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Case report

Three-dimensional imaging forced oscillation technique to assess position-dependent airway obstruction in relapsing polychondritis: A case report

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ABSTRACT

Relapsing polychondritis (RP) is characterized by recurrent systemic inflammation of the cartilages and is accompanied by central airway collapse. We report a case wherein three-dimensional imaging of respiratory system resistance (Rrs) and respiratory system reactance (Xrs) by using MostGraph (CHEST M.I., Tokyo, Japan), a forced oscillation system, revealed that Rrs and Xrs in the inspiratory and expiratory phases correlated with proximal airway collapse. The degree of difference in Rrs and Xrs between the supine and sitting positions reflected airway collapse more closely than did the pulmonary function test. MostGraph could be a useful tool for assessing airway collapse in RP.

Abbreviation: 3D, three-dimensional; CT, computed tomography; FEV_1, forced expiratory volume in one second; FOT, forced oscillation technique; FVC, forced vital capacity; IOS, impulse oscillation system; OSAS, obstructive sleep apnea syndrome; PEFR, peak expiratory flow rate; PFT, pulmonary function test; RP, relapsing polychondritis; Rrs, respiratory system resistance; V25, flow rate at 25% of FVC; V50, flow rate at 50% of FVC; Xrs, respiratory system reactance

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1. Introduction

Relapsing polychondritis (RP) is a rare disease characterized by recurrent systemic inflammation of the cartilages and proteoglycan-rich structures, such as the joints, skin, eyes, nose, auricles, central nervous system, trachea, and bronchi [1]. Patients with RP and airway involvement are reported to have a poor prognosis [1].

The forced oscillation technique (FOT) can determine respiratory system impedance, which is a measurable estimate of breathing mechanics. MostGraph (CHEST M.I., Tokyo, Japan) was developed as an FOT that could visualize respiratory system resistance (Rrs) and reactance (Xrs) as three-dimensional (3D) color images with time and signal frequencies. The obtained pattern can be useful for differential diagnosis; pathophysiologic analysis; and assessment of severity and therapeutic effects in airway diseases, especially in chronic obstructive pulmonary disease and asthma [2–4]. In addition, unlike the pulmonary function test (PFT), MostGraph allows for measurements at rest respiration. Further, the obtained results can be analyzed separately for expiration and inspiration. Compared with the impulse oscillation system (IOS) (MasterScreen IOS; Jaeger, Hoechberg, Germany), MostGraph produces a 3D color image that allows patients to interpret the results more easily.

Airway collapse in RP can be clinically assessed by bronchoscopy and chest computed tomography (CT) [5]. However, since these techniques are performed in the supine position only, differences in the degree of airway collapse between the supine and sitting positions cannot be assessed. We hypothesized that MostGraph would enable quantitative evaluation of the status of RP through measurements of the difference in Rrs and Xrs between the supine and sitting positions, and by analyzing these results separately for expiration and inspiration.

2. Case report

A 71-year-old man was referred to our hospital because of fever (37.5 °C), productive cough, and bilateral ear pain that persisted for 2 months. On spirometry, the forced expiratory volume in one second (FEV_1) was 2.47 L, and FEV_1/forced vital capacity (FVC) was 70.0%. Bronchodilators and inhaled corticosteroids were prescribed to treat his condition.

One year later, he was hospitalized for the assessment and treatment of dyspnea in the supine position and rapid deterioration of FEV_1 to 1.35 L and FEV_1/FVC to 38.0%, which suggested airway obstruction. The peak expiratory flow rate (PEFR) was 3.23 L/s and the flow rate at 50% of FVC (V50) per flow rate at 25% of FVC (V25), i.e., (V50/V25) was 1.53. The flow-volume curve showed a sharp decline immediately after the peak, with a long expiratory phase and trembling (Fig. 1a).

Physical examination did not reveal any ear or nose deformity. On auscultation in the supine position during rest respiration, low-pitch rumbles were noted during deep expiration, most prominently around the trachea. Serum chemistry values were within normal ranges. C-reactive protein level and rheumatoid factor were within normal limits, but anti-collagen 2 antibody was positive at 28.0 IU/mL. Arterial partial pressures of oxygen and carbon dioxide at room air were 73.6 mmHg and 43.3 mmHg, respectively. Pathologic examination of a biopsy specimen from an irregularly shaped nasal septum revealed lymphocyte-dominant inflammation on the cartilage surface. A diagnosis of RP was determined on the basis of McAdams' criteria [6], which included nasal chondritis, tracheal chondritis, and elevation of serum anti-collagen 2 antibody level.

CT images were acquired separately during the inspiratory and expiratory phases. Compared with the inspiratory phase, the expiratory phase was characterized by collapsed tracheal wall and bronchi. Bronchoscopy showed that the airway did not collapse during the inspiratory phase; however, during the expiratory phase, the flattening of the ventral tracheobronchial

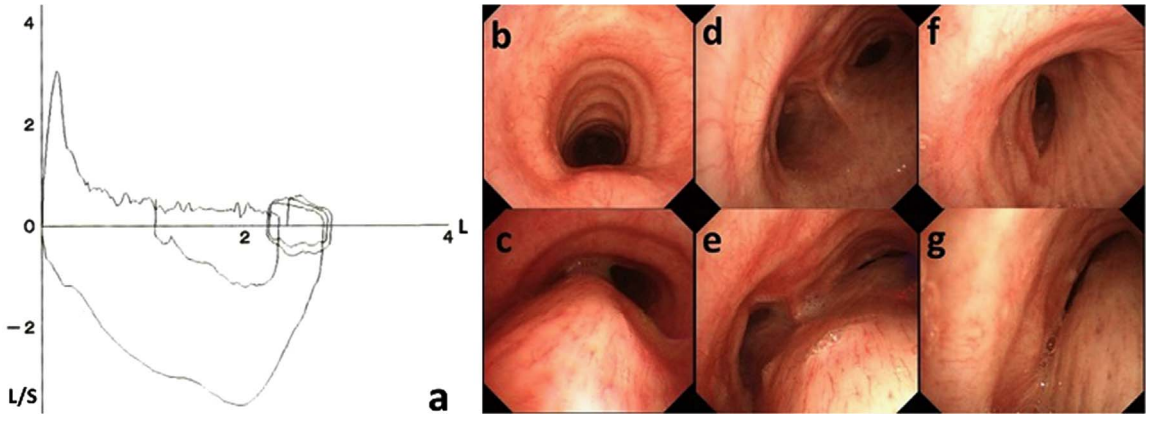

Fig. 1 – (a) The flow-volume curve shows a sharp decline immediately after the peak with a long expiratory phase and trembling. (b)–(g) Bronchoscopy in the supine position. (b), (c) trachea; (d), (e) carina; and (f), (g) right main bronchus. Images (b), (d), and (f) acquired during the inspiratory phase and (c), (e), and (g) acquired during the expiratory phase show luminal narrowing secondary to the flattening of the ventral tracheobronchial cartilages and elevated dorsal membranous portion during the expiratory phase.

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