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Letter to the Editor

Early diagnosis and management of tracheomalacia with invasive bronchopulmonary aspergillosis in an adult

Dear Editor,

A 59-year-old male patient had suffered from chronic obstructive pulmonary disease (COPD) for over 5 years, but never attempted to obtain formal diagnosis and treatment. He reported a history of repeated medication with intravenous and oral corticosteroids. He was admitted to our Respiratory Intensive Care Unit (RICU) due to severe breathing difficulties. He presented fever (body temperature of 38.1°C), bloody sputum, respiratory distress, and difficulty in lying down at night. The follow-up chest computerized tomography (CT) showed a few exudation changes in two segments in the inferior area (Fig. 1A), FEV1/FEV of 40% and a WBC of $12.8 \times 10^9/L$. After admission, he was given antibiotics, inhalation for asthma, combined with bilevel positive airway pressure (BIPAP) and oxygen therapy. Five days after the medication was started, the patient had not clearly improved, so a bronchoscopy was performed and found that the tracheal cartilage rings had disappeared and forced expiratory airway collapsed significantly (Fig. 1B). There was white mural necrotic tissue on the ventricular wall of the basal segments in the lower left lung, easily bleeding upon instrument touch (Fig. 1C). A basal mucosal biopsy of necrotic tissue was performed, pathologically confirming pulmonary *Aspergillus* infection (Fig. 1D). The patient was then diagnosed with tracheomalacia complicated with invasive bronchopulmonary aspergillosis (IBPA), and had the medication immediately adjusted, starting with intravenous voriconazole 200 mg bid (doubling the first dose) for 21 days, and switching to the oral formulation for 28 days, with combination of continuous positive airway pressure (CPAP), plus intravenous nutrition and methylprednisolone 40 mg IV q12h. The temperature returned to normal on day five; symptoms of cough, expectoration and dyspnea improved on day six. The patient was discharged due to the substantial improvement after 28 days of hospitalization.

Although diagnostic techniques and criteria for tracheomalacia are not standardized, thorax CT and bronchoscopy are the preferred methods in all published studies. Asymptomatic patients should be closely observed without treatment, and there is no uniform standard treatment

for severe patients. Generally speaking, the patients would be treated on individual basis. Internal physicians usually prescribe glucocorticoid and immunosuppressive agents as a conservative treatment that has some effects. CPAP¹ shows a better therapeutic effect by increasing tidal volume and lessening airway collapse. Surgical treatments, including stent implantation,² airway reconstruction³ (e.g. tracheostomy), and relieving airway obstruction⁴ (e.g. aortic fixation) show some therapeutic effects, but should be carefully considered on an individual basis.

To our knowledge, the reports of tracheomalacia complicated with IBPA in adult patients are rare. We suggest that the probable reasons may be: 1) long-term glucocorticoid therapy; 2) treatment with immunosuppressive agents; 3) repeated use of antibiotics. IBPA patients show no specific signs in CT imaging,⁵ and the symptoms of respiratory tract infections – such as cough, expectoration and fever – lack characteristic features and are easily concealed by the symptoms of underlying diseases. Thus, the diagnosis of IBPA, especially when complicated with adult tracheomalacia, is difficult and often leads to misdiagnosis and delayed treatment, increasing medical costs and clinical risk. Early diagnosis of IBPA should be based on clinical features and laboratory examinations. Our understanding is that when patients have risk factors for fungal infection, especially aspergillosis, respiratory symptoms that do not match imaging findings, and antibiotics (anti-bacterial) do not work well, performing bronchoscopy immediately is critical for correct diagnosis and successful treatment of IBPA, as well as to enhance the awareness and recognition of IBPA by physicians.

In conclusion, IBPA is a very difficult and dangerous disease, especially for adult tracheomalacia patients, who have high mortality. Critical management to achieve a successful outcome in such cases involves quick diagnosis and early treatment. Physicians should closely observe patients' clinical features, carefully investigate their respiratory symptoms, and identify bronchopulmonary aspergillosis by BLAF and bronchial mucosal biopsy. Voriconazole⁶ combined with other treatments such as CPAP, corticosteroids, nutritional support and methylprednisolone, are very effective in alleviating the disease.

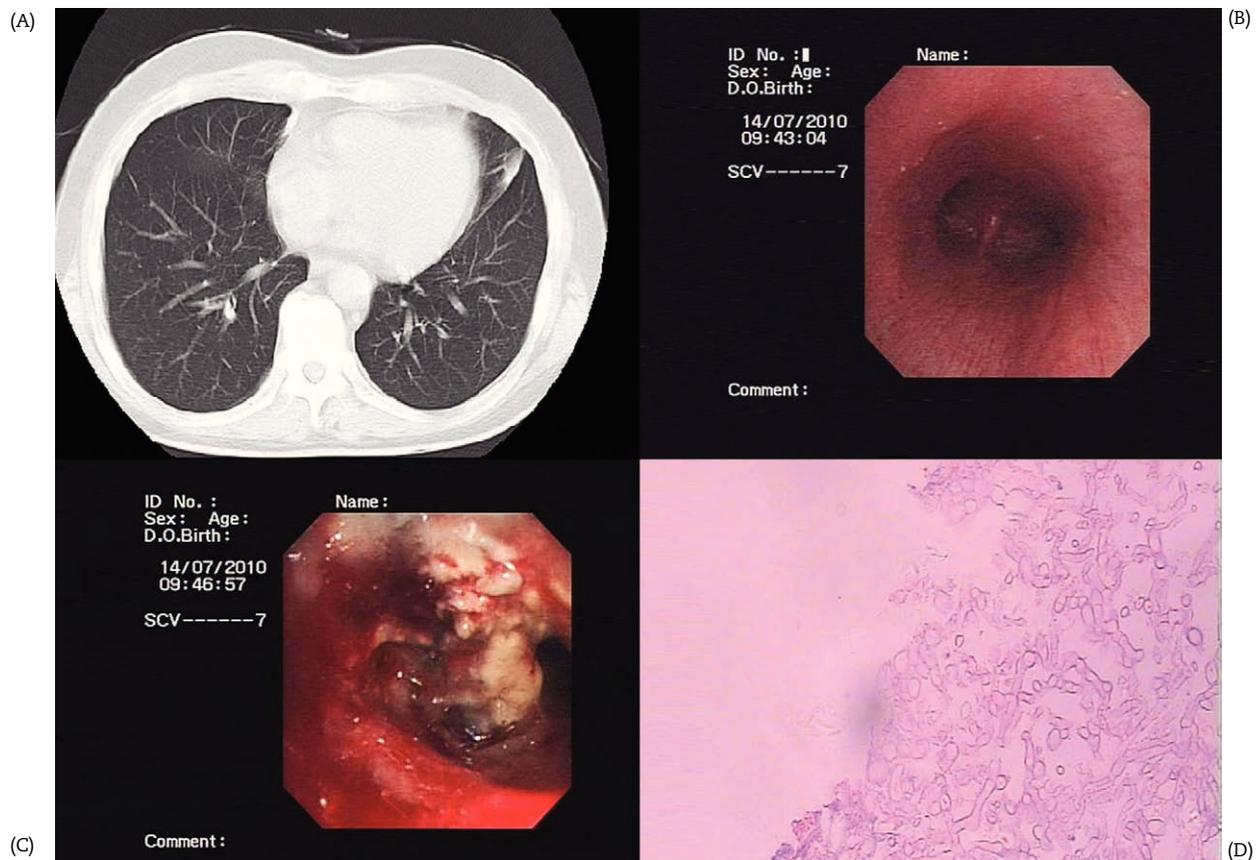


Fig. 1 - Early diagnosis and management of tracheomalacia with invasive bronchopulmonary aspergillosis in an adult. (A) CT showed few exudation changes in two segments in inferior area. (B) Bronchoscopy showed that tracheal cartilage rings disappeared and forced expiratory airway collapsed significantly. (C) Bronchoscopy showed white mural necrotic tissue on the ventricular wall of the basal segments in the lower left lung, easily bleeding upon instrument touch. (D) Pathologically confirmed pulmonary *Aspergillus* infection.

Conflict of interest

All authors declare to have no conflict of interest.

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