Received: 29 May 2024 | Accepted: 2 August 2024

DOI: 10.1002/rcr2.1451

CASE REPORT

Respirology Case Reports OPEN ACCESS APSR WILEY

The use of overlapping self-expandable covered stents in the management of long-segment tracheobronchomalacia: A case report

Therese Pauline F. Yap[1] | **Wen-Jue Soong**[2,3,4]

[1]Department of Pediatrics, Section of Pediatric Pulmonology, Cardinal Santos Medical Center, San Juan City, Philippines

[2]Divison of Pediatric Pulmonology, Children's Hospital, China Medical University, Taichung, Taiwan

[3]Department of Pediatrics, Taipei Veterans General Hospital, Taipei, Taiwan

[4]Department of Pediatrics, Triservice General Hospital, National Defense Medical Center, Taipei, Taiwan

Correspondence
Therese Pauline F. Yap, Department of Pediatrics, Section of Pediatric Pulmonology, Cardinal Santos Medical Center, San Juan City, Philippines.
Email: thereseyapmd@gmail.com

Associate Editor: Daniel Ng

Abstract

Tracheomalacia is a condition where the tracheal wall is abnormally soft and prone to collapse during increased respiratory efforts. Airway malacia can manifest as segmental conditions like laryngomalacia, tracheomalacia and bronchomalacia, or as diffuse conditions such as tracheobronchomalacia (TBM). Unlike long-segment congenital tracheal stenosis, where surgery may be the preferred treatment, the management of long-segment TBM remains controversial.

KEYWORDS
airway, flexible bronchoscopy, stent, tracheobronchomalacia

INTRODUCTION

Tracheomalacia (TM) is a condition characterized by abnormal weakness and easy collapsibility of the tracheal lumen, particularly during periods of increased respiratory effort. It can be localized or diffuse, depending on the extent of the disorder. Primary diffuse TM, or TBM, is a rare congenital condition characterized by immature cartilaginous rings, most commonly affecting the distal third of the trachea, leading to weakness of the entire tracheal structure.[1,2] Congenital TM often occurs in preterm infants and may be associated with other congenital malformations such as congenital heart disease, vascular rings, tracheoesophageal fistula, oesophageal atresia and gastroesophageal reflux. The rigidity of the tracheal cartilage prevents collapse during expiration.[2] Common symptoms observed in patients with tracheal collapse include stridor, apnea, brassy cough, cyanotic spells, and recurrent pneumonia.

The management of TM remains controversial. Focal tracheomalacia can be addressed with an airway stent; however, in cases of severe and diffuse TM, tracheostomy is preferred, to maintain airway patency with continuous positive airway pressure.[3] This case illustrates a successful treatment using multiple overlapping stents in a paediatric patient with long-segment tracheobronchomalacia.

CASE REPORT

A 14-year-old female was referred from another institution for further evaluation and management. She was born full term, and developed stridor, recurrent choking, vomiting, and failure to thrive. Eventually, she was diagnosed with large atrial septal defect post-Amplatzer septal occluder placement, tracheoesophageal fistula and oesophageal atresia post-surgical repair, severe gastroesophageal reflux post-surgical repair, laryngeal cleft type II, and severe long-segment TM with bronchomalacia.

With parental preference, the long-segment TBM was initially managed by the referring hospital by inserting two

balloon-expandable stents in the trachea and RMB. The tracheal stent was eventually removed due to deformity caused by gradual balloon dilatations and the growing lumen. Retrieval of the RMB stent proved to be challenging; therefore, it was kept patent and in place using balloon dilatation. She was discharged without stridor or feeding issues.

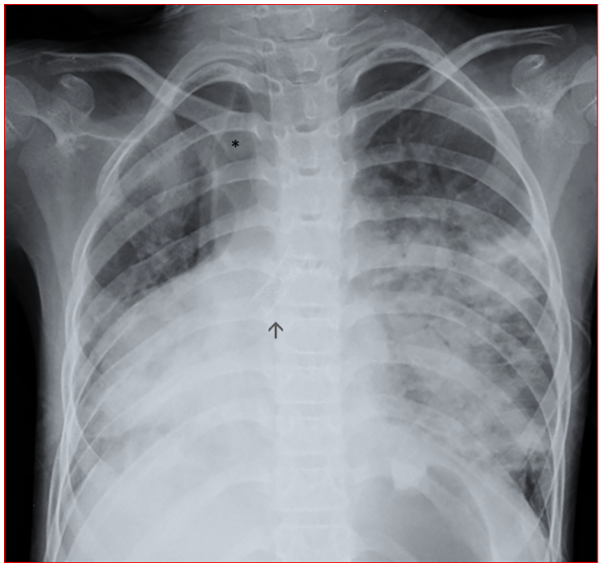

FIGURE 1 Chest radiograph. Bilateral pneumonia with dextroposition of heart because of right low lung collapse; a huge air-filled oesophagus* and a metallic stent in the right main bronchus.

Recommendations include using oxygen as necessary and prescribing diuretics, pulmonary vasodilators, inhaled bronchodilators, and anti-reflux medications.

The patient was admitted to our paediatric intensive care unit for unilateral opacification of the right hemithorax (Figure 1). A chest CT scan showed decreased right lung volume with multiple lung cysts and increased ground glass opacities in the left lower lobe, with atelectasis in the lingular lobe.

The team proceeded with flexible bronchoscopy. Topically, 2% lidocaine was instilled in both nostrils, larynx, trachea, carina, and bronchi. The patient received procedural sedation by titrating intravenous midazolam (0.3 mg/kg/dose), ketamine (1.5 mg/kg/dose), and fentanyl (1.5mcg/kg/dose). With proper sedation and Soong's ventilation[4,5] for airway oxygenation and ventilation support, a short-length flexible fiberoptic bronchoscope (FFB) (*Olympus*, ENF-VT2, Japan) was used to evaluate the entire airway.

The examination revealed severe gastroesophageal reflux with airway aspiration, purulent material pooling throughout all visible airway lumen, total collapse of the trachea (approximately 10 cm, from subglottis to carina), carinal trifurcation (Figure 2), and a fracture in the RMB stent. Initially, a 10 mm balloon catheter *(Boston Scientific)*, was used for dilatation and repair to restore the patency in the RMB lumen and stent. Subsequently, a self-expandable covered stent (Bonastent, 14 mm diameter × 40 mm length) was deployed using the same FFB from the carina to the mid-portion of the trachea. However, the proximal tracheal

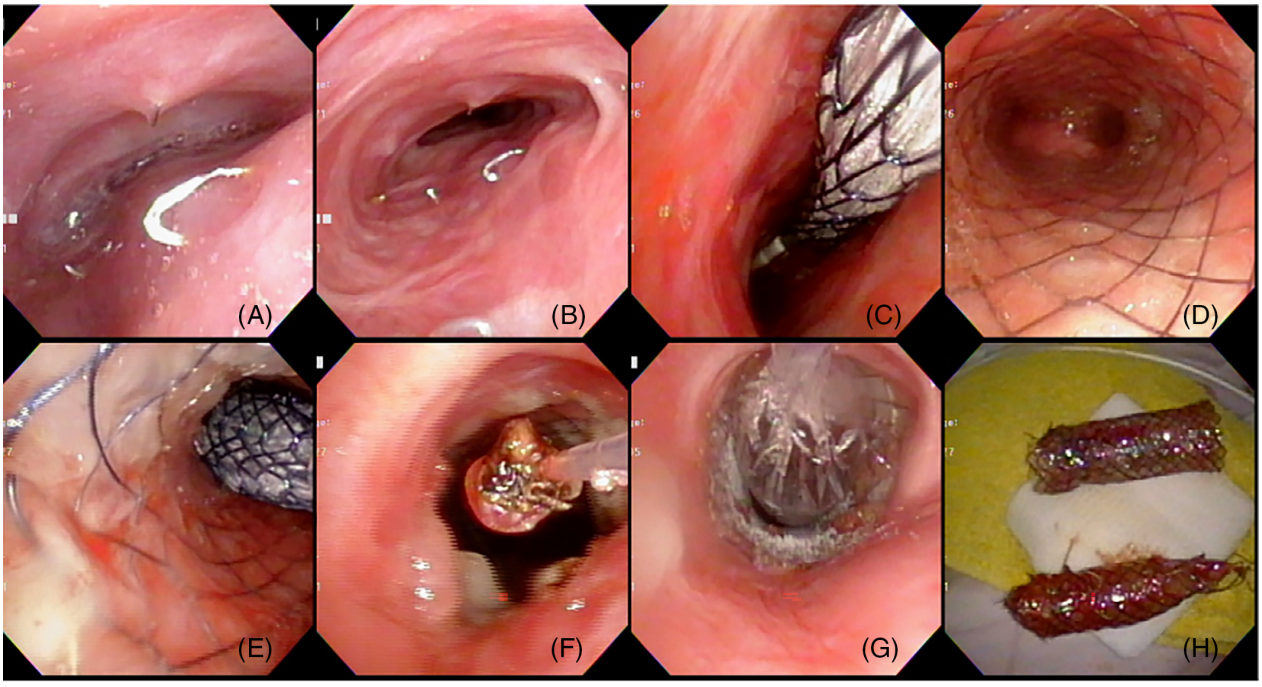

FIGURE 2 Flexible bronchoscopy showed (A) and (B) severe dynamic tracheomalacia, (C) self-expandable metallic covered stent insertion on the distal trachea, (D) deployed and fully expanded stent on the distal trachea, (E) positioning the proximal tracheal stent overlapping with the distal tracheal stent, (F) removal of stent granuloma with laser ablation, (G) balloon dilatation on malapposed stent, and (H) retrieved self-expandable covered metallic stents. The entire procedure of flexible interventional bronchoscopy was supported by Soong's ventilation.

lumen remained collapsed. Therefore, a second tracheal stent of the same type was inserted, overlapping approximately 10 mm with the proximal portion of the first distal stent.

The position and patency of the two stents were checked with FFB, revealing luminal patency from 15 mm below the vocal cords to the level of the carina and bronchi. The patient's vital signs, including the heart rate, respiratory rate, blood pressure and oxygen saturation were continuously monitored throughout the procedure and during recovery. She exhibited normal vital signs and remained clinically stable without encountering any complications such as desaturations, apnea, cyanosis, arrhythmia, or bronchospasm. The patient was discharged without any immediate complications associated with FFB or stent placement.

The patient was closely followed up with pulmonary function tests, FFB evaluations and regular replacement of the tracheal stents every 3–4 months.

DISCUSSION

The gold standard in diagnosing airway malacia is direct visualization of the lumen during spontaneous respiration with FFB. This method can demonstrate dynamic expansion and collapse during breathing cycles and also evaluate nasal tracts, adenoids, tonsils, vocal cords, and other lesions such as cysts, clefts and tracheoesophageal fistulas.[6] While TM is expected to improve spontaneously as the airway matures, some patients may require treatment depending on the severity of the condition. Patients with severe symptoms such as apneic spells, dyspnea, failure to thrive, recurrent pneumonia, and cyanosis are more likely to need surgical interventions such as tracheostomy and aortopexy.[1] These procedures will not be discussed in this report.

Airway stenting is a less invasive option for severe TM, and most experts recommend performing it with rigid bronchoscopy under general anaesthesia. The choice of stent is challenging, especially in younger children. Currently, only vascular balloon-expandable metallic stents with a diameter less than 10 mm are available for use in paediatric airways.[7]

As the 'ideal' stent has not yet been developed, the authors believe that the material of the stent is not the most crucial factor to consider when choosing which type to utilize. Paediatric patients who receive silicone stent implantation, particularly in the trachea, face a high risk of stent migration due to the poor fixation. Silicone stents also reduce the inner airway diameter and pose a risk of mucus plugging due to poor mucociliary clearance. Metallic stents are easier to deploy using FFB, provide better fixation, and are easier to detect with chest imaging during follow-up due to their radio-opacity. Regular and close follow up of patients should be conducted to anticipate granulation tissue formation, obstruction and stent fracture.

For long-segment TBM, the most common approach to management is surgical. In our patient, after undergoing multiple surgical procedures, the parents opted for a more conservative approach in addressing her complex airway, along with associated pulmonary hypertension, severe gastroesophageal reflux and aspiration, and cardiac abnormalities.

Conventional approaches to respiratory support during FFB include oxygenation with a face mask, laryngeal mask airway (LMA) or endotracheal tube (ETT).[4,8] In this patient, the procedure was safely carried out using a nasopharyngeal catheter delivering warmed and humidified 100% oxygen at a flow rate of 0.5–1 L/kg/min. Soong's ventilation, a non-invasive ventilation method involving pharyngeal oxygen catheter, intermittent nose closure and abdominal compressions,[4,9–11] was accessible and beneficial as needed by both the patient and FFB operator.

The major issue associated with the stent observed in our patient was the formation of granulation tissue at both edges, which are the most vulnerable injury sites of the mucosa. This was managed by laser ablation and balloon compression, facilitated using FFB with Soong's ventilation as respiratory support. Malposition or migration of the covered-stent was easily readjusted with grasping forceps during FFB. If a metallic stent fracture occurs, it can be managed by removing and implanting another stent if necessary. However, due to the difficulty in removing the fractured RMB metallic stent in this patient, patency was maintained with balloon dilatation to expand and accommodate the growing airway lumen, with plans to retrieve it at a more opportune time.

In conclusion, flexible fiberoptic bronchoscopy (FFB) in a patient with severe pulmonary involvement and a complex medical history presents significant challenges. Given the patient's complex medical history and compromised cardiopulmonary status, a collaborative effort among specialties including pulmonology, cardiology, intensive care, anesthesiology and thoracic surgery is crucial for thorough risk assessment and management during the procedure.

Overlapping tracheal and bronchial stent placement using an FFB, supported by Soong's ventilation, can be a safe alternative for the management of patients with long-segment tracheomalacia.

AUTHOR CONTRIBUTIONS

In this case report, Therese Pauline F. Yap and Wen-Ju Soong jointly conducted patient assessments, analysed medical data, and crafted the clinical narrative. Both authors contributed expertise and provided insights, ensuring a comprehensive examination and presentation of the case. Both authors equally participated in drafting and revising the manuscript.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

ORCID

Therese Pauline F. Yap https://orcid.org/0009-0001-5580-0342

REFERENCES

1. Yang D, Cascella M. Tracheomalacia. StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024. https://www.ncbi.nlm.nih.gov/books/NBK553191/
2. Vinograd I, Filler RM, Bahoric A. Long-term functional results of prosthetic airway splinting in tracheomalacia and bronchomalacia. J Pediatr Surg. 1987;22:38–41. https://doi.org/10.1016/S0022-3468(87)80011-8
3. Mok Q. Airway problems in neonates—a review of the current investigation and management strategies. Front Pediatr. 2017;5:60. https://doi.org/10.3389/fped.2017.00060
4. Soong W-J, Jeng M-J, Lee Y-S, Tsao P-C, Harloff M, Matthew Soong Y-H. A novel technique of non-invasive ventilation: pharyngeal oxygen with nose-closure and abdominal-compression—aid for pediatric flexible bronchoscopy. Pediatr Pulmonol. 2015;50:568–75. https://doi.org/10.1002/ppul.23028
5. Soong C, Yu-Sheng Lee YS, Lin CH, Chen CH, Soong WJ. Sustained pharyngeal inflation in infant airway—flexible bronchoscopy measurements. PLoS ONE. 2023;18(11):e0294029. https://doi.org/10.1371/journal.pone.0294029
6. Varela P, Torre M, Schweiger C, Nakamura H. Congenital tracheal malformations. Pediatr Surg Int. 2018;34:701–13. https://doi.org/10.1007/s00383-018-4291-8
7. Yang Y, Li X, Jiao A. Stent intervention for children with CHD and tracheal stenosis. Cardiol Young. 2020;30(10):1532–4. https://doi.org/10.1017/S1047951120002504
8. Midulla F, de Blic J, Barbato A, Bushz A, Eber E, Kotecha S, et al. Flexible endoscopy of paediatric airways. Eur Respir J. 2003;22:698–708.
9. Soong W-J, Tsao P-C, Lee Y-S, Yang C-F. Flexible endoscopy for pediatric tracheobronchial metallic stent placement, maintenance and long-term outcomes. PLoS one. 2018;13(2):e0192557. https://doi.org/10.1371/journal.pone.0192557
10. Soong WJ. Pediatric interventional flexible bronchoscopy. Pediatr Resp Crit Care Med. 2018;2:38–44. http://www.prccm.org on Monday, October 29, 2018, IP: 203.64.247.4.
11. Soong WJ, Tsao PC, Lee YS, Yang CF. Therapeutic flexible airway endoscopy of small children in a tertiary referral center—11 years' experience. PLoS one. 2017;12(8):e0183078. https://doi.org/10.1371/journal.pone.0183078

How to cite this article: Yap TPF, Soong W-J. The use of overlapping self-expandable covered stents in the management of long-segment tracheobronchomalacia: A case report. Respirology Case Reports. 2024;12(8):e01451. https://doi.org/10.1002/rcr2.1451