Weber et al. Clin Med Rev Case Rep 2015, 2:12
ISSN: 2378-3656

Clinical Medical Reviews and Case Reports

Case Report: Open Access

Long-Segment Slide Tracheo-Bronchoplasty (LSTB) with Contralateral Lung Agenesis: Case Report and Review of the Literature

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Abbreviations

LSTB: Long-segment slide tracheo-bronchoplasty, LSTS: Long-segment tracheal stenosis, STB: Slide tracheo-bronchoplasty

The management of long-segment tracheal stenosis (LSTS) can be quite challenging, particularly in the presence of other pulmonary anomalies. We present a complicated case of LSTS and left bronchial stenosis in a 4-month-old infant with a congenitally absent right lung. This case emphasize that a slide tracheo-bronchoplasty(STB) in the setting of congenital absence of the contralateral lung is complicated but can be performed with reasonable results.

Clinical Summary

A 4-month-old girl with LSTS and congenital absence of the right lung had been a noisy breather but doing quite well until she suddenly developed respiratory failure requiring emergent intubation and ventilatory support. She was transferred to our center where a CT scan demonstrated a LSTS (Figure 1A, Figure 1B and Figure 1C) with and absent right lung. Bronchoscopy revealed complete tracheal rings with a funnel shaped trachea starting at the third tracheal ring and tapering down into the distal trachea and left bronchus. The patient was urgently taken to the operating room for a STB. After mobilization of the trachea down to the left main stem bronchus we noted that the left bronchus was 50% narrowed down to the left upper lobe takeoff and had complete bronchial rings. A STB was performed from the third tracheal ring to the left upper lobe takeoff. Due to systemic pulmonary artery pressures when initially separating from cardiopulmonary bypass as well as some mild hemodynamic instability, the sternum was initially left open and the skin was closed. Five days later the sternum was closed after her severe pulmonary hypertension resolved.

Her postoperative course was complicated by recurrent stenosis toward the distal aspect of the repair due to transverse aortic arch

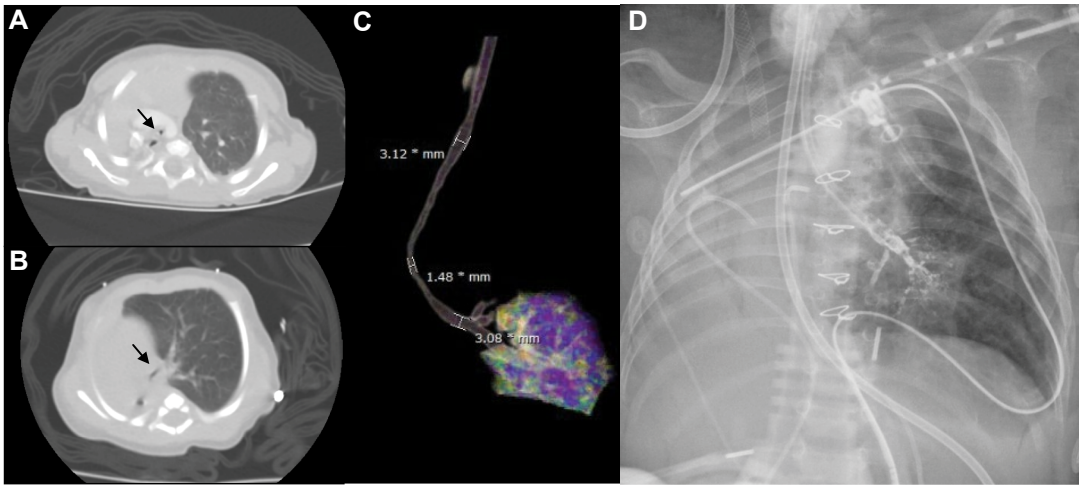

Figure 1: Representative computed tomographic images of long-segment tracheal and left mainstem bronchial stenosis with right lung agenesis. (A) Axial panel with severe retroaortic stenosis of distal trachea (arrow); (B) Axial panel with stenosis extending into left main-stem bronchus (arrow); (C) Three-dimensional airway reconstruction with measurements of proximal and distal trachea as well as left bronchus showing the morphology of the stenosis; (D) Fluoroscopic study with contrast instilled via endotracheal tube showing likely stenosis in distal left mainstem airway.

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Citation: Weber DJ, Rodefeld MD, Matt BH, Brown JW (2015) Long-Segment Slide Tracheo-Bronchoplasty (LSTB) with Contralateral Lung Agenesis: Case Report and Review of the Literature. Clin Med Rev Case Rep 2:077

Received: September 26, 2015: **Accepted:** December 15, 2015: **Published:** December 18, 2015

Table 1: Summary table of recent papers on tracheal stenosis with relevant findings.

Authors, Year (Reference)	Study Design	Relevant Findings
Manning et al., 2011, [1]	Single institution review of 80 patients undergoing slide tracheoplasties over an 8-year period	- 95% overall survival (2 early deaths and 2 late deaths) - Lower complication and reintervention rates with slide tracheoplasty compared to previous techniques
Butler et al., 2014, [2]	Single institution review of 101 patients undergoing slide tracheoplasties over a 17-year period	- 88% overall survival - Largest series of slide tracheoplasties reported - Preoperative ECMO, bronchomalacia, and bronchial stenosis predicted worse survival - Single lung not predictive of worse survival
Fanous et al. 2010, [3]	Single institution review of 26 patients undergoing anterior pericardial tracheoplasties over a 28-year period	- 81% overall survival (3 early deaths and 2 late deaths) - Advantage of extended long-term follow-up (3 months to 22 years)
Anton-Pacheco et al., 2003, [4]	Single institution review of 13 patients with congenital tracheal stenosis looking at predictors of outcome including the morphology and location of stenosis	- 77% overall survival with a variety of techniques - Short segment stenosis can be treated with resection while long segment disease is best treated with slide tracheoplasty - High mortality (50%) with single lung
Backer et al. 2009, [6]	Single institution review of 71 patients with congenital tracheal stenosis and unilateral lung agenesis or hypoplasia undergoing 4 different surgeries over a 30-year time period	- Similar overall survival among patients with two lungs compared to those with a hypoplastic or absent lung (83% vs 84%) - Slide tracheoplasty is the procedure of choice in those with bilateral lungs as well as those with a hypoplastic or absent lung

anterior compression with her heart in the far right chest. When her endotracheal tube was inadvertently pulled back and could not properly replaced she required 3 days of neck ECMO support due to airway compression and was taken back to the operating room for a pericardial patch tracheoplasty for the distal tracheal obstruction (Figure 1D) which healed without issues. She was weaned from ECMO support, had a custom tracheostomy tube placed through the patch site, and was discharged home with ventilator support under the care of her family.

Discussion

The slide tracheoplasty has become the preferred method for repair of long-segment tracheal stenosis in children. Studies have demonstrated low mortality rates in the face of a very challenging problem (Table 1). When compared with previous techniques, it offers a versatile approach with documented mortality rates in the range of 5-20% from recent reports out of two large volume centers [1,2]. These rates appear even better than the results we published in 2009 using the anterior pericardial patch technique. However, it should be noted that we looked at long-term results over more than twenty years where we found 16 of the 26 patients were still alive [3]. In the largest series of slide tracheoplasties recently reported, 47% of the slide tracheoplasties were carried into the proximal bronchus and 9% of patients presented with a single lung. Using multivariate analysis, neither the presence of a single lung nor extending the tracheoplasty into the proximal bronchus was associated with increased risk of death or stent requirement post-operatively [2].

Survival with long-segment tracheal stenosis in the setting of lung agenesis is documented to be exceedingly poor with mortality rates around 30% to 50% from small case series over the last fifteen years [4,5]. More recently, successful tracheoplasties have been documented in patients with unilateral lung agenesis or severe hypoplasia [6]. Outcomes from 11 single-lung patients (7 with a congenitally absent lung and 4 with a hypoplastic lung) were found to be similar to 60 patients with two lungs. In particular, the mortality of the single lung group was 18% while it was 24% from the double lung group. Of the 11 with single lungs, 3 underwent slide tracheoplasty (the more recent cases) while 8 of the 60 underwent slide tracheoplasty in the double lung group. Interestingly, slide tracheoplasty was not carried into the bronchus in any of the single lung patients as was necessary in our patient. We have previously encountered two similar cases of an absent contralateral lung but this was our first patient where bronchoplasty was required as well.

Certainly extension of stenosis into the bronchus in the setting on an absent contralateral lung adds to the complexity of the repair and likely increases the morbidity and mortality of the slide tracheoplasty. Consequently, post-operative vigilance is required for complications as well as the development of pulmonary hypertension. Ultimately, good results can be obtained in this challenging group of children. This reinforces recent reports demonstrating that improved outcomes are being obtained with the slide tracheoplasty technique even with coexisting complex anomalies.

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