

# Long-term pneumatic stenting with positive expiratory pressure therapy for severe expiratory central airway collapse

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## Abstract

Expiratory central airway collapse (ECAC) comprising excessive central airway collapse (EDAC) and tracheobronchomalacia. Treatment is challenging for severe cases that are not candidates for surgical management. We report a case of severe ECAC successfully managed with continuous positive airway pressure (CPAP) therapy. A 75-year-old female patient status post right pneumonectomy, presented with chronic cough. Dynamic bronchoscopy evaluation showed severe EDAC which improved with intraprocedural noninvasive positive pressure (NIPPV) therapy. Due to patients' comorbidities, she was not a candidate to surgical tracheobronchoplasty. Therefore, we attempted pneumatic stenting with long-term CPAP therapy resulting in improvement of symptoms and functional capacity. The long-term efficacy of pneumatic stenting has not been clearly established yet. Literature review of management of ECAC with NIPPV consist primarily of case reports and there is only one clinical trial being conducted to assess the efficacy of CPAP therapy in ECAC. While NIPPV arises as a sufficient alternative for management of severe ECAC, larger scale studies are needed to prove the real efficacy of NIPPV in this setting.

## KEYWORDS

CPAP, expiratory central airway collapse, noninvasive positive pressure ventilation, pneumatic stent

## INTRODUCTION

Expiratory central airway collapse (ECAC) is pathological dynamic narrowing of the central airways during expiration that includes excessive dynamic airway collapse (EDAC) and tracheobronchomalacia (TBM). EDAC is defined as a forward bulging of the posterior membranous tracheal wall into the airway lumen during expiration secondary to atrophy of the smooth muscle fibres. TBM is characterized by the weakness of the anterior or lateral tracheobronchial cartilage. Although a disease prevalence of 13% has been suggested, a lack of awareness of the condition coupled with a consensus definition suggests the actual prevalence may be greater. (1–3). Diagnosis requires a high index of suspicion, as the presenting symptoms of chronic cough and dyspnea are common and non-specific. Additionally, patients often have comorbid conditions including asthma, chronic obstructive pulmonary disease, gastroesophageal reflux, and

obstructive sleep apnea (OSA) to which symptoms are ECAC are incorrectly ascribed delaying an accurate diagnosis. Dynamic flexible bronchoscopy is the current gold standard for diagnosing ECAC (1,3,4).

Initial treatment for ECAC includes supportive measures, pulmonary hygiene in those who complain of inability to clear respiratory secretions, and maximization of medical therapy for concomitant diseases. Mild to moderate disease can be managed with pneumatic stenting with noninvasive positive pressure ventilation (NIPPV) in the mode of intermittent continuous positive pressure (CPAP) or bilevel positive pressure (BiPAP). Patients with severe ECAC are generally best managed with surgical repair by tracheobronchoplasty (TBP) if an airway stent trial suggests clinical benefit of maintaining structural integrity of the airway (1,5). We report a case of severe ECAC presenting with chronic cough that was successfully managed with long-term pneumatic stenting with CPAP and a literature review on this therapeutic approach.

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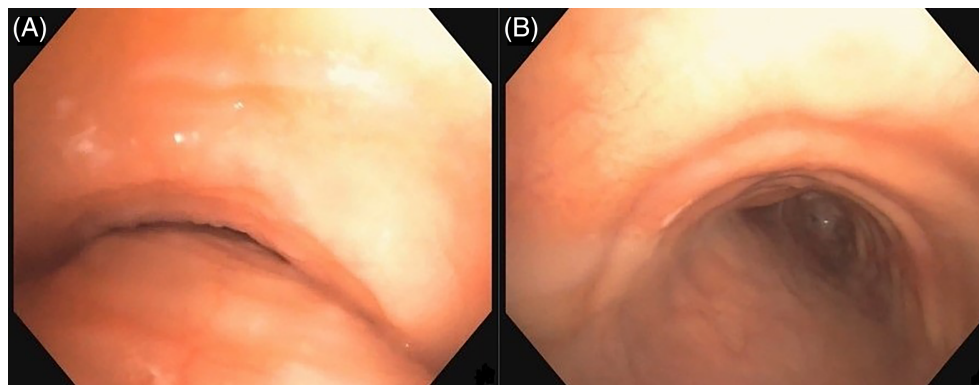
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## CASE REPORT

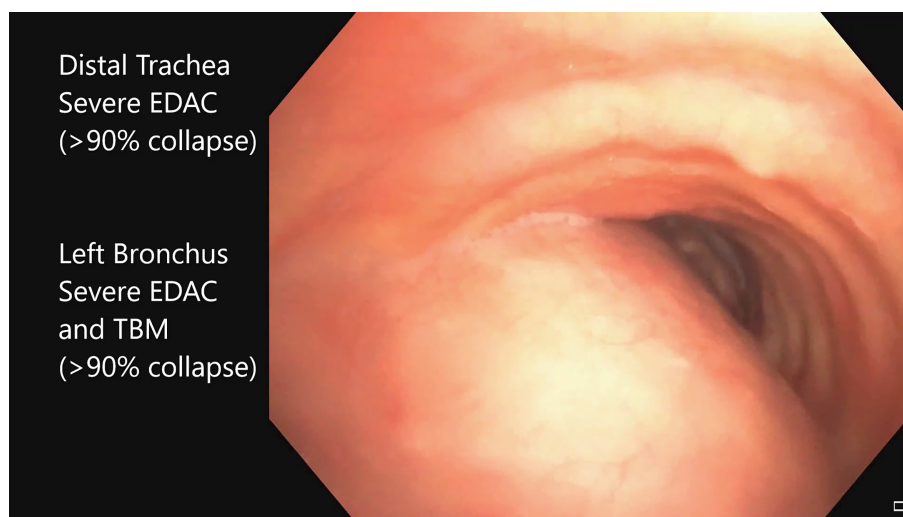
A 75-year-old female, with past medical history of right pneumonectomy secondary to squamous cell carcinoma, presented with a one-month history of severe, persistent non-productive cough. A chest computed tomography (CT) showed a focus of tree in bud nodularity in the left lower lobe likely reflective of mild infectious or inflammatory airway process. One month later, symptoms persisted despite treatment with prednisone, levofloxacin, nebulized ipratropium, bromide/albuterol, and bronchial hygiene. Other medications included home oxygen requirement at 2 L/min on exertion.

Airway inspection with a standard flexible bronchoscopy suggested moderate collapse in the mid and distal trachea and moderate collapse in the left mainstem bronchus with significant mucopurulent secretions. Bronchial wash was negative for infection. Dynamic bronchoscopy showed mild

EDAC (70%–80% collapse) in the proximal trachea, moderate to severe EDAC (80%–90% collapse) in the mid trachea, severe EDAC (> 90% collapse) in the distal trachea (Figure 1A), and severe ECAC (> 90% collapse) in the variety of EDAC and TBM in the left mainstem bronchus (Video 1; Figure 2A). The overall severity index for EDAC was 9 points (6), indicating a classification of overall severe EDAC. CPAP titration for pneumatic stenting was performed during the procedure: at 8–10 cmH<sub>2</sub>O, there was no clinically evident ECAC with tidal breathing and dynamic exhalation (Video 1; Figures 1B and 2B). These findings were likely present prior to her chronic cough presentation and potentially exacerbated in the setting of a new infectious process. A stent trial was not pursued as the patient was not a surgical candidate due to her prior right pneumonectomy. TBP is usually performed through a right sided thoracotomy and upon evaluation, it was determined an excessive surgical risk due to the surrounding scar tissue in the potential

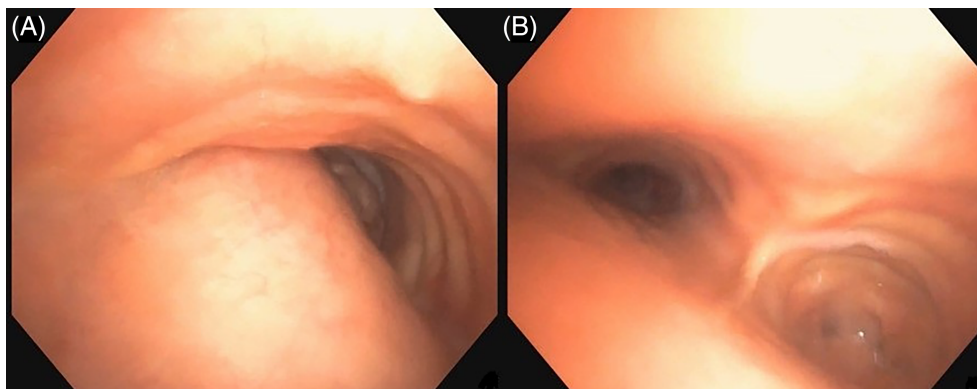


**FIGURE 1** Dynamic bronchoscopy demonstrating (A) Severe EDAC (>90% collapse) of the distal trachea; (B) CPAP titration at 8–10 cmH<sub>2</sub>O in distal trachea, maintaining airway patency.



**VIDEO 1** Dynamic bronchoscopy footage demonstrating severe ECAC and subsequently CPAP titration with pneumatic stenting resolution throughout the distal trachea and the left mainstem bronchus.

Video content can be viewed at <https://onlinelibrary.wiley.com/doi/10.1002/rcr2.1367>



**FIGURE 2** Dynamic bronchoscopy demonstrating (A) Severe ECAC (>90% collapse) of left mainstem bronchus; (B) CPAP titration at 8–10 cmH<sub>2</sub>O in left mainstem bronchus, maintaining airway patency.

surgical area. In addition to her home medications and nocturnal CPAP use, airway clearance therapy using nebulized albuterol followed by 3% saline and intermittent CPAP sessions of 30–60 min at 10cmH<sub>2</sub>O three times a day, respectively, were recommended. Patient compliance with this therapy was excellent.

As a former physician, the patient created a journal describing symptoms and functional capacity during the positive expiratory pressure daily sessions, measured by two fixed points, treadmill rate (MPH), O<sub>2</sub> flow (L/Min) need, and one variable point, pulsimeter saturation (SpO<sub>2</sub>). Prior to CPAP use, SpO<sub>2</sub> decreased during treadmill use at 3.5 MPH from 93%–94% to 85%–87%, and O<sub>2</sub> flow at 4 L/min flow continuously was required to maintain adequate saturation. With adherence to this treatment plan, her symptoms gradually improved over a two-month period, and she attained her baseline pulmonary functional status. A trial of medical therapy consistent with aggressive bronchial hygiene protocol three times per day consistent with nebulized albuterol and nebulized 3% saline, without CPAP sessions, was attempted to decrease treatment burden but lead to worsened symptom control and oxygenation. CPAP therapy was reinstated, and over the next 7–8 days, her respiratory status was restored to her baseline. No further interventions were performed. The patient remained compliant with her CPAP therapy and bronchial hygiene with appropriate control of her symptoms for a total of 6 months, until her passing due to a non-pulmonary malignancy.

## DISCUSSION

Commonly, ECAC can coexist in patients with other respiratory conditions. Identification of ECAC and institution of specific therapy is indicated. ECAC is associated with higher morbidity and lower survival rates in elderly patients who have undergone bronchial or bronchovascular sleeve resections for lung cancer (7).

Currently, treatment of ECAC is dependent upon severity of the disease, comorbid conditions, patient preferences, and suitability for surgery. Optimization of medical therapy

for concomitant pulmonary conditions must be incorporated into the management of ECAC. For appropriate patients with severe ECAC, definitive surgical repair with TBP can be markedly beneficial. Robotic TBP or bronchoscopic laser therapy may prove to be less invasive but equally efficacious interventions for severe ECAC compared with open TBP, but further studies are needed to assess the safety, efficacy, and durability of these interventions (1,5,8).

CPAP has been shown to improve dyspnea, cough, and secretion management in selected patients with TBM (7). The amount of positive pressure necessary to maintain airway patency is likely varied among individuals. Airway pressure titration during dynamic bronchoscopy is currently suggested as an appropriate method to identify needed positive pressure application. (2,9). In select patients who are not surgical candidates, CPAP may be a feasible alternative for long-term management or adjunctive to TBP for those with residual symptoms (10). Nonetheless, the long-term efficacy of NIPPV has not been unequivocally established (2,7,9,10). The mechanism behind NIPPV is pneumatic stenting, which is defined by increased transmural pressure in the central airway due to increased intraluminal pressure on the trachea from NIPPV, expanding the cross-sectional area of the airway lumen (11). CPAP titration can be performed during the assessment of collapsibility during dynamic bronchoscopy. After segmental severity is assessed and the overall ECAC severity score determined, the CPAP mask is attached to the bronchoscope and the patient is instructed to perform manoeuvres to evaluate airway collapse on tidal respiration and forced expiration. In this case report, CPAP pressure was initiated at 6 cmH<sub>2</sub>O and titrated up to 2 cm until collapsibility improved. A pressure of 8–10 cmH<sub>2</sub>O stabilized the airway with tidal breathing and dynamic exhalation.

The patient's journal provided invaluable insight into the impact of CPAP therapy on her symptoms, functional capabilities, and physiological status. The subjective nature of her symptom recording is acknowledged as a limitation. However, her detailed analysis of her clinical course forms the basis for further exploration of the therapeutic approach in a more systematic format.

Literature review of non-invasive management for severe ECAC consists primarily of case reports of patients treated with NIPPV or high-flow nasal oxygen therapy (HFNOT). In 2023, a case report of severe ECAC by Barrios-Ruiz, et al., showed a resolution of cough and decreased exercise-related perceived exertion with the use of CPAP during a cardiopulmonary exercise test (12). Olley et al., reported a case of EDAC managed with pneumatic stenting with nocturnal CPAP which significantly improved quality and duration of sleep, daytime symptoms and quality of life. (13) Park et al. reported a case of severe EDAC managed with HFNOT as a measure of applying intermittent positive end expiratory pressure to successfully overcome the collapse of the airway, improving CO<sub>2</sub> retention and expectoration (14). There is an ongoing clinical trial studying the effect of CPAP on 6-minute walk test outcomes in patients with ECAC that could provide further details into the physiologic effects of pneumatic stenting (15). Most of the studies regarding CPAP titration and airway collapse were done in the setting of OSA. There are no current guidelines that describe how to perform CPAP titration and we envision this report as a contribution to further elaborate a systematic approach to execute this procedure.

ECAC is an underdiagnosed disease that commonly decreases functional capacity and quality of life. The management for mild to moderate ECAC includes a noninvasive approach with maximization of pulmonary therapy, secretions clearing, and NIPPV. For severe cases, therapy involves invasive procedures such as stent trials and TBP. Meanwhile, NIPPV arises as an option for those with severe ECAC especially in those with chronic cough or frequent infections phenotype, and are not candidates for surgical TBP. However, due to the lack of well-designed large-scale trials, the benefits or NIPPV for patients with severe ECAC are uncertain and require further rigorous investigation.

#### AUTHOR CONTRIBUTIONS

RFF was responsible for writing the main draft, patient data collection, literature review, video and figures editing; ABR was involved in manuscript writing; AYLM revised the manuscript; MMJ was responsible for manuscript editing; SFB was responsible for manuscript review and scientific checking, and final approval, DAT was responsible for conceptualization, manuscript review, scientific checking and final approval.

#### CONFLICT OF INTEREST STATEMENT

None declared.

#### DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

#### ETHICS STATEMENT

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

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