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Case Report

Successful step-by-step diagnosis and management of expiratory central airway collapse

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ABSTRACT

A 45-year-old woman with recurrent dyspnea for 40 years was previously diagnosed with bronchial asthma and spasmodic dysphonia. On admission, the patient was diagnosed with expiratory central airway collapse (ECAC) due to expiratory dynamic airway collapse based on radiographic examination, chest computed tomography, and bronchoscopy. After continuous positive airway pressure and temporal airway stenting, surgical tracheobronchoplasty and tracheal membranous portion reinforcement using polypropylene mesh successfully relieved the respiratory symptoms. In patients with airway obstructive disease refractory to conventional therapies, ECAC should be considered.

1. Introduction

Expiratory central airway collapse (ECAC) includes tracheobronchomalacia (TBM) and expiratory dynamic airway collapse (EDAC) [1,2]. TBM, which is caused by cartilage weakness, affects the anterolateral portion of the tracheobronchial tree. EDAC is an excessive inward bulging of the tracheobronchial posterior membrane during expiration, caused by weakness of the longitudinal muscle fibers. Although the etiology of TBM and EDAC is not fully understood, congenital disorders, external pressure, and inflammation may possibly lead to airway wall weakness, predisposing the trachea to collapse.

Associations between ECAC and high body mass index (BMI) [3,4], tobacco use [5], gastroesophageal reflux [6], and inhaled corticosteroid (ICS) [7] use have been reported.

The threshold level of ECAC diagnosis is sometimes controversial. At least 70% of expiratory collapse on dynamic computed tomography (CT) might be an appropriate threshold level for considering ECAC to avoid false negatives. However, the criteria of 50% airway collapse in the horizontal cross-sectional area of the airway at expiration or during coughing, has been widely used [8]. CT does not replace the gold standard of bronchoscopy. When ECAC diagnosis is made, continuous positive airway pressure (CPAP), stenting, and tracheobronchoplasty (TBP) are candidate treatments [9].

Owing to expiratory flow obstruction, patients can present with non-specific symptoms, such as cough, dyspnea, wheezing, secretion retention, and recurrent respiratory infections, which can contribute to common misdiagnoses. Moreover, sudden inability to ventilate could lead to life-threatening hypoxemia.

We encountered a patient with ECAC due to EDAC whose symptoms were similar to those of bronchial asthma and spasmodic dysphonia. In this case, eventual diagnosis of ECAC allowed for successful surgical management and resolution of symptoms.

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2. Case presentation

A 45-year-old woman presented with fever, sore throat, stridor, cough, dyspnea, and speech disturbance. The patient reported a history of similar episodes since childhood, with symptoms such as hoarse voice; barking cough with stridor; impaired exercise tolerance; and dyspnea provoked by coughing and forced expiration. There was a tendency of SpO2 level to decrease. The patient was initially diagnosed with bronchial asthma and treated with ICS. Temporary difficulty in speaking led to a tentative diagnosis of spasmodic dysphonia. The symptoms and episodes resulted in deteriorated quality of life over time, while diagnosis remained unclear. A history of smoking for 17 years and predisposition to obesity with a BMI of 30.8 was noted.

Upon admission, laryngoscopy revealed right-dominant, swollen tonsils with a white coating. No subglottic stenosis or laryngeal edema was noted. Empiric antibiotics and steroids were administered intravenously, with the presumption of a diagnosis of suppurative tonsillitis. Although the inflammation improved, the patient reported stridor and dyspnea while walking down the hospital hallway. Expiratory dominant stridor was obvious in the cervical area, although the lungs were relatively clear to auscultation. Thus, considering other differential diagnoses was warranted.

Chest radiography revealed narrowing of the bronchi on exhalation (Fig. 1), which was further confirmed by CT at the end of forced expiration/inspiration, revealing a collapsed trachea, leading to a diagnosis of ECAC. Virtual endoscopic images were generated from the CT data (Fig. 2A and B), showing protrusion of the membranous portion of the trachea at exhalation. The patient was discharged once the condition stabilized. However, soon after, the patient experienced rapid recurrence of dyspnea with a gradually deteriorating condition.

The patient was referred to a university hospital, where a diagnosis of hypoxic respiratory failure was made, and admitted to the intensive care unit. Upon admission to the intensive care unit, clinical impression was airway collapse combined with bronchial asthma. Thus, we administered steroids and applied CPAP. This management successfully stabilized the patient's condition that was unclear whether ECAC alone led to it. Bronchoscopy was first performed immediately before rigid bronchoscopy followed by stent insertion. The bronchoscopic procedure was performed under mild sedation at the end of forced expiration/inspiration immediately before general anesthesia induction. Bronchoscopy revealed protrusion of the membranous portion of the central airways into the tracheal lumen (Fig. 2C and D). Reduction in the cross-sectional area of the trachea at the aortic arch level measured by repeated CT scanning at expiration was 64% compared with that at inspiration, supporting ECAC diagnosis. Although CPAP was effective in initial stabilization, it did not provide sufficient therapeutic effects in the patient to be discharged or improve quality of life. TBP was also considered as a treatment option. To estimate the efficacy of this surgical procedure, a dynamic Y tracheobronchial stent was placed (Fig. 3), following an algorithm commonly used to manage ECAC [2]. The Y-stent significantly improved the patient's symptoms. However, the stent was not tolerated because of irritation and removed 5 days later. Two weeks later, the patient underwent successful TBP.

The approach was a standard posterolateral thoracotomy through approximately 20 cm skin incision. A long spiral single-lumen tube was used for ventilation and moved back and forth depending on surgical steps to avoid cuff injury and confirm surgical safety. The endotracheal tube was first placed in the upper trachea, where bronchial observation confirmed significant airway collapse by exerting negative pressure via suction. Then the tube was advanced to the left main bronchus to collapse the right lung. Once the right main bronchus was dissected and encircled, an aortic clamp was applied to occlude the right bronchus for later procedures. After the membranous portion of the trachea was fixed, the endotracheal tube was moved back to the upper trachea to confirm surgical efficacy. After fixation of the trachea, airway collapse did not occur by applying negative pressure at all. While keeping the endotracheal

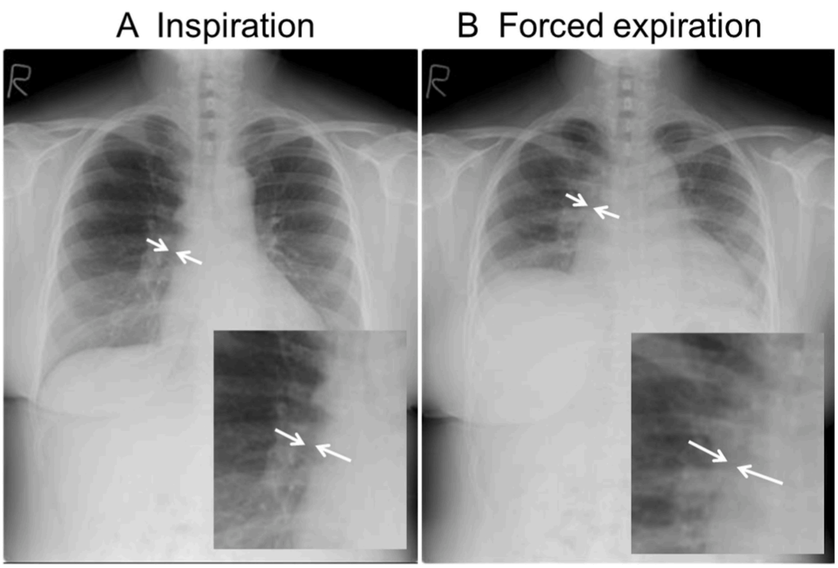

Fig. 1. Chest X-ray at end-inspiratory (A) and forced expiratory phases (B). Narrowing of the bronchi is evident at the forced expiratory phase (arrows).

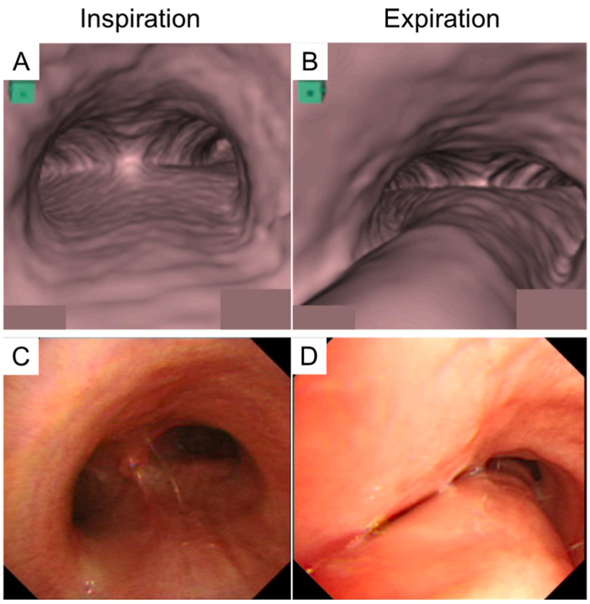

Fig. 2. Virtual endoscopic images generated by CT data at end-inspiratory (A) and forced expiratory phases (B). Virtual endoscopic image created by CT data showing intrusion of the tracheobronchial posterior membrane during expiration, attributed to the weakness of the longitudinal muscle fibers (B). Bronchoscopy (C) (D) reveals airway collapse at expiration, affecting the trachea and opening of the bronchi. The degree of narrowing in the anteroposterior diameter is apparent, compared to that of luminal narrowing.

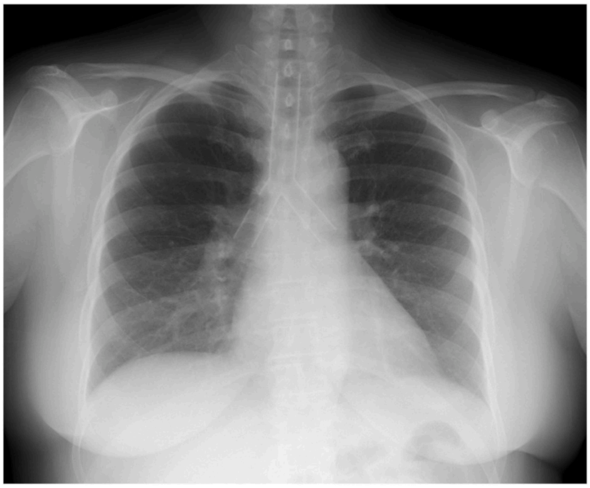

Fig. 3. Chest X-ray taken after placing a silicone Y stent under rigid bronchoscopy. As expected, stenting improves the patient's airway symptoms initially, suggesting potential benefit from surgical fixation. However, the patient cannot tolerate the irritation cause by the stent.

tube kept in the middle trachea, the left main bronchus was fixed. Then the endotracheal tube was advanced to the left main bronchus. Additionally, the aortic clamp on the right bronchus was removed to fix the right main bronchus and bronchus intermedius. An appropriately trimmed polypropylene mesh was sutured with polydioxanone sutures (Johnson and Johnson, New Brunswick, NJ, USA), to the membranous portion of the airway. This fixation was from the upper trachea through the carina, down to the left main bronchus close to the secondary carina, as well as down the right main bronchus and bronchus intermedius (Fig. 4). Stitches were placed to plicate the membranous portion. A total of 64 stitches were placed to complete the procedure that lasted for 453 min without complications. On CT imaging, the airways at segmental levels appeared intact. Otherwise, surgical indication would have been doubtful.

Recovery after surgery was uneventful, with a significant improvement in symptoms. The patient was discharged 16 days after surgery. Six months later, CT demonstrated good outcomes of surgery (Fig. 5). The patient reported resolution of symptoms. The recovery time from exertion (for example, upon exercise) was significantly reduced after TBP.

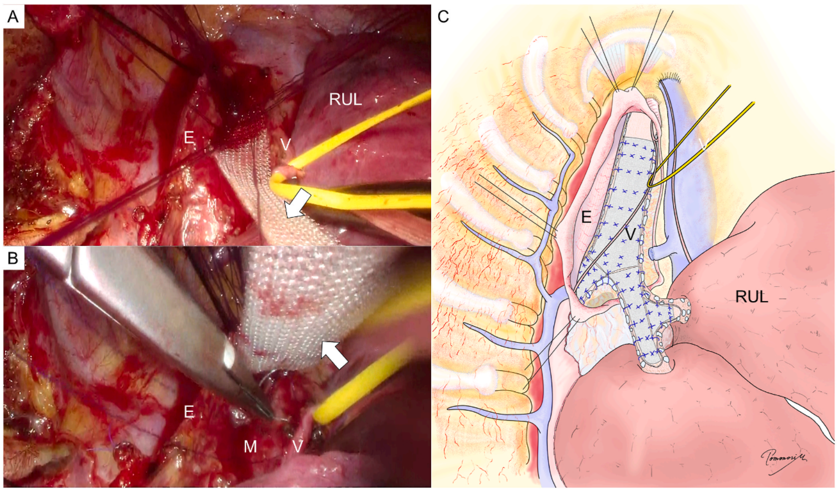

Fig. 4. Intraoperative findings. (A, B) Photos showing a rigid polypropylene mesh (arrows) sutured onto the membranous portion of the trachea. (C) Illustration showing the polypropylene mesh splinting the tracheobronchial tree. E, esophagus; M, membranous portion; V, Vagus nerve; RUL, right upper lobe of the lung.

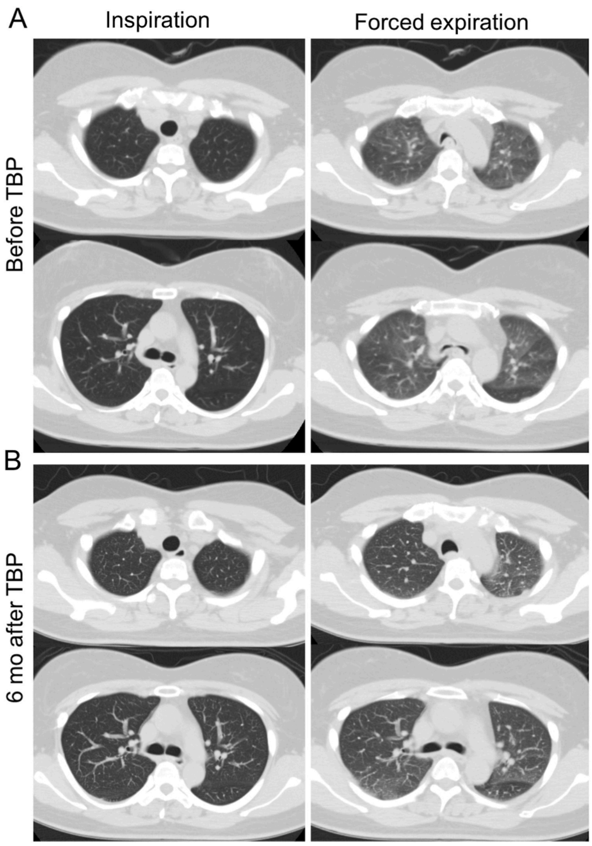

Fig. 5. Comparison of CT images at end-inspiratory and forced expiratory phases before tracheobronchoplasty (TBP) (A) and 6 months after TBP (B) at the level of the aortic arch and at the level caudal to the carina. Protrusion of the posterior membranous portion of the central airway and resulting airway collapse is evident at expiration before TBP. However, these findings are significantly improved after TBP.

3. Discussion

A large number of conditions can obstruct the airway both reversibly and irreversibly. Even if patient presents with clinical features and laboratory findings of ECAC, ECAC can be commonly misdiagnosed as other upper airway disorders and lower airway obstructive diseases that may mimic the symptoms of ECAC.

Spasmodic dysphonia is a task-specific voice dysfluency resulting from intrinsic laryngeal musculature hyperfunction exhibiting uncontrolled voice breaks [10]. In our patient, these voice breaks occurred once every few sentences, which led to a diagnosis of spasmodic dysphonia. However, this diagnosis did not explain her cough and stridor during exercise. Inducible laryngeal obstruction, presenting as recurrent variable airflow obstruction in the larynx, should also be considered in patients with sudden onset of breathlessness and wheezing on inspiration [11].

Common mimickers of ECAC in adult lower airway diseases are bronchial asthma [12] and chronic obstructive pulmonary disease (COPD) [13]. Bronchial asthma is a variable expiratory airflow limitation with wheezing, dyspnea, and cough, which may vary over time. In this case, speech disturbance was inconsistent with the diagnosis of bronchial asthma. COPD, often associated with ECAC, is characterized by dyspnea, cough, excessive sputum production, and a smoking history. Bronchial asthma and COPD are syndromes comprising several endotypes and phenotypes. Furthermore, mixed ECAC phenotypes should also be considered in the diagnostic procedure.

The diagnostic processes for ECAC tend to be so complex and misleading. Therefore, inclusion of ECAC in the list of differential diagnosis and radiographic diagnostic procedures are important. Since a wide range of functional airway collapses have been observed among healthy individuals [14], clear cut-off value between physiological and pathological narrowing is yet to be determined. This makes determining the clinical significance of ECAC difficult. To make diagnosis of ECAC, not only the CT images but also the clinical symptoms that are not fully explained by other airway diseases are important.

Diagnostic images in each case may differ according to the affected portion and severity of airway collapse. In our case, tracheal narrowing in the frontal view of the aerogram during expiration was not detected because of inconspicuous change in the lateral diameter of the trachea. Although bronchial narrowing was noted (Fig. 1), indicating the importance of careful radiographic examination as an initial step in diagnosis, it does not replace paired inspiratory/expiratory CT analysis or fiberscopy [15] in terms of precise assessment of airway collapse necessary for diagnosing TBM or EDAC.

Noninvasive virtual endoscopic assessment generated using CT data is helpful in indirect visualization of the airway (Fig. 2 A, B).

Once the diagnosis of ECAC is made, therapeutic strategies need to be established in a stepwise manner. Although the pathophysiology and morphology of TBM and EDAC are distinct, the current evidence shows that the therapeutic work-ups are quite similar [9].

Current evidence proposes using initial noninvasive therapeutic workups such as CPAP or noninvasive ventilation to estimate the benefit of supporting airway patency [16]. Once the benefit is suggested, stents can be placed to maintain airway patency [17]. However, since airway stents can cause multiple problems including airway irritation, infection, and mucus plugging, the role as a solution of ECAC is more limited to those who are not good candidates for more invasive surgical procedures. In a relatively fit patient, similar to our case, airway stenting plays a more important role in a patient who benefits from surgical splinting of the central airways or TBP. Our patient was found to be a good surgery candidate by temporal placing of a Y-stent (Fig. 3), and underwent successful TBP (Fig. 4). As demonstrated in a large cohort study, TBP was shown to be the most possibly reliable therapeutic strategy for ECAC [18,19].

When evaluated by the FEMOS classification [20], an acronym for the words 'function', 'extent of abnormality', 'morphology', 'origin of disease' and 'severity of airway collapse', the etiology and morphology were idiopathic and EDAC with crescent shaped, respectively. This revealed a functional status, F3; extent of disease, E4; and severity, S4. Although the degree of collapse on CT images was not severe, we failed to maintain the patient's condition by CPAP and stenting. We prioritized symptoms relief. We suggested that although images captured in the setting of the end of forced expiration or inspiration on CT could estimate the degree of severity, they did not reflect or correlate with the patient's daily symptoms. Subsequently, TBP led to improving not only CT findings, but also the patient's quality of life. This suggests that symptom-oriented decision on the therapeutic strategy might have contributed to the good results.

In this case, starting from complex processes until reaching ECAC diagnosis, a series of therapeutic trials including noninvasive positive-pressure ventilation, stenting, and TBP, led to successful step-by-step case management and eventual symptomatic relief.

4. Conclusion

In patients with a deviant clinical course of airway obstructive disease refractory to conventional therapies, ECAC should be considered. In diffuse, moderate-to-severe symptomatic ECAC, TBP should be considered as a definitive therapy for maintaining airway patency.

Informed consent

Written informed consent was obtained from the patient for the publication of this case report.

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Declaration of compting interest

The authors report no conflicts of interest.

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None.

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