

**Case
Report**

Tracheoplasty for Congenital Tracheal Stenosis with Bilateral Tracheal Bronchus

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Introduction: Congenital tracheal stenosis (CTS) with a bilateral tracheal bronchus (TB) has not been reported as a subtype of CTS. A novel technique to manage CTS in patients with a bilateral TB is described.

Case Report: An infant with tetralogy of Fallot underwent repair of cardiac anomaly at age 1 month. He experienced numerous cyanosis and episodes of transient respiratory arrest. Chest computed tomography (CT) demonstrated an aberrant bilateral upper lobe bronchus arising directly from the trachea and a stenotic trachea connecting the pseudo-carina to the true carina between the common right lower and left lower bronchus. On bronchoscopy, the diameter of the lumen of the narrowed segment was estimated to be less than 2 mm. Tracheal reconstruction was undertaken when he was 2 years of age. The surgical technique using a modified slide tracheoplasty for the correction of this anomaly are described. After surgery, the patient was extubated and has had no respiratory symptoms.

Discussion and Conclusion: The patient had unique anatomic considerations that made reconstruction challenging. Our technique of covering a stenotic section by normal trachea is a modification of the slide tracheoplasty technique and is useful for CTS with a unilateral and a bilateral TB.

Keywords: congenital tracheal stenosis, tracheal bronchus, slide tracheoplasty

Introduction

Congenital tracheal stenosis (CTS) is characterized by complete tracheal rings and is associated with high post-operative mortality.¹⁾ The presence of a tracheal bronchus (TB) is a relatively rare subtype of CTS. Previous studies

reported that an end-to-end anastomosis or conventional or lateral slide tracheoplasty after isolation or resection between the TB and the bifurcation is performed in patients with combined unilateral bronchial stenosis.^{2–4)}

However, bilateral TB has not been reported as a subtype of CTS. Bilateral TB constitutes a unique surgical challenge because the lower trachea is fixed due to the relationship between the bronchus and the lung, and it can be associated with vascular anomalies, making reconstruction challenging.

A novel technique to manage tracheal stenosis in patients with a bilateral TB, using a modified slide tracheoplasty, is described.

Case Report

A 37-week-male premature twin with tetralogy of Fallot and CTS with a bilateral TB underwent repair of a ventricular septal defect and right ventricular outflow

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Received: July 17, 2019; Accepted: November 23, 2019
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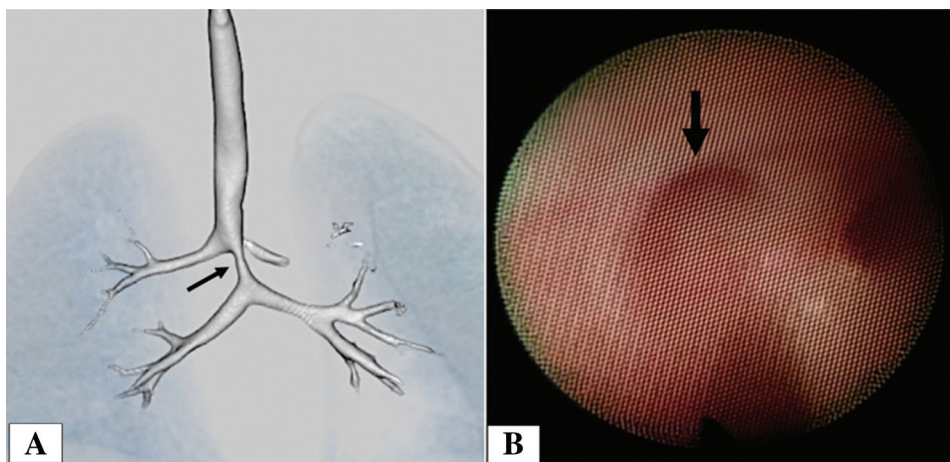


Fig. 1 Preoperative findings. **(A)** Preoperative three-dimensional computed tomography scan demonstrating a congenital tracheal stenosis with a bilateral tracheal bronchus. Black arrows: Narrowest point of trachea. **(B)** Preoperative bronchoscopic view. Black arrows: Narrowest point of trachea.

tract muscle resection at age 1 month. He was ultimately extubated and discharged home, but he experienced numerous respiratory infections, cyanosis, and episodes of transient respiratory arrest in the ensuing months. Computed tomography (CT) showed a narrowed segment of tracheal stenosis with a bilateral TB. Three-dimensional chest CT demonstrated an aberrant bilateral upper lobe bronchus arising directly from the trachea and a stenotic trachea connecting the pseudo-carina to the true carina between the common right lower and left lower bronchus (**Fig. 1**). On bronchoscopy, the diameter of the lumen of the narrowed segment was estimated to be less than 2 mm.

Surgical intervention was undertaken when the patient was 2 years of age and weighed 8.95 kg. Through a median sternotomy, cardiopulmonary bypass (CPB) was initiated. Wide dissection of the trachea was carried out from the level of the lower thyroid to the level of the true carina inferiorly. Then, the trachea, carina, right upper lobe bronchus, and intermediate bronchus were all identified (**Fig. 2A**). After the institution of CPB, the endotracheal tube was removed. The extent and degree of stenosis and the complete cartilage ring were confirmed. The stenotic segment was noted to be approximately 1.5 cm long; its luminal diameter was 1 mm. The trachea was transected above the bilateral TB. The distal trachea was cut at the front side to the bifurcation of the lower lobe bronchi. The proximal segment was cut at the back side, up into the trachea of the corresponding length for

slide tracheobronchial plasty. The incision range was 2 cm on both the proximal side and the distal side (**Fig. 2B**). The anastomosis was completed with a 5-0 polydioxanone suture (PDS II, Ethicon, Somerville, NJ, USA) (**Fig. 2C**). The bypass time was 152 minutes. The patient was weaned from the ventilator and successfully extubated 14 days after the operation. He was discharged 4 weeks later. A CT scan (**Fig. 3A**) and bronchoscopy (**Fig. 3B**) performed 6 months later disclosed a well-reconstructed trachea and bronchi. He is well-being for 3 years after surgery without any supplementary oxygen or mechanical ventilation.

Discussion and Conclusion

CTS was previously associated with mortality rates as high as 50%.¹⁾ Various surgical techniques have been developed for the treatment of CTS, with acceptable rates of operative mortality (7–13%).⁵⁻⁷⁾ Slide tracheoplasty has been hailed as a revolution in the management of CTS, achieving significantly better results.⁵⁾ For the treatment of long segment lesions, slide tracheoplasty has largely supplanted tracheal patching with either costal cartilage or pericardium because of concerns about high rates of re-intervention with the latter techniques. On the other hand, end-to-end anastomoses are reserved for patients with short-segment CTS. However, surgical management of tracheal stenosis with abnormal arborization or involvement of the bronchi remains challenging.⁸⁾

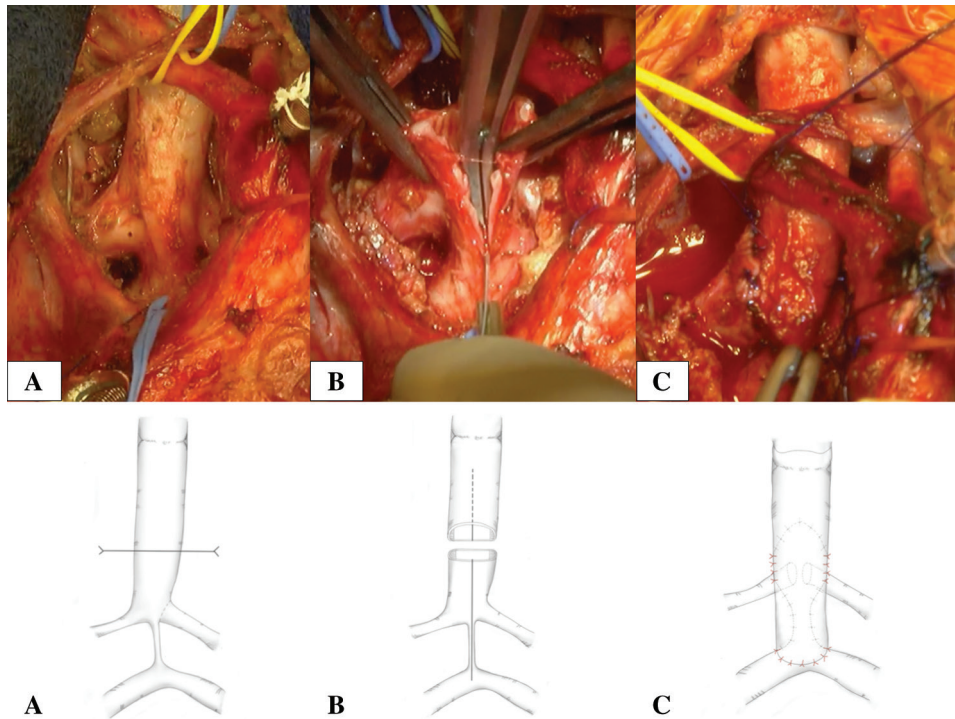


Fig. 2 Operative technique. (A) Sternotomy approach: an aberrant bilateral upper lobe bronchus arising directly from the trachea and a stenotic trachea connecting the pseudo-carina to the true carina between the common right lower and left lower bronchus are seen. (B) The stenotic trachea is transected at the trachea above the bilateral tracheal bronchus. The distal trachea is cut at the front side to the bifurcation of the middle and lower lobe bronchi. The proximal segment is cut at the back side, up into the trachea of the corresponding length for slide tracheobronchial plasty. (C) The anastomosis is completed.

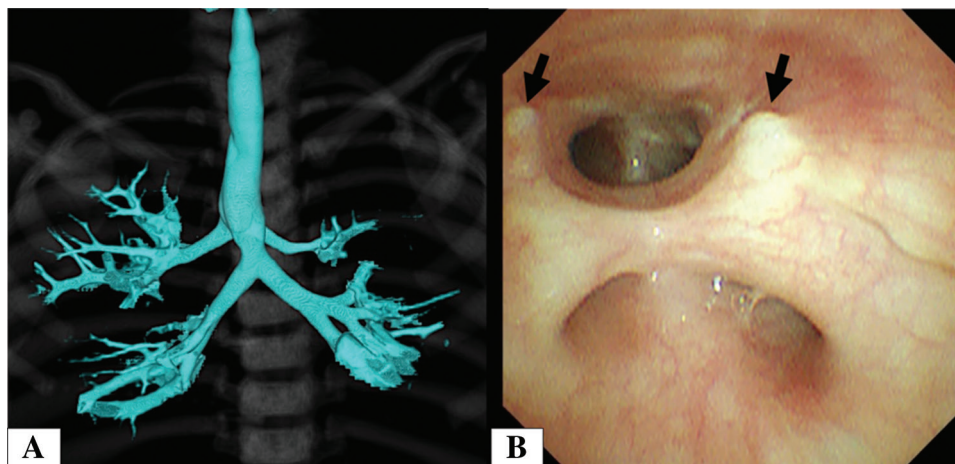


Fig. 3 (A) Follow-up three-dimensional computed tomographic view on postoperative 6 months showing no stenosis. (B) Follow-up bronchoscopic view on postoperative 6 months, showing clear-appearing distal airway with no evidence of granulation tissue. Black arrows: Anastomosis.

In the present case, the stenotic trachea was fixed with a bilateral TB and bilateral bronchi. Therefore, it seemed that postoperative anastomotic leakage, consequent bronchomalacia, and bronchial flexion might occur if the method of resection at the center of the stenotic trachea and performance of an end-to-end anastomosis were chosen. Furthermore, because the most stenotic part was very narrow, it was thought that the formation of the slide tracheoplasty of the narrowed part would not result in a large diameter, and the possibility of restenosis was high. As a technique that can be passive and can secure a large diameter, it was divided with a normal trachea on the proximal side of the narrowed part, and slide tracheoplasty was performed.

We propose that if the length of the anastomosis is appropriate, surgery to anastomose a thick trachea and a stenotic segment is more stable than slide tracheoplasty with anastomosis between stenotic segments for CTS with a bilateral TB. However, we consider that this technique is also useful for CTS with a TB on one side. In our previous experience, we performed sliding trachea formation on CTS with a TB on one side that was also cut at the upper border of the stenotic trachea, which provided a good result.

In conclusion, the resection and reconstruction in this case were challenging because of the patient's unique anatomy. The patient had unique anatomic considerations that made reconstruction challenging. Our technique of covering a stenotic section by normal trachea is a modification of the slide tracheoplasty technique and is useful in patients with a unilateral and a bilateral TB.

Disclosure Statement

The authors report no conflicts of interest.

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