


# Temporary stenting in a case of bronchomalacia due to relapsing polychondritis

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## Abstract

Airway involvement in relapsing polychondritis (RP) can often be debilitating and life threatening. Interventional procedures such as stenting can be useful to improve airway stenosis. This case was diagnosed with RP with a circumferential obstruction at the left main bronchus. We determined that a silicone airway stent would be placed. The silicone stent was removed after 22 months due to granulation tissue. After stent removal, bronchoscopic findings revealed a collapsing left main bronchus during exhalation, but airway patency was maintained during inhalation without any respiratory symptoms. In this case, bronchomalacia remained after stent removal. However, since there were no severe respiratory symptoms, we decided that stent replacement was unnecessary. In general, it is difficult to remove airway stents in severe tracheobronchomalacia; however, temporary stenting might be a useful procedure in cases with unilateral main bronchial stenosis.

## KEYWORDS

airway stents, relapsing polychondritis, tracheobronchomalacia

## INTRODUCTION

Airway involvement in relapsing polychondritis (RP) is an important predictive factor for prognosis. It has been reported that tracheobronchomalacia (TBM), when caused by RP, can be debilitating and life threatening.<sup>1</sup> In addition, immunosuppressive therapy is often inadequate for airway disorders in RP, and interventional procedures such as airway stenting are often required.<sup>1</sup> Metallic stents were reported to be useful to improve airway stenosis in RP patients.<sup>2</sup> However, metallic stents are not recommended for benign central airway stenosis due to several complications such as respiratory tract infections, granulation tissue and difficulty in expectorating. In this case, temporary stenting with a silicone stent was able to dilate the left main bronchial stenosis on inhalation and her respiratory symptoms improved.

## CASE REPORT

A 50-year-old woman with no smoking history was diagnosed with bronchial asthma 8 years ago. She has been

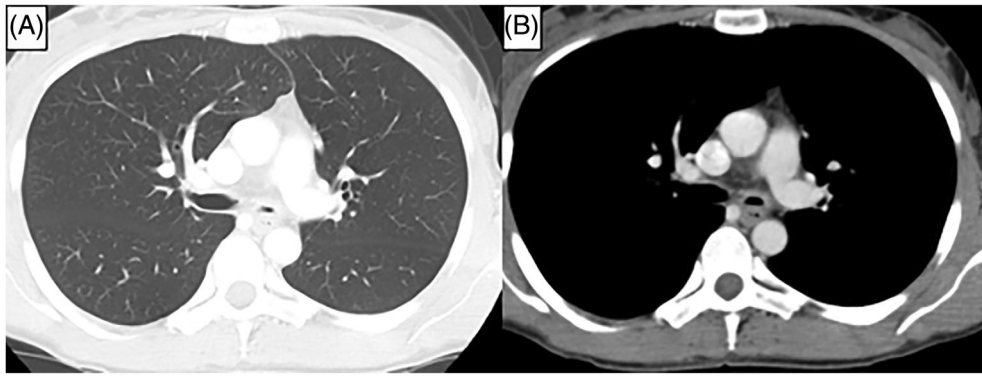
treated repeatedly with oral corticosteroid therapy. Five years earlier, CT findings revealed a narrowing of the left main bronchus with airway wall thickness, and she was referred to our hospital (Figure 1A, B). Bronchoscopic findings revealed a circumferential obstruction at the left main bronchus (Figure 2A, B), and flow-volume curves showed a decreased expiratory flow (Figure 3A). Since she also had a hearing impairment, she satisfied the diagnosis criteria by Damiani and Levine for a diagnosis of RP, which included respiratory tract chondritis, cochlear and vestibular dysfunction, and a positive response to administration of corticosteroid.<sup>3,4</sup>

A Dumon straight stent (10 × 42 mm) was placed at the left main bronchus and airway patency was maintained after placement (Figure 2C, D). After stenting, prednisolone 30 mg per day, and cyclosporine 150 mg per day were administered. One year later, prednisolone was reduced to 10 mg per day and self-administered tocilizumab (162 mg every 2 weeks) was added.

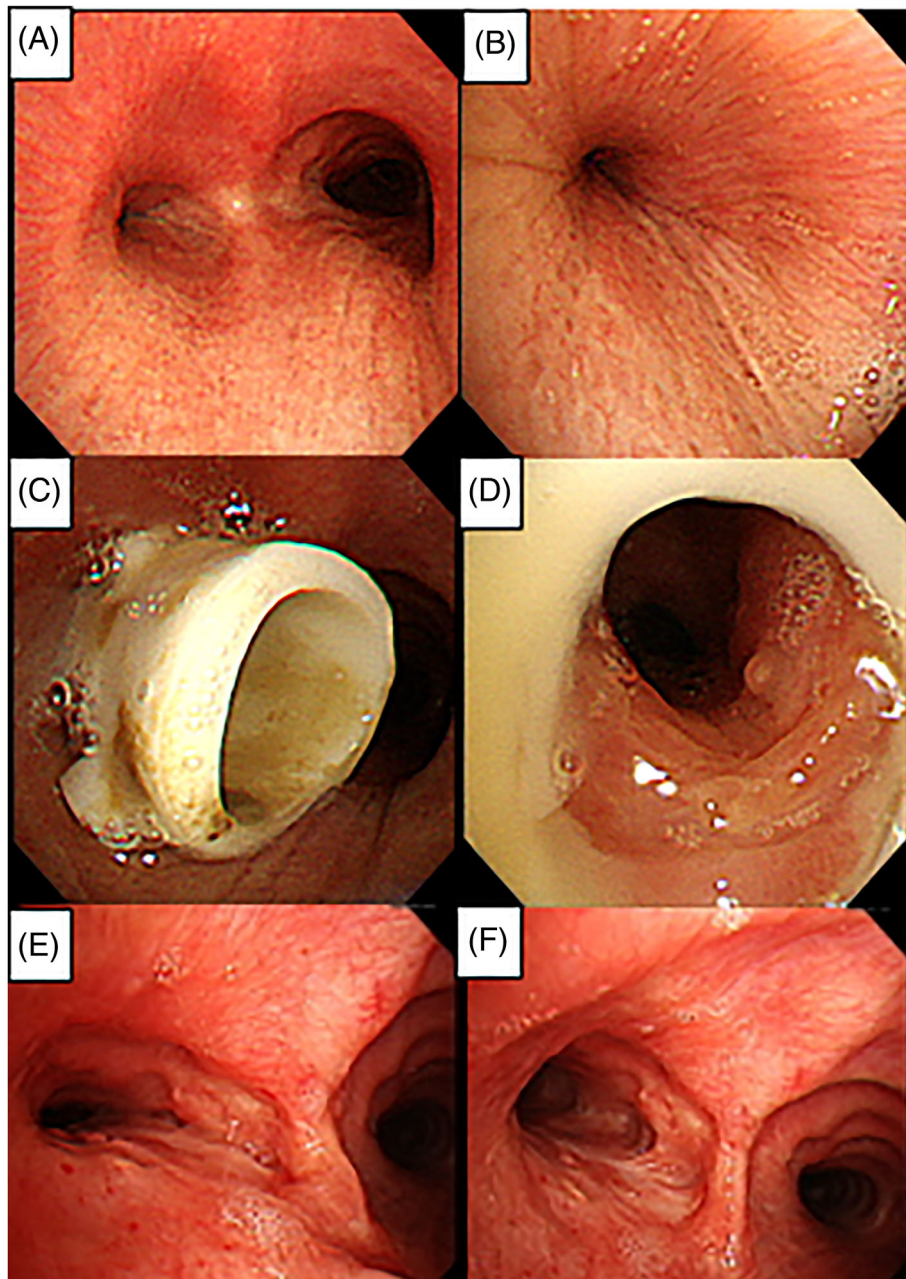
Twenty-two months after stenting, she had trouble expectorating due to granulation tissue at the distal end of the stent, and her flow-volume curves worsened on spirometry (Figure 3B). The stent was subsequently removed. After

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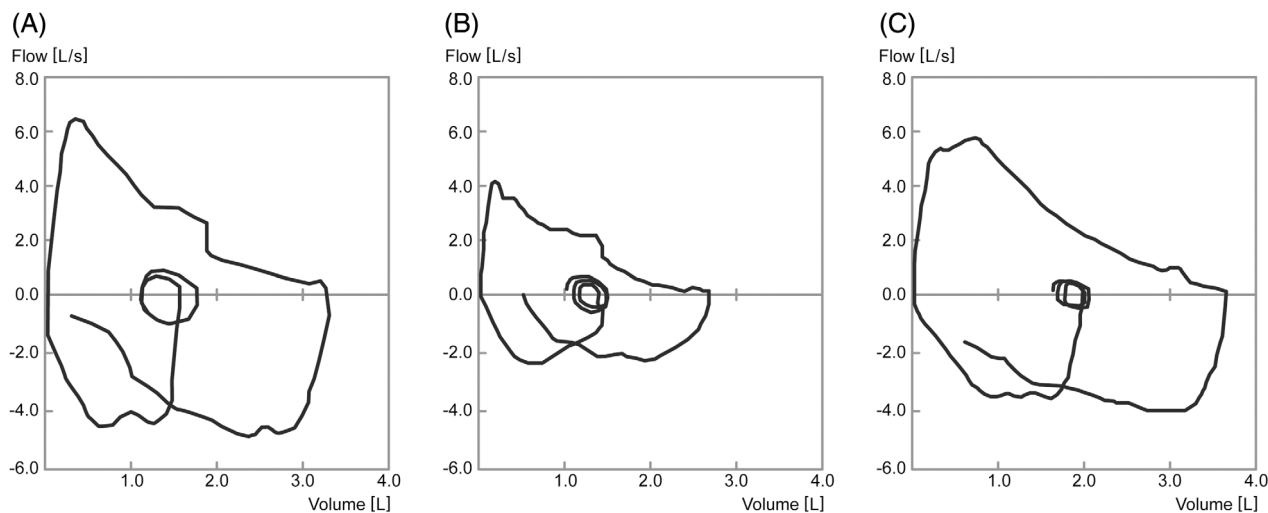
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**FIGURE 1** Chest CT findings. (A) Pre stenting, we can see a narrowed left main bronchus under lung window and (B) Mediastinal window



**FIGURE 2** Bronchoscopic findings. (A) Pre-stenting at the carina, and (B) the left main bronchus with a circumferential stenosis. (C) 22 months post-stenting at the carina, and (D) the distal end of the stent. (E) 30 months after stent removal at the carina on exhalation, and (F) on inhalation



**FIGURE 3** Flow volume curves. Pre stenting (A), post stenting (B), and 30 months after stent removal (C).

stent removal the left main bronchus was well maintained during inhalation; therefore, we decided that stent replacement was unnecessary. Thirty months after stent removal, bronchoscopic findings revealed the left main bronchus remained collapsed during exhalation (Figure 2E), but patency was maintained during inhalation without any respiratory symptoms (Figure 2F). In addition, granulation tissue at left main bronchus disappeared. After stent removal, flow volume curves improved, but expiratory flow limitation remained (Figure 3C).

## DISCUSSION

Airway stenting is often indicated for severe tracheal and/or bronchomalacia in RP patients. Metallic stents, although not recommended for benign central airway obstruction,<sup>5</sup> are often placed for circumferential type TBM due to airway narrowing. However, it has been reported that silicone stents are a suitable choice for airway involvements other than circumferential type TBM in RP patients.

While airway stents can maintain airway patency, complications such as granulation tissue, difficulty in expectorating, and re-stenosis often reduce the patients' quality of life. In this case, we thought the left main bronchial stenosis could have been caused by both inflammation and bronchomalacia by RP. The stent was able to be removed 22 months after placement since the pre-treatment pin hole-sized obstruction was dilated by steroid and biologic therapy. Although the left main bronchial malacia did not improve on exhalation after stent removal, she did not suffer from dyspnoea. Therefore, we decided stent replacement was unnecessary. Furthermore, airway patency was maintained during inhalation over 33 months after stent removal and granulation tissue disappeared. If her respiratory symptoms had worsened, non-invasive positive pressure ventilation would have been considered.<sup>6</sup>

To our knowledge, this is the first study to report a successful stent removal in an RP patient. In RP patients with TBM, it is difficult to remove airway stents due to the complete collapse of the central airway; however, temporary stenting might be a useful procedure for cases with severe unilateral main bronchial stenosis.

## AUTHOR CONTRIBUTIONS

Hiroshi Handa contributed to conception of the work and drafting the article. Hiroshi Handa, Hajime Tsuruoka, Shin Matsuzawa, Shinya Azagami and Masamichi Mine-shita participated in the broncoscopic intervention of this case. All authors approved the final version to be published.

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## CONFLICT OF INTEREST

None declared.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

## 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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