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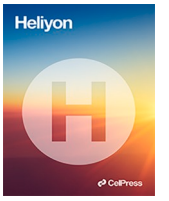

Case report

Comprehensive Interventional treatment for severe central airway collapse caused by Relapsing Polychondritis: A case report

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ABSTRACT

Relapsing Polychondritis (RP) is a rare systemic inflammatory disease. One major cause of death for patients with RP is severe tracheobronchial tree collapse. Treatment guidelines for RP are mainly based on case reports. We report a rare and challenging case of RP in a patient who experienced asphyxia due to severe central airway collapse. The patient had previously been misdiagnosed with refractory asthma due to recurrent wheezing. Following interventions including bronchoscopic laser tracheobronchoplasty, stent placement, corticosteroid therapy, and both invasive and non-invasive mechanical ventilation, the patient was successfully stabilized and subsequently discharged from the hospital. Notably, after discharge, the patient did not require rehospitalisation for worsening respiratory symptoms and was managed with only a gradually tapering glucocorticoid regimen. In our case report, stent placement rapidly relieved asphyxia due to severe tracheobronchial stenosis, and laser tracheobronchoplasty may be a potential cure for diffuse airway collapse due to RP.

1. Introduction

Relapsing Polychondritis (RP) is a rare, immune-mediated systemic inflammatory disease primarily affecting the cartilaginous structures of the ears, nose, respiratory tract, and joints [1]. For better management, several groups have categorized RP into phenotypic groups based on the affected organs. The respiratory subtype has been recognized most wildly, with airway involvement being a hallmark of this subtype [2,3]. The incidence rate of RP is approximately 0.71–4.5 per 1000000 per year, with 20–50 % of patients experiencing airway involvement [2]. More than one-fifth of these patients present with airway focal and diffuse malacia [3]. This condition results from inflammation and even destruction of the tracheobronchial cartilage, leading to a decrease in the hardness of the tracheobronchial tree or cartilage oedema, manifesting as airway collapse or stenosis [2]. Based on the reduction proportion of cross-sectional area, airway stenosis can be classified as mild (70%–80 %), moderate (81%–90 %), or severe (>90 %) [4]. Clinical symptoms of RP patients with tracheobronchial involvement typically include progressively worsening respiratory distress, cough, wheezing, and even respiratory failure. However, these manifestations are not specific and often lead to misdiagnosis and missed diagnosis, posing challenges to clinical diagnosis and treatment. Historically, airway collapse or stenosis caused by RP was a significant cause of death [5]. Although a clustering analysis showed no deaths caused by airway collapse or stenosis [6], in China, respiratory

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failure caused by airway collapse or stenosis remains a primary cause of mortality in RP patients [7]. Due to advancements in treatment and the evolution of bronchoscopic intervention techniques, there has been a significant reduction in the acute mortality rate of RP patients with airway involvement [8]. The fundamental treatment for RP is glucocorticoids (GCs). Patients with RP experiencing respiratory failure due to airway collapse or stenosis require tracheotomy, ventilatory support, and bronchoscopic stent placement to alleviate respiratory distress. Here, we report a rare and challenging case of severe central airway stenosis due to RP. The patient survived after bronchoscopic intervention, including laser tracheobronchoplasty, straight tube stent placement, L-shaped stent placement, combined with corticosteroid therapy, and airway positive pressure support ventilation.

2. Case presentation

2.1. Chief Complaints and history of present Illness

On May 9, 2023, a 58-year-old male patient with severe central airway collapse was transferred to our hospital. The patient had been misdiagnosed as having refractory asthma due to recurrent coughing and wheezing for over a year. During this period, the patient exhibited no additional systemic manifestations, including thoracic pain, arthralgia, or otolaryngological symptoms, and had intermittent hospital admissions for symptomatic treatment to relieve respiratory symptoms. The patient's medical history includes hypertension and a traumatic cranial injury resulting from a foreign object (a stick) penetrating the right orbital cavity and entering the cranial vault. Upon the latest hospital admission, the patient's chest computed tomography (CT) images showed pronounced tracheal and bronchial wall thickening, with luminal stenosis exceeding 95 % in the narrowest segment of the central airway. This was accompanied by complete atelectasis of the left lung and severecentral airway stenosis, necessitating emergency inter-hospital transfer via ambulance for advanced management.

2.2. Emergency Interventions and initial Recovery phase

Upon the patient's arrival, the patient's respiratory distress notably worsened, presenting with cyanosis of the face and lips, and an oxygen saturation ranging between 85 and 90 % under high-flow nasal oxygen therapy set at 35 L/min with an oxygen concentration

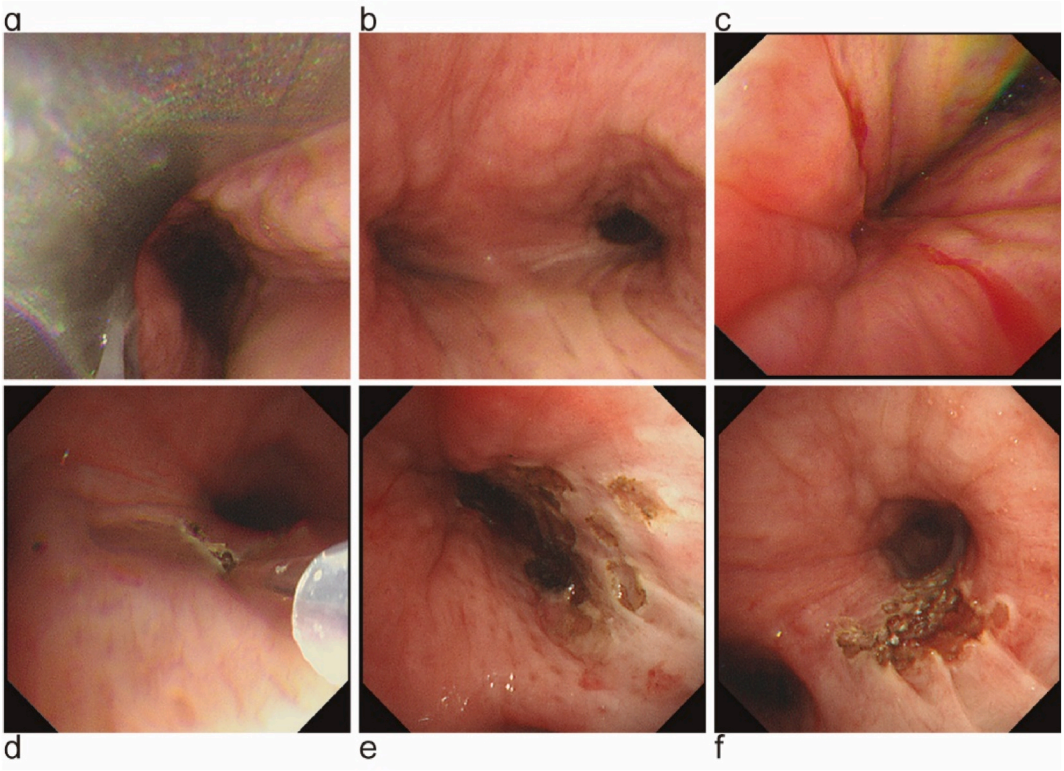

Fig. 1. Findings of the bronchoscopy and laser intervention. [(a,b) Bronchoscopy showed severe congestion, oedema of the tracheobronchial mucosa, disappearance of cartilage ring, collapse of the tracheobronchial lumen, while the membranous portion was unaffected; (c) The narrowest section (left main bronchus) was approximately 95 % narrowed in cross-sectional area; (d) Laser ablation was performed from distal to proximal on the mucosa between the narrow sections of the left and right main bronchi and the membranous part; (e,f) After treatment, the lumen of the left and right main trachea was more patent than before.].

of 40 %. Given the critical condition of the patient, emergency surgery was deemed necessary. A rigid bronchoscopy was performed at 13:54, revealed significant findings: There was severe congestion, oedema of the tracheobronchial mucosa and collapse of the tracheobronchial lumen, while the membranous portion was unaffected (Fig. 1a and b). The cartilage rings were no longer visible, and pronounced mucosal ridges led to a severe narrowing of the lumen. The narrowest section (left main bronchus) was approximately 95 % narrowed in cross-sectional area (Fig. 1c). Laser ablation was performed from distal to proximal on the mucosa between the narrow sections of the left and right main bronchi and the membranous part (Fig. 1d), followed by multiple submucosal injections of 5mg dexamethasone. Tissue samples were also taken above the carina of trachea for pathological examination. After treatment, enhanced patency of the tracheal lumen was observed compared to pre-intervention status (Fig. 1e and f). Considering the possibility of post-operative airway oedema and necrotic obstruction, the patient was transferred to the ICU for further treatment. ICU bedside chest X-rays showed complete atelectasis of the left lung (Fig. 2a). The patient was given invasive mechanical ventilation via nasal tracheal intubation (PC mode: FiO2 100 %, PEEP 7cmH2O, fpm 15 breaths/min), and intravenous infusion of methylprednisolone sodium succinate 80mg for anti-inflammatory treatment, with a slight improvement in SpO2. At 18:00, the patient's breathing difficulties intensified, becoming agitated with progressively decreasing SpO2, fluctuating between 70 and 85 %. Given the significant collapse of the tracheobronchial tract, with predominant involvement of the left main bronchus, an urgent transfer to the bronchoscopy suite was undertaken to facilitate the placement of a straight membrane-covered metallic stent (12–40mm, Micro-tech Co., Ltd, Nanjing, China) into the left main bronchus. The stent was well deployed, with the upper edge at the opening of the left main bronchus and the lower edge at the distal end of the left main bronchus (Fig. 2b and c), significantly improving the patient's breathing difficulties post-operation.

2.3. *Further surgical Adjustments*

On May 11th, the patient's bedside chest X-rays revealed partial re-expansion of the left lung, indicating improvement compared to previous assessments (Fig. 2d). In an effort to further address airway constriction, the previously placed stent was surgically removed and the tracheal tube was exchanged. Following this, an L-shaped membrane-covered metallic stent (16–60mm/12-35mm, Micro-tech Co., Ltd, Nanjing, China.) was inserted into the left main bronchus and the lower third of the trachea. This stent was successfully deployed, positioning its upper edge within the mid-segment of the trachea and its lower edge at the distal terminus of the left main bronchus, ensuring full exposure of the left main bronchus (Fig. 2e). The patient continued to receive invasive mechanical ventilator support and medication treatment. On May 13th, the patient's bedside bronchoscopy revealed that the L-shaped stent had shifted and overlapped inside the tracheal tube (Fig. 2f), likely due to compression from the collapsed and narrowed left main bronchus. Using

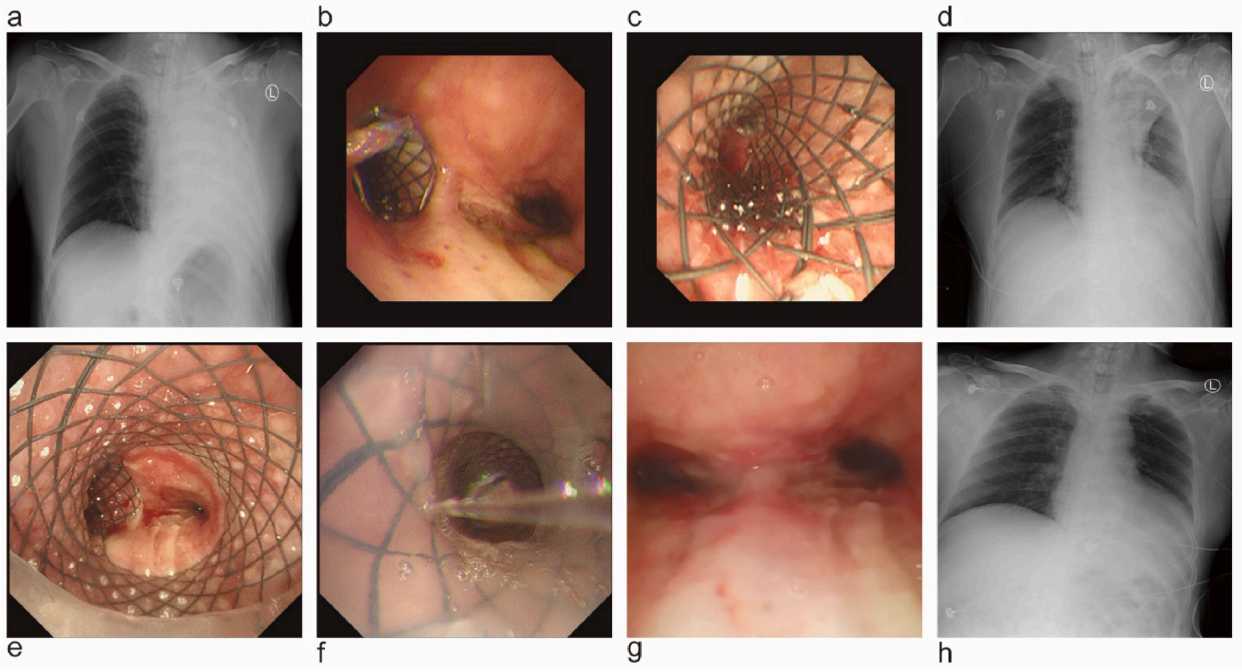

Fig. 2. Bronchoscopic stent placement. [(a) Chest X-rays shows a complete opacity of the left side of the hemithorax; (b,c) A straight of membrane-covered metallic stent was placed in the left main bronchus; (d) Chest X-rays shows partial translucency in the upper lobe of the left lung after stent placement; (e) L-shaped membrane-covered metallic stent was placed in trachea and left main bronchus; (f) L-shaped stent displaced and embedded in the tracheal tube; (g,h) After comprehensive treatment, bronchoscopy showed that the lumen of the right and left main bronchi was more patent than before; review of chest X-rays shows the translucency of the left lung was increased, and the ventilation function of the left lung was improved than before.].

biopsy forceps, the extraction loop at the lower end of the metal-covered stent was grasped and gradually separated from the tracheal tube by continuous winding and pulling. The stent was removed with the biopsy forceps at the same time as the tracheal tube was pulled out. Further inspection revealed significant improvement in the narrowing of tracheobronchial lumen compared to before (Fig. 2g). A re-examination of bedside chest X-rays showed an improved state of partial lung collapse compared to before (Fig. 2h). After treatment with GCs, laser, stent support, and mechanical ventilator support, the patient had a noticeable improvement in central airway narrowing and experienced no significant breathing difficulties, and was possible to wean off the machine. After three days of weaning training, the tracheal tube was successfully removed, and the patient was discharged, with significantly improved coughing and difficulty breathing compared to admission.

The patient's histopathological analysis indicated both acute and chronic inflammation of the mucosal tissues, with significant compression observed. Congo red and Periodic Acid-Schiff staining yielded negative results. The whole treatment process is shown in Fig. 3.

2.4. Follow-up

During the six-month follow-up, the patient demonstrated considerable improvement, requiring only intermittent supplemental oxygen via a nasal cannula. Notably, there were no subsequent hospital admissions related to exacerbated respiratory difficulties.

3. Discussion

RP predominantly impacts middle-aged individuals, aged 40–60 years, as a systemic inflammatory condition [9]. Dion and colleagues have classified RP into several subtypes: the hematologic type, recently considered as VEXAS syndrome; the respiratory subtype; and a milder variant, typically associated with a more favorable prognosis [1,6]. The approach to management and the expected outcomes differ across these categories. The respiratory subtype distinguished by the tracheobronchial cartilage's involvement, which can cause the cartilage to inflame or disintegrate, leading to varying levels of airway collapse or stenosis. Such conditions progressively exacerbate inspiratory breathing difficulties, potentially culminating in asphyxiation, thus highlighting tracheobronchial involvement as a significant mortality risk in RP. In China, the reported mortality rate for RP with airway involvement has reached up to 67 % [7]. Prompt and appropriate intervention can significantly mitigate the extent of airway constriction and halt its progression. Thus, identifying RP at an early stage is critical, particularly when the airway is affected.

3.1. Diagnosis and Differential diagnosis

Since RP lacks specific laboratory markers and the utility of histological biopsy is limited, the diagnosis largely depends on the clinical presentation. The most widely accepted diagnostic criteria include McAdam criteria; Damiani and Levine criteria; and Michet criteria, with the latter having the highest sensitivity [10]. However, the lack of specificity in the respiratory symptoms presented by the patient and the absence of involvement of other organs always led to a misdiagnosis of intractable bronchial asthma prior to admission [11–14]. Therefore, in addition to clinical manifestations, diagnosis for airway-involved RP should encompass bronchoscopic examination, imaging findings, pulmonary function test results, and histological characteristics. In terms of imaging, particularly chest CT scans, typically reveals laryngeal soft tissue enlargement, thickening of the tracheobronchial walls, narrowing or obstruction of the lumen, retention of air during expiration, and calcification of tracheal cartilage. Pulmonary function tests predominantly show obstructive ventilation dysfunction. Depending on the disease stage, flow-volume loops may show fixed airway

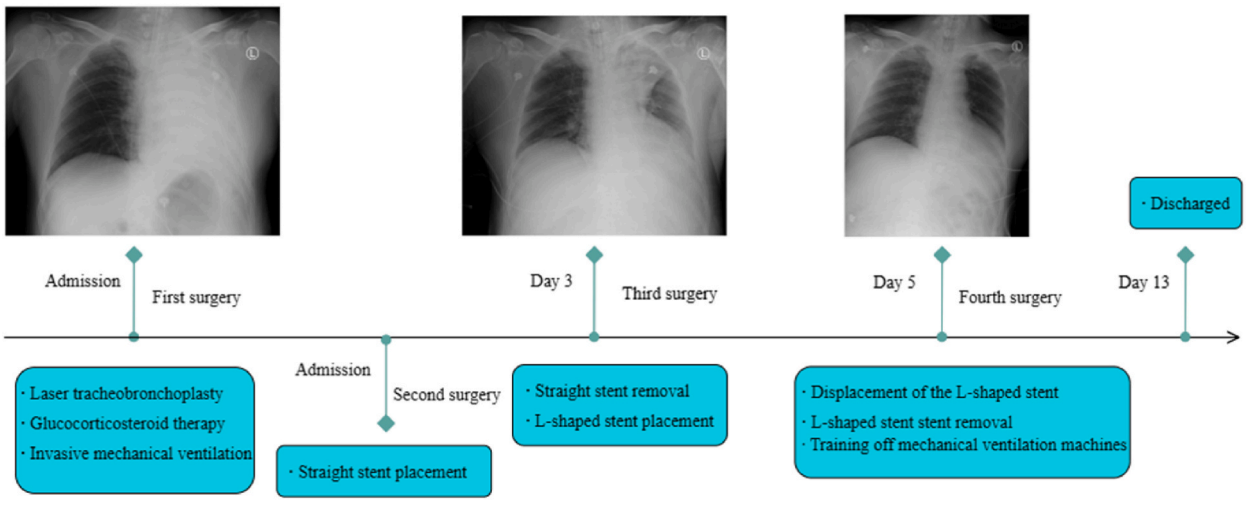

Fig. 3. Patient treatment progression and outcome during Hospitalisation.

obstruction, variable intrathoracic obstruction and variable extrathoracic obstruction. Bronchoscopic examination commonly identifies mucosal erythema, hypertrophy, lumen narrowing or collapse, with the membranous part remaining normal [15]. During acute inflammation, the histological manifestation of the trachea and bronchi mucosa is acute inflammatory oedema. With recurrent episodes of inflammation over time, the cartilaginous structures can collapse, resulting in tracheomalacia and bronchomalacia, along with thickening, fibrosis, and calcification of the tracheobronchial walls [16].

Similar to other studies [17,18], we made a clinical diagnosis after excluding other differential diagnoses. The differential diagnosis includes tracheal intubation, tracheostomy, tuberculosis, amyloidosis, granulomatosis with polyangiitis (GPA), tracheobronchopathia osteochondroplastica (TPO). Tracheal intubation and tracheostomy are the main causes of traumatic conditions, with tracheobronchial stenosis usually appearing 3–6 weeks after extubation. Bronchoscopy typically reveals fibrotic or scarred stenosis [19]. In cases of tracheobronchial tuberculosis, chest CT often shows the "tree-in-bud" appearance, segmental bronchial narrowing with concentric wall thickening, and possible evidence of extrinsic obstruction by affected lymph nodes. Bronchoscopy typically shows fibrotic stenosis or inflammatory ulcerative stenosis [20]. Tracheobronchial amyloidosis on CT imaging typically presents with circumferential (including membranous trachea) or unilateral thickening of the tracheobronchial walls with calcified plaques within the thickened areas or diffuse nodular thickening of the trachea and main bronchi [21,22]. Pathological biopsy shows submucosal amyloid deposits, positive Congo red staining, and yellow-green birefringence under polarized light [23]. GPA-associated stenosis typically features circumferential stenosis, primarily involving the subglottic area without calcification [24]. TPO presents similarly to RP with long-segment tracheobronchial stenosis, sparing the membranous part. However, TPO is characterized by multiple submucosal osteocartilaginous nodules protruding into the lumen on bronchoscopy and chest CT [25]. Our patient initially presented with symptoms of tracheobronchial involvement with no involvement of other organs. Due to the urgency of the patient's condition, pulmonary function tests were not conducted. Bronchoscopic findings supported the CT results before admission, showing inflammatory oedema of the tracheobronchial mucosa and severe collapse of the airway lumen, with no affection of the membranous parts. Histopathology showed non-specific inflammation with no amyloid deposition, and Congo red and periodic acid-Schiff staining were negative. Consequently, the patient was diagnosed with RP involving the tracheobronchus.

3.2. Treatment

Given the rarity of RP, a standardised treatment protocol remains undefined. Contemporary clinical guidelines advocate the use of nonsteroidal anti-inflammatory drugs (NSAIDs), GCs, disease-modifying antirheumatic drugs (DMARDs), and biological agents [1]. GCs are considered the first-line treatment, with combination intensive therapy involving DMARDs and biologics typically reserved for cases that are dependent on or resistant to GC treatment [1]. The use of DMARDs often aims to reduce the required dosage of GCs. Recent studies have shown that biologics may improve survival in some RP patients and that early use may delay disease progression [26,27]. However, adverse reactions such as respiratory infections led to a treatment discontinuation rate of around 20 % [26]. The risk of infections, particularly respiratory infections, which are a leading cause of mortality in patients with tracheobronchial RP [28,29]. Biologics also only reduced the average GC dose by approximately 5 mg of prednisone equivalent [26]. Additionally, a UK study indicated that patients with tracheobronchial involvement in RP often do not respond well to biologics [14]. Therefore, a careful risk-benefit assessment of intensive therapy for late-stage RP involving airway is necessary.

Upon admission, the patient immediately received intravenous and submucosal tracheal corticosteroid treatment. Nevertheless, medical therapies including GCs or DMARDs did not provide immediate relief for symptoms in patients with severe airway collapse due to RP [3,30]. In patients with RP presenting with life-threatening severe central airway collapse, interventions extending beyond pharmacological management are essential. These include tracheostomy, support via mechanical ventilation employing bilevel positive airway pressure (BiPAP), and the insertion of bronchoscopic stents [15]. The application of bronchoscopic stent placement during acute RP flare-ups has been shown to promptly relieve the symptoms attributable to tracheobronchial constriction [31], significantly enhancing patients' quality of life. Among the stent options, silicone stents or membrane-covered metallic stents, especially those of bifurcated designs (L or Y-shaped), are preferred for significant airway narrowing. A Y-shaped silicone stent would typically be more suitable for this patient due to its stability. Nevertheless, in the hospital, we could not find an appropriately sized silicone stent based on the measurements from the CT scan and bronchoscopy in the short period of time. To quickly relieve asphyxiation due to diffuse airway collapse, a straight stent was initially placed and later replaced with an L-shaped stent for better airway management. Although these were not our first choice, they effectively stabilized the patient's condition by rapidly improving blood oxygen levels. Nevertheless, the post-placement period of stents is associated with potential complications such as retention of secretions, obstruction due to granulomatous growth, or complications arising from stent migration and fracture under the forces of airway collapse, leading to pneumothorax or further stenosis. Hence, the long-term deployment of stents is not advocated. In addition, throughout the process, we used positive-pressure ventilation on the ventilator improve blood oxygenation and the utilisation of mechanical ventilatory support is considered beneficial during acute phases [32]. Tracheostomy should be reserved as a last recourse, following the ineffectiveness of other modalities, due to its potential to harm to tracheal cartilage and walls, thereby aggravating the condition of airway collapse.

To our knowledge, we are the first to report the use of laser ablation for severe airway stenosis caused by RP. During the initial surgical procedure, laser tracheobronchoplasty was performed on the posterior wall of the patient's tracheobronchial tree. It is a novel and effective technique for improving tracheobronchial collapse [33–35]. This technique involves the application of laser to the membranous sections of the tracheobronchial wall, promoting fibrosis and subsequent stiffening of the tissue, which aids in reducing collapse [36]. Laser tracheobronchoplasty works primarily through two mechanisms: photobiomodulation and photothermal reactions [37]. (1) Photobiomodulation involves the absorption of light by cytochrome *c* oxidase in cells, which increases levels of reactive oxygen species and activates transcription factors for cell repair. This process also promotes the release of nitric oxide (NO), improving

blood flow by dilating blood vessels; (2) Photothermal reactions occur when light energy is converted to heat, causing a localized increase in temperature within the tissues. This heating effect can cause controlled damage that triggers a healing response [37,38]. In vitro studies have shown that laser therapy on tracheobronchial tissues can create a "thermal injury effect" that promotes tissue repair and increases fibroblast activity and collagen deposition without causing full tissue erosion [39,40]. These processes suggest that laser treatment encourage tissue proliferation and fibrosis, which are key steps in wound healing involving fibroblast proliferation, collagen synthesis, and extracellular matrix formation. Signaling molecules like transforming growth factor (TGF-β), platelet-derived growth factor (PDGF), and interleukin-1 (IL-1) are critical in promoting fibrosis [41,42]. However, the specific molecular mechanisms in laser tracheobronchoplasty require further study, as most of the current evidence comes from animal models or studies of skin injuries.

Notably, after discharge, our patient did not require rehospitalisation for worsening respiratory symptoms and was managed with only a gradually tapering GC (prednisone, 10 mg) regimen. This suggests that laser tracheobronchoplasty may provide long-term benefits in cases of severe airway collapse due to RP, as it may reduce respiratory symptoms by stabilising the airway. This approach also minimizes the need for additional immunosuppressants or biologics, reducing the risk of infection and avoiding the need for long-term stent placement. However, serious adverse reactions, such as tracheal fistula or airway fire, can occur during laser ablation due to factors like the angle of the laser probe or excessive laser power. To avoid adverse reactions, we set the energy at 12–15W, with each laser application lasting less than 3 seconds. We also maintained the inhaled oxygen concentration below 40 %. After laser ablation, airway oedema at the ablation site may cause difficulty in breathing and sputum clearance. Therefore, we retained tracheal intubation for more than one week. Daily bedside bronchoscopy was performed to care for the airway, including clearing tracheal secretions and necrotic tissue. Intravenous steroids were administered to reduce inflammatory oedema. Additionally, in other subsequent procedures for severe airway collapse, we performed laser tracheobronchoplasty alongside stent placement to achieve more effective airway stabilisation meanwhile reduce the impact of inadvertently aggravated airway inflammation.

4. Conclusion

In conclusion, for RP cases confined to the airways, comprehensive early detection incorporating clinical manifestations, bronchoscopic examination, radiological assessments, pulmonary function test results, and histological characteristics is paramount for initiating prompt and regulated therapy to avert further tracheal constriction. Apart from anti-inflammatory treatment with GCs, the implementation of bronchoscopic laser tracheobronchoplasty and stent insertion were pivotal for the treatment. The immediate relief of tracheobronchial narrowing symptoms through stent placement, alongside the potential of laser tracheobronchoplasty for effective airway collapse intervention, underscores their significance.

Ethics statement

The research was conducted in accordance with the World Medical Association Declaration of Helsinki. The patient provided written informed consent for the publication of this case report and any accompanying images. This study protocol was reviewed and approved by [Ethics Committee of Dongzhimen Hospital affiliated to Beijing University of Chinese Medicine], approval number [2024DZMEC-207-01]. This study has obtained written informed consent from the participants. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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CRediT authorship contribution statement

Youqiang Wu: Writing – original draft, Investigation. **Mingzhe Wang:** Writing – review & editing, Conceptualization. **Hongwu Wang:** Project administration, Conceptualization. **Chengjun Ban:** Conceptualization. **Xuechun Tang:** Writing – review & editing. **Yi Luo:** Investigation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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References

[1] P. Mertz, J. Sparks, D. Kobrin, S.A. Ogbonnaya, E. Sevim, C. Michet, et al., Relapsing polychondritis: best practice & clinical rheumatology, Best Pract. Res. Clin. Rheumatol. 37 (1) (2023) 101867.
[2] S. Rednic, L. Damian, R. Talarico, C.A. Scirè, A. Tobias, N. Costedoat-Chalumeau, et al., Relapsing polychondritis: state of the art on clinical practice guidelines, RMD Open 4 (Suppl 1) (2018) e000788. Published 2018 Oct 18.
[3] A. Ernst, S. Rafeq, P. Boiselle, A. Sung, C. Reddy, G. Michaud, et al., Relapsing polychondritis and airway involvement, Chest 135 (4) (2009) 1024–1030.
[4] Tracheomalacia in adults: clinical features and diagnostic evaluation - UpToDate. https://www.uptodate.com/contents/tracheomalacia-in-adults-clinical-features-and-diagnostic-evaluation, 2024. (Accessed 2 April 2024).
[5] N. Hazra, A. Dregan, J. Charlton, M.C. Gulliford, D.P. D'Cruz, Incidence and mortality of relapsing polychondritis in the UK: a population-based cohort study, Rheumatology 17 (2015) kev240. Published online July.
[6] J. Dion, N. Costedoat-Chalumeau, D. Sène, J. Cohen-Bittan, G. Leroux, C. Dion, et al., Relapsing polychondritis can Be characterized by three different clinical phenotypes: analysis of a recent series of 142 patients, Arthritis Rheumatol. 68 (12) (2016) 2992–3001.
[7] D.F. Lin, W.Q. Yang, P.P. Zhang, Q. Lv, O. Jin, J.R. Gu, Clinical and prognostic characteristics of 158 cases of relapsing polychondritis in China and review of the literature, Rheumatol. Int. 36 (7) (2016) 1003–1009.
[8] S. Rafeq, D. Trentham, A. Ernst, Pulmonary manifestations of relapsing polychondritis, Clin. Chest Med. 31 (3) (2010) 513–518.
[9] A. Horváth, N. Páll, K. Molnár, T. Kováts, G. Surján, T. Vicsek, et al., A nationwide study of the epidemiology of relapsing polychondritis, CLEP 8 (2016) 211–230.
[10] T. Rose, U. Schneider, M. Bertolo, J. Klotsche, V. Casteleyn, R. Biesen, et al., Observational study and brief analysis of diagnostic criteria in relapsing polychondritis, Rheumatol. Int. 38 (11) (2018) 2095–2101.
[11] E. Sheffield, B. Corrin, Fatal bronchial stenosis due to isolated relapsing chondritis, Histopathology. 20 (5) (1992) 442–443.
[12] C. Gorard, S. Kadri, Critical airway involvement in relapsing polychondritis, BMJ Case Rep. 2014 (2014) bcr2014205036.
[13] S. Winstanley, A. Boyde, R. Attanoos, Fatal relapsing tracheobronchial polychondritis diagnosed at autopsy, BMJ Case Rep. 2015 (2015) bcr2015209483.
[14] S. Dubey, C. Gelder, G. Pink, A. Ali, C. Taylor, J. Shakespeare, et al., Respiratory subtype of relapsing polychondritis frequently presents as difficult asthma: a descriptive study of respiratory involvement in relapsing polychondritis with 13 patients from a single UK centre, ERJ Open Res 7 (1) (2021) 170–2020.
[15] N. De Montmollin, D. Dusser, C. Lorut, J. Dion, N. Costedoat-Chalumeau, L. Mouthon, et al., Tracheobronchial involvement of relapsing polychondritis, Autoimmun. Rev. 18 (9) (2019) 102353.
[16] J. Catano, Y. Uzunhan, R. Paule, J. Dion, A. Régent, P. Legendre, et al., Presentation, diagnosis, and management of subglottic and tracheal stenosis during systemic inflammatory diseases, Chest 161 (1) (2022) 257–265.
[17] T. Yanagihara, M. Ohgushi, T. Setoguchi, N. Ogo, Y. Inutsuka, H. Fujiwara, et al., Early recognition and treatment of relapsing polychondritis, Cureus 14 (1) (2022) e21463.
[18] S. Suzuki, A. Ikegami, Y. Hirota, M. Ikusaka, Fever and cough without pulmonary abnormalities on CT: relapsing polychondritis restricted to the airways, Lancet 385 (9962) (2015) 88.
[19] L. Freitag, A. Ernst, M. Unger, K. Kovitz, C.H. Marquette, A proposed classification system of central airway stenosis, Eur. Respir. J. 30 (1) (2007) 7–12.
[20] T. Shahzad, M. Irfan, Endobronchial tuberculosis—a review, J. Thorac. Dis. 8 (12) (2016) 3797–3802.
[21] J.H. Chung, J.P. Kanne, M.D. Gilman, CT of diffuse tracheal diseases, Am. J. Roentgenol. 196 (3) (2011) W240–W246.
[22] F. Czeyda-Pommersheim, M. Hwang, S.S. Chen, D. Strollo, C. Fuhrman, S. Bhalla, Amyloidosis: modern cross-sectional imaging, Radiographics 35 (5) (2015) 1381–1392.
[23] X. Lu, B. He, G. Wang, B. He, L. Wang, Q. Chen, Bronchoscopic diagnosis and treatment of primary tracheobronchial amyloidosis: a retrospective analysis from China, BioMed Res. Int. 2017 (2017) 1–7.
[24] C. Girard, P. Charles, B. Terrier, G. Bussonne, P. Cohen, C. Pagnoux, et al., Tracheobronchial stenoses in granulomatosis with polyangiitis (Wegener's): a Report on 26 Cases, Medicine (Baltim.) 94 (32) (2015) e1088.
[25] N. Kamangar, Tracheobronchopathia Osteochondroplastica, Mayo Clin. Proc. 94 (6) (2019) 949–950.
[26] G. Moulis, G. Pugnet, N. Costedoat-Chalumeau, A. Mathian, G. Leroux, J. Boutémy, et al., Efficacy and safety of biologics in relapsing polychondritis: a French national multicentre study, Ann. Rheum. Dis. 77 (8) (2018) 1172–1178.
[27] A. Petitdemange, C. Sztejkowski, L. Damian, T. Martin, L. Mouthon, Z. Amoura, et al., Treatment of relapsing polychondritis: a systematic review, Clin. Exp. Rheumatol. 40 (5) (2022) 81–85. Suppl 134.
[28] D. Wang, L. Guan, X. Dong, X. Zhu, Z. Tong, Comparison of relapsing polychondritis patients with and without respiratory involvement based on chest computed tomography: a retrospective cohort study, BMC Pulm. Med. 22 (1) (2022) 222.
[29] N. Hazra, A. Dregan, J. Charlton, M.C. Gulliford, D.P. D'Cruz, Incidence and mortality of relapsing polychondritis in the UK: a population-based cohort study, Rheumatology 54 (12) (2015) 2181–2187.
[30] A. Alaga, H. Mohammed, A. Ishida, M. Oki, H. Saka, Multiple endobronchial airway stents in a case of relapsing polychondritis - A rare entity, Respiratory Medicine Case Reports 36 (2022) 101583.
[31] S.A.M. Nashef, C. Dromer, J.F. Velly, L. Labrousse, L. Couraud, Expanding wire stents in bengin tracheobronchial disease: Indications and complications, Ann. Thorac. Surg. 54 (5) (1992) 937–940.
[32] M. Adliff, D. Ngato, S. Keshavjee, S. Brenaman, J.T. Granton, Treatment of Diffuse Tracheomalacia Secondary to Relapsing Polychondritis With Continuous Positive Airway Pressure, Chest 112 (6) (1997) 1701–1704.
[33] H. Dutau, F. Maldonado, D.P. Breen, A. Colchen, Endoscopic successful management of tracheobronchomalacia with laser: apropos of a Mounier-Kuhn syndrome, Eur. J. Cardio. Thorac. Surg. 39 (6) (2011) e186–e188.
[34] J. Plojoux, S. Laroumagne, T. Vandemoortele, P.J. Astoul, P.A. Thomas, H. Dutau, Management of Benign Dynamic "A-Shape" Tracheal Stenosis: A Retrospective Study of 60 Patients, Ann. Thorac. Surg. 99 (2) (2015) 447–453.
[35] Y. Wang, Y.P. Gao, L. Jing, X.L. Li, F. Qing, J.L. Zhang, K. Liu, et al., Therapeutic effect analysis of excessive dynamic airway collapse treated by laser (13 cases), China J. Endosc. 30 (3) (2024) 73–80. Chinese.
[36] P. Castellanos, M. Mk, I. Atallah, Laser tracheobronchoplasty: a novel technique for the treatment of symptomatic tracheobronchomalacia, Eur. Arch. Oto-Rhino-Laryngol. 274 (3) (2017) 1601–1607.
[37] S.P. Gangadharan, F. Mathew, Thermoablative Techniques to Treat Excessive Central Airway Collapse, Thorac. Surg. Clin. 33 (3) (2023) 299–308, https://doi.org/10.1016/j.thorsurg.2023.04.016.
[38] G. Insero, F. Fusi, G. Romano, The safe use of lasers in biomedicine: Principles of laser-matter interaction, J Public Health Res 12 (3) (2023) 22799036231187077.
[39] A. de Lima, B. Vidal, F. Kheir, P.A. VanderLaan, P.S. Mallur, S.P. Gangadharan, et al., Thermoablative Techniques for Excessive Central Airway Collapse: An Ex Vivo Pilot Study on Sheep Tracheal Tissue, J Bronchology Interv Pulmonol 27 (3) (2020) 195–199.
[40] F. Kheir, D. Ospina-Delgado, J. Beattie, R. Singh, B. Vidal, P.A. VanderLaan, et al., Argon Plasma Coagulation (APC) for the Treatment of Excessive Dynamic Airway Collapse (EDAC): An Animal Pilot Study, J Bronchology Interv Pulmonol 28 (3) (2021) 221–227.
[41] H.N. Wilkinson, M.J. Hardman, Wound healing: cellular mechanisms and pathological outcomes, Open Biol 10 (9) (2020) 200223.
[42] J. Larouche, S. Sheoran, K. Maruyama, M.M. Martino, Immune Regulation of Skin Wound Healing: Mechanisms and Novel Therapeutic Targets, Adv. Wound Care 7 (7) (2018) 209–231.