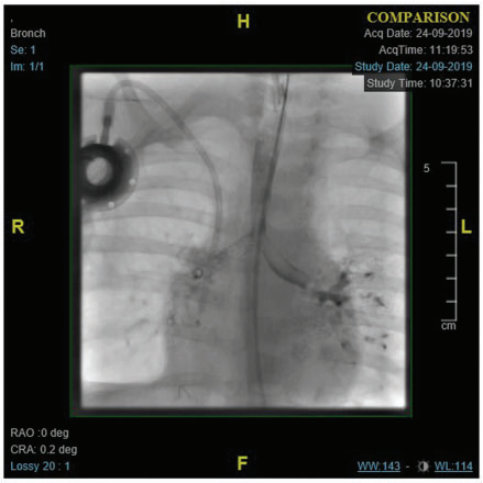

Figure 4. Bronchoscopy bronchogram image showing patent and well-developed tracheobronchial tree 3 months following discharge.

Bronchoscopy and bronchography prior to discharge showed an intact repair site with laryngomalacia and dilation to 6 mm.

The patient underwent three sessions of follow-up bronchoscopy and bronchography at the Tracheal Clinic of Great Ormond Street Hospital with good clinical progress (Figure 4). The most recent bronchoscopy and bronchography was performed 8 months postoperatively and showed a mild in-stent stenosis, which was post-dilated to 5 mm. The child remained very well clinically.

Case presentation 2

An 11-month-old boy presented with suspicion of foreign body ingestion or aspiration 1 week prior to admission. He was noticed to be unwell with difficulty breathing and fever, which worsened after 3 days. He also exhibited drooling of saliva and decreased oral intake. On examination, the child showed mild respiratory distress with biphasic noisy breathing and crepitations.

A chest radiograph showed a button battery in the mediastinum. Thoracic CT showed a dilated oesophagus, mediastinal collection, and collapse/consolidation changes in the right lung apex with damage to the carina and posterior wall destruction of both the proximal right main bronchus and left main bronchus (Figures 5–7). Bronchoscopy at the referral hospital showed that a button battery had eroded through both main bronchi and the oesophagus. There was extensive injury to the trachea, extending to the left main bronchus and the oesophagus.

The patient underwent emergency repair of the tracheo-bronchio-oesophageal fistula and removal of the impacted button battery from the oesophagus. The operation was done under beating-heart CPB with a run time of 213 minutes. Intraoperative findings included right pneumothorax upon opening the sternum, multiple paratracheal nodes, and a button battery impacted in the oesophagus. There was a large tracheoesophageal fistula at the distal trachea involving the carina and extending to the left main bronchus and proximal right main bronchus.

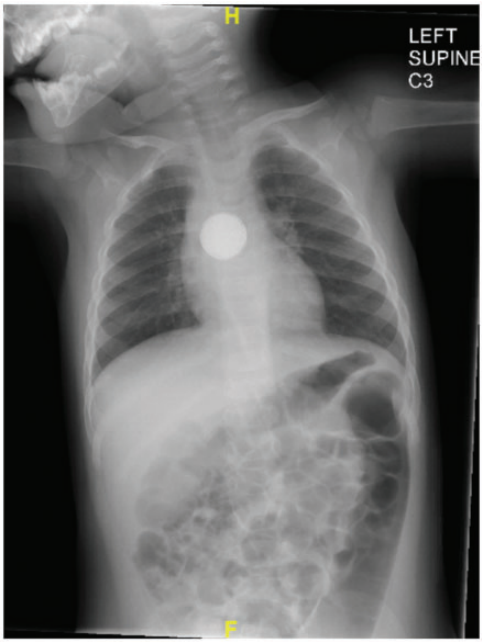

Figure 5. Chest radiograph showing a button battery in the mediastinum.

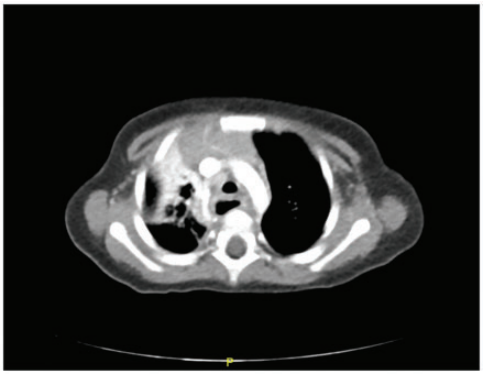

Figure 6. Chest computed tomography showing the upper trachea and dilated oesophagus with mediastinal collection and collapse/consolidation changes in the right lung apex.

The posterior wall of the distal trachea, carina, entire right main bronchus, and two-thirds of the left main bronchus were reconstructed with pedicled right and left pericardial patches. On completion of the

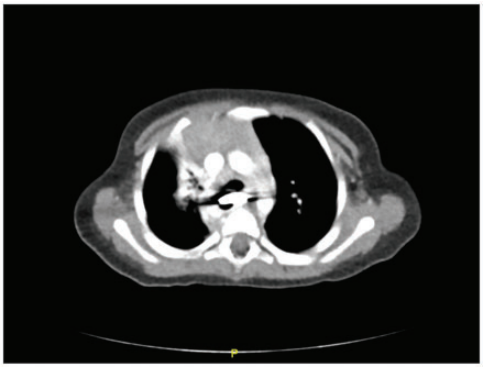

Figure 7. Chest computed tomography showing the button battery in profile with damage to the carina and posterior wall destruction of both the proximal right main bronchus and left main bronchus.

reconstruction, the right main bronchus and left main bronchus allowed a size 4 Hegar dilator. Our general surgical team performed direct closure of the oesophageal injury with trans-anastomotic tube insertion.

The patient was paralysed for 5 days in accordance with the general surgery oesophageal reconstruction protocol. Betadine mediastinal irrigation was instituted. Repeat bronchoscopy and bronchography showed that the central airway was intact with no leak. However, oesophagography confirmed oesophageal repair breakdown, and the patient underwent emergency cervical oesophagostomy and gastrostomy.

The patient was discharged to the referring hospital without respiratory support on postoperative day 17, tolerating gastrostomy feeding. On follow-up bronchoscopy 2 months postoperatively, tracheal granulation was noted over the posterior wall; this was removed endoscopically, and additional systemic steroid therapy was administered.

Case presentation 3

A 5-year-old previously well girl was playing on a swing that tipped over, and the girl

fell onto her chest. She was initially transferred to a local hospital cyanotic and hypoxic with a Glasgow Coma Scale score of 7. She was intubated and ventilated and was noted to have bilateral pneumothorax, for which chest drains were inserted. Thoracic and abdominal CT showed features suggestive of avulsion of the right main bronchus with bilateral lung contusion and extensive emphysema throughout the anterior chest wall. No evidence of vascular injury was found.

The patient was then transferred to a Level I trauma centre, where she was found to have a persistent air leak at the site of the right chest drain; the drain was therefore replaced. The chest CT was again reviewed, and the child was thought to have right main bronchus avulsion, severe soft tissue contusions, fracture of the first left posterior rib, extensive subcutaneous emphysema, and pneumomediastinum. She was transferred to our hospital for further management including ECMO back-up in view of the continued air leak at the right chest drain and increasingly difficult ventilation.

The patient was admitted to our cardiac intensive care unit paralysed, intubated, and ventilated. She was haemodynamically stable with a pH of 7.12, PCO_2 of 12.5, PO_2 of 12, and FiO_2 of 100%. However, she had a persistent massive air leak bilaterally. Bedside echocardiography revealed good biventricular function. Further chest CT was performed within 12 hours of admission to re-assess her injury and implement surgical planning. The CT showed right bronchial avulsion, tracheal disruption, pneumomediastinum, and a dropped right lung (Figure 8).

Emergency repair was performed under beating-heart CPB with a run time of 174 minutes. Drastic respiratory decompensation occurred following anaesthetic induction with severe air leaks necessitating urgent institution of CPB. Intraoperative findings included complete avulsion of the right main bronchus and carinal tracheal disruption with rupture of the posterior tracheal membrane up to the cervical portion. A thin fibrous attachment was present between the trachea and the left main bronchus. No obvious external oesophageal injury was noted.

Figure 8. Chest computed tomography showing bilateral neck subcutaneous emphysema, right bronchial avulsion, tracheal disruption, pneumomediastinum, and a dropped right lung.

Pedicled pericardium was used to reconstruct the damaged posterior tracheal wall. The right and left main bronchi were re-anastomosed side by side to create a neo-carina, and the posterior wall of the neo-carina was anastomosed to the pericardial patch to complete the posterior wall of the trachea. The anterior wall of the neo-carina was anastomosed to the anterior wall of the distal trachea with interrupted polydioxanone sutures, thus completing the tracheoesophageal reconstruction (Figure 9).

Intraoperative bronchoscopy showed good repair and patent bronchial openings. The patient was transferred to the cardiac intensive care unit intubated and paralysed

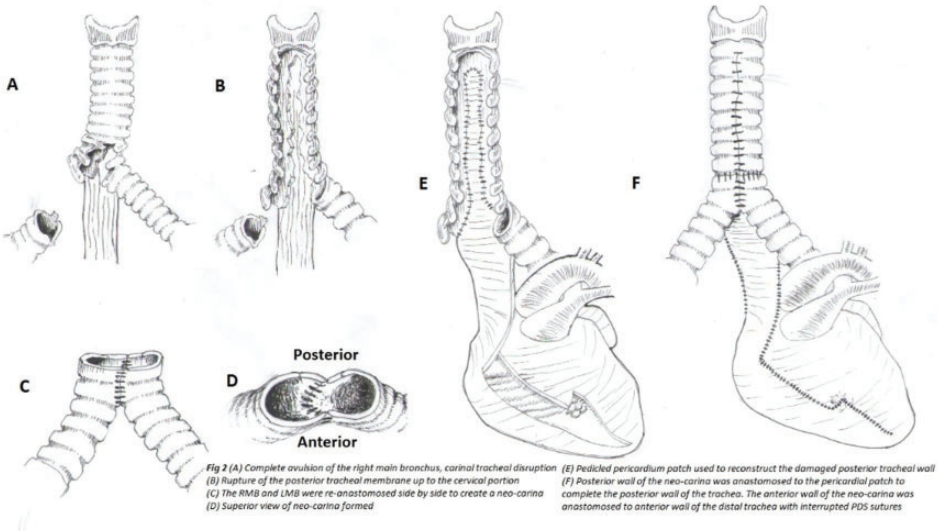

Figure 9. Detailed graphical illustration of pedicled pericardial repair of the complex tracheobronchial defect in the third patient.

in accordance with the tracheal protocol with mediastinal betadine irrigation. Muscle relaxation and intubation were continued for 72 hours, and pre-extubation bronchoscopy and bronchography on postoperative day 5 showed good repair with no evidence of leakage. She was extubated on postoperative day 6 and transferred to high-flow nasal cannula therapy, and her chest drains were removed on postoperative day 7. She subsequently made steady clinical progress and was discharged home clinically well, 16 days postoperatively.

Discussion

In this series, autologous pedicled pericardial patch repair was a versatile technique for treatment of large tracheobronchial defects arising from various scenarios. The technique is amenable to primary tracheobronchial repair, as in our second and third patients, and airway salvage following failed repair, as in our first patient. Tension-free anastomosis of healthy tracheobronchial edges is a prerequisite for successful repair but may not be obtainable in complex situations such as our cases. The defects in our patients were too large to close primarily without compromise of the airway, and they were too extensive to safely perform the required reconstruction without sacrificing the left lung in the second case and right lung in the third case. Other authors have reported lung resection in patients with tracheobronchial injuries possibly because of extensive devitalisation of tissues and limitations of repair.[2,17] However, surgical repair with lung preservation is preferred over pneumonectomy because of the higher mortality associated with performing post-traumatic pneumonectomy.[17] Utilisation of a pedicled pericardial flap with repair under CPB therefore offers a viable option for airway reconstruction in these complex cases. This technique provides autologous tissue and an airtight seal. The pericardial flap is based on the phrenic artery as previously described.[13,14]

Briefly, an adequate strip of a superiorly based pedicled pericardial flap is harvested

from the right side of the pericardium. The right phrenic nerve is carefully visualised and preserved. The flap is based on the vascularity of the phrenic vessels. Its superiorly based reflection facilitates the transposition of the flap toward the airway and makes it a versatile tissue for reconstruction of airway defects irrespective of its location. Dissection behind the superior vena cava (SVC) is performed to create a tunnel through which the patch is threaded beneath the SVC. The pedicled patch can also be passed over the SVC to avoid tension on the harvested tissue when extending to the site of the tracheobronchial defect. The free end of the patch is anastomosed to the tracheal or bronchial wall (the edges of the airway defect) with continuous 5-0 polydioxanone sutures, taking full-thickness bites in the patch and the trachea or bronchi. The smooth visceral surface of the pericardium is oriented to lie in the luminal side of the airway. The endotracheal tube is adjusted under bronchoscopic control so that it is located at the centre of the repair and immediately above the carina. Water immersion is used to test for air leakage under 40 cm H_2O pressure to ensure an airtight repair. Fibrin sealant glue is applied to the suture line and covered with an oxidised regenerated cellulose absorbable haemostat. An irrigation catheter is placed in the mediastinum and exits from the subclavicular skin as part of our tracheal protocol. Mediastinal irrigation is performed with povidone-iodine for 48 hours and with normal saline for 72 hours. Endoscopy at the conclusion of the operation is mandatory to assess the repair and ensure a stable airway without collapse on inspiration or expiration.

Notably, most blunt tracheobronchial injuries are located within 2 cm of the carina, as seen in our second and third patients.[2,17–19] Several authors have therefore suggested a right or left posterolateral thoracotomy approach for this reason.[2,7,17,18,20] However, in injuries involving the carina, lower trachea, and left mainstem bronchus, as in all three of our patients, median sternotomy affords the best operative access.[15,20] Furthermore, the highest mortality rate in patients with airway trauma occurs in patients with bilateral bronchial injuries.[7] We therefore posit that repair under CPB for efficient management of physiology, and with ECMO back-up as described in this series, is another option with possibly improved outcomes. Our review of the literature suggests limited awareness and use of this important live-saving option. Such beating-heart surgery under CPB is performed under reduced pressure with enough time for complex repairs.[15]

Follow-up of these patients using bronchoscopy and bronchography is necessary to identify complications, including bronchial stenosis. This enables early endobronchial intervention in the form of mechanical dilatation with or without stents, as in our first case. Moreover, primary endobronchial intervention is increasingly being explored for patients with severe associated injuries or as a follow-up to conservative management in patients with limited tracheal tears complicated by fibrotic stenosis.[6,21] Stents are usually placed via a rigid bronchoscope under general anaesthesia. Custom-made biodegradable stents made of polydioxanone material are helpful in reducing the risks of erosion and in-stent stenosis posed by earlier metallic stents.[22] The airway integrity is well maintained for 2 months, beyond which the biodegradable stents start to dissolve; they are completely dissolved by 3 months, thereby avoiding the need for permanent stenting.

In conclusion, paediatric airway surgery in the setting of severe tracheobronchial injuries is complex and requires a dedicated interdisciplinary team. We believe that adding the option of pedicled pericardial repair not only enriches the surgical

armamentarium but also gives room for tracheobronchial growth. In addition, this repair technique reduces the incidence of dehiscence because of the continuous blood supply to the harvested pericardium. The early results appear promising, and further study is required to demonstrate the extent of possible reformation of this vascularised pericardial flap within the respiratory endothelium and the capacity for ciliary formation.

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Conflict of interest

The authors declare that there is no conflict of interest.

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