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## Case Report

## Pneumatic stenting in the surgical candidacy assessment for cough variant expiratory central airway collapse

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## ABSTRACT

Expiratory Central Airway Collapse (ECAC) is a multifactorial, underdiagnosed entity that poses unique challenges. Airway stenting is used as a predictor for successful outcomes after central airway stabilization surgery via tracheobronchoplasty (TBP). This approach may pose suboptimal performance in certain ECAC variants.

We hypothesize that Continuous Positive Airway Pressure (CPAP), used as a pneumatic stent, could be a non-invasive alternative to evaluate surgical candidacy in cough-predominant ECAC presentations.

We report on a 67-year-old female with a history of chronic cough and confirmed ECAC. After optimization of medical therapy without significant relief and unsuccessful stent trial. We opted to perform CPAP trial during exercise, the patient exercised on a treadmill, and CPAP was applied at two levels (9 cmH<sub>2</sub>O, 11 cmH<sub>2</sub>O). The use of CPAP was associated with resolution of cough and a decrease in exercise-related perceived exertion.

Applying CPAP during exercise may be a promising alternative to stent trials to determine patients' candidacy for surgical management of cough-predominant ECAC.

## 1. Introduction

Expiratory central airway collapse (ECAC), characterized by excessive narrowing of the airway lumen during expiration, is a significant cause of functional impairment and decreased health-related quality of life (HRQOL) [1]. This tracheal or main bronchi obstruction could precipitate chronic cough, dyspnea, wheezing, increased sputum production, and/or recurrent respiratory infections. The complexity in determining the true prevalence of ECAC relies on the absence of standardized guidelines, unreported differentiation within the disease spectrum, and overlapping comorbidities.

Therapeutic options are limited to pharmacologic treatment and definite surgical central airway stabilization through tracheobronchoplasty (TBP), the outcome(s) of which are variable. Temporary airway stenting is the preferred alternative to estimate the potential benefit of TBP. However, this method can yield unreliable data for cough ECAC variant as airway stents may exacerbate cough. Noninvasive positive pressure ventilation may pose as a promising alternative to determine surgical candidacy for cough-

*Abbreviations:* BiPAP, bilevel positive airway pressure; CPAP, continuous positive airway pressure; ECAC, expiratory central airway collapse; EDAC, expiratory dynamic airway collapse; HRQOL, health-related quality of life; PEEP, positive end-expiratory pressure; TBM, tracheobronchomalacia; TBP, tracheobronchoplasty.

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predominant ECAC [2]. Continuous positive airway pressure (CPAP) provides pneumatic stenting of the central airway and maintains a fixed positive transmural pressure through the breathing cycle preventing its collapse [3].

Herein, we report on a patient with chronic cough and confirmed ECAC in whom application of CPAP during exercise led to a reduction in self-reported symptoms and subsequently, referral for TBP.

## 2. Case description

A 67-year-old woman with a BMI of 31.8 kg m<sup>2</sup> presented to our outpatient clinic with productive refractory cough (worse on exertion), wheezing, and shortness of breath. Her past medical history was significant for asthma, gastroesophageal reflux disease, obstructive sleep apnea on CPAP, and recurrent pulmonary infections.

Dynamic computed tomography suggested severe expiratory collapse of the trachea, mainstem bronchi, and bronchus intermedius. Complementary findings with dynamic bronchoscopy showed collapsibility of 70% at the cricoid, 90% at mid trachea, 80% at distal trachea, 90% at right mainstem bronchus, 100% at bronchus intermedius, and 90% at left mainstem bronchus. These findings are consistent with severe ECAC in the subtype of EDAC, as portrayed in Fig. 1. Subsequently, a stent trial was attempted to assess surgical candidacy; however, it was not feasible due to complex anatomy consisting of a short epiglottis and anterior larynx.

Given the persistence of symptoms and quality of life impairment, an alternate method for surgical candidacy evaluation was pursued before committing the patient to TBP. We opted for pneumatic stenting of the airways during exercise.

We had the patient perform submaximal treadmill exercise (1.6 mph, 16% gradient) while CPAP was applied at 0 cmH<sub>2</sub>O (control), 9 cm H<sub>2</sub>O, and 11 cmH<sub>2</sub>O; each CPAP level was applied for 3 min (Fig. 2). At rest, the patient rated her 'exertion' (RPE) as 9 on the Borg 6–20 scale. Following the first 3 minutes of exercise at 0 cmH<sub>2</sub>O, the patient's RPE increased to 13. The exercise was also associated with worsening of the patient's cough. When applied at 9 cmH<sub>2</sub>O, the use of CPAP was associated with a decrease in RPE back to the baseline value and complete resolution of the patient's cough was achieved. Further titration of CPAP to 11 cmH<sub>2</sub>O was associated with a slight increase in RPE, but this could also reflect the duration of exercise and upward drift in heart rate rather than the lack of efficacy of the CPAP itself (Table 1). Overall, the patient and our team noticed a significant reduction in cough and shortness of breath when CPAP was applied, which led to the multidisciplinary joint decision to undergo surgical tracheal stabilization. TBP was performed without complications and resulted in an improvement of respiratory symptoms. Following this multidisciplinary management, the patient reported no recurrence of symptoms at a one-year follow-up.

## 3. Discussion

ECAC encompasses two main entities: 1) excessive dynamic airway collapse (EDAC) and 2) tracheobronchomalacia (TBM). Airway collapse in ECAC develops when extraluminal pressure exceeds intraluminal pressure, comparable to *physiologic dynamic intrathoracic tracheal collapse*, which increases with force expiration and cough. Despite having a similar clinical presentation, the two entities of ECAC have different mechanisms. EDAC is the anterior bulging of the tracheal wall due to the destruction of smooth muscle fibers that form the posterior wall of the tracheobronchial tree. By comparison, in TBM, collapsibility extends beyond the mainstream bronchi and results from the destruction of the anterolateral cartilaginous support of the trachea [4].

It has proven challenging to define clear thresholds for the identification of ECAC. Studies reporting an abnormal airway collapse of 50–60% have shown an increased risk of false positives, overlapping with normal individuals [2,5]. Thus, a pragmatic criterion of mild (70%–79%), moderate (80%–89%), and severe (> 90%) has been described and implemented in this case [6]. Further literature with an overall collapsibility assessment is needed.

While ECAC pathophysiology remains unclear, prevailing hypotheses rely on the changes in airway resistance, loss of elastic recoil, and two central mechanisms: a) equal pressure point (EPP) and b) wave speed theory. EPP is present at forced expiration when alveolar pressure parallels intrapleural pressure. In the setting of ECAC, displacement of EPP with increased resistance farthest from the EPP point develops collapsibility in the airway with lower intrapleural pressures. Wave speed theory describes a choke point when airflow and wave dispersion velocity match [7,8]. Furthermore, this decrease in cross-sectional area impairs mucociliary clearance, perpetuating the chronic inflammatory response [8].

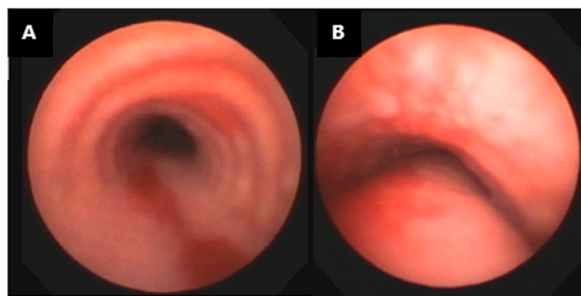


Fig. 1. Bulging of posterior membrane of tracheal wall into the airway lumen. A, inspiratory view of the proximal trachea; B, expiratory view of the proximal trachea collapse.



Fig. 2. Clinical and Cardiopulmonary Lab set up depicting motorized treadmill and rating of perceived exertion (RPE) scale used in this case study.

Table 1

Patient response during CPAP protocol.

	CPAP			
	Rest	0 cmH <sub>2</sub> O	9 cmH <sub>2</sub> O	11 cmH <sub>2</sub> O
Heart rate, bpm	69	132	139	160
BP, mmHg	130/85	136/78	167/89	168/84
SpO <sub>2</sub> , %	96	92	91	90
RPE (Borg 6–20 scale)	9	13	9	12

BP, blood pressure; RPE, rating of perceived exertion.

Treatment options for severe ECAC stages comprise conservative ventilation methods (CPAP, BiPAP, PEEP), airway clearance techniques, and invasive airway stabilization procedures, including stent placement and TBP. The role of positive airway pressure methods in the diagnostic phase has not been explored.

CPAP overcomes airway collapse throughout the respiratory cycle by increasing alveolar pressure above atmospheric pressure. On the contrary, short-term stenting is the preferred predictor for potential improvement after TBP due to its immediate central airway stabilization [9]. Significant objective improvement following stent trial has been reported in 65–75% of patients [10,11]. However, some patients may be unsuitable for this invasive trial and present related complications such as granulation tissue formation, intractable cough, valve migration, mucus plugs, or impaction, that could lead to the narrowing of the lumen or worsening symptoms, especially in cough phenotypes.

Exercise, as an ECAC exacerbation factor, has been previously reported in the literature; even so, the discussion has focused more on the diagnostic approach than on the management [3,7,8]. Since it was also a precipitating condition for cough in our patient, we decided to perform the CPAP pneumatic stent test trial during exercise.

We acknowledge that the main limitation of our study was the absence of quantitative data for symptomatic assessment during pneumatic stenting. Nonetheless, there is no standardized evaluation to assess cough severity during exercise. The challenges faced by certain phenotypes of ECAC could be overcome with accessible and less invasive procedures when a stent trial is not feasible, ensuring

timely management and reducing the risks of invasive procedures without compromising diagnostic value. Further research in larger cohorts is needed to assess its hypothesized overestimation [9]. Development of a systematic approach depending on patient phenotype can effectively enhance patient compliance, decrease diagnostic delays, and impact patient QOL.

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### Abstract presentation

Part of this data will be presented as a poster at the 7th European Congress for Bronchology and Interventional Pulmonology 2023 meeting in Madrid, Spain. With the title of: Pneumatic stenting pilot for surgical management assessment in expiratory central airway collapse.

### Author contributions

A.B.R.: Writing- Original draft preparation. A.Y.L.M.: Writing- Reviewing and Editing. B.N.H.: Investigation. K.L.W.: Visualization. S.F.B.: Conceptualization. V.A.:Supervising. B.C.: Supervising. B.J.T.: Investigation, Writing- Reviewing and Editing. D.A.T.:Writing- Reviewing and Editing.

### Declaration of competing interest

The authors have no relevant financial or other relationships to disclose.

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