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Argon gas knife combined with cryotherapy for amyloidosis leading to severe airway stenosis

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ABSTRACT

Objective: This case report shows that bronchoscopy is an important method to treat severe airway stenosis caused by bronchial amyloidosis. Bronchoscopic forceps were used to incise the intra-tracheal lump repeatedly. The incision was frozen with a cryosurgery probe, argon knife was used to stop the bleeding until the airway lumen stenosis was reduced to approximately 40%, after which, it continued to enter the lumen. We used bronchoscopic biopsy forceps to repeatedly clamp the lumps in the tracheal carina and left and right main bronchial tumors until the lumen was completely unobstructed.

Results: The symptoms of severe dyspnea and wheezing were significantly improved after two interventions with the bronchoscope.

1. Introduction

Systemic amyloidosis (AM) refers to a group of diseases, in which insoluble proteins are deposited in tissues or organs, causing their dysfunction [1]. AM rarely causes respiratory system involvement. If airway involvement occurs, it often endangers life and has a poor prognosis. At present, there is no standard and effective treatment for amyloidosis. We present a case of severe airway stenosis caused by tracheobronchial amyloidosis.

2. Case presentation

A 51-year-old Chinese female presented with complaints of recurrent wheezing, cough, and yellow sputum production for the past seven years. The patient was hospitalized several times with the diagnosis of bronchial asthma or pneumonia. During her non-hospitalization, she received intermittent inhalation of budesonide/formoterol dry powder as maintenance therapy. During 2016 and 2017, she was repeatedly admitted to the intensive care unit (ICU) with acute respiratory failure, and underwent invasive mechanical ventilation with endotracheal intubation twice. On February 7, 2018, she developed type II respiratory failure due to aggravation of dyspnea and was hospitalized again. Physical examination revealed a middle-aged woman with poor health status characterized by dyspnea, cyanosis of lips, hoarseness, and dysphonia, with a temperature of 36.4 degree centigrade, a pulse rate of 144 beats per minute, respiratory rate of 32 breaths per minute, blood pressure 128/65 mmHg. On lung examination, the lungs were full of wheezing sound, mainly in the inspiratory phase, accompanied by serrated and intermittent breath sounds. Her heart rate was 144 beats per minute, with regular heart rhythm, and grade III systolic murmur at the cardiac apex. Her WBC was elevated to 25.17×109/L with neutrophilic predominance up to 92.31%. The arterial blood gas analysis report showed a pH of 7.272, pCO_2 of 48 mmHg, and pO_2 of 56.6 mmHg. Lung CT revealed that the upper middle wall of the trachea was thickened and the lumen was severely narrow. She received antibiotics, anti-asthmatic medication, and symptomatic treatment, and her clinical symptoms improved after five days. However, lung CT re-examination showed no significant improvement. Bronchoscopy was recommended but after her family refused, the patient was discharged. On February 28, 2018, the woman presented to emergency department with dyspnea, orthopnea, profuse sweating, and irritability. When the patient underwent pulmonary CT examination, dyspnea was further aggravated, and the results of pulmonary CT examination showed that upper airway stenosis was further aggravated. With the consent of the patient and her family, emergency interventional therapeutic bronchoscopy was performed.

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The patient underwent the first interventional therapeutic bronchoscopy at 9:00 on February 28, 2018. The patient was pushed into the operating room under high-flow oxygen inhalation, the No. 4 laryngeal mask was inserted after intravenous induction anesthesia, and the bronchoscope was inserted through the laryngeal mask. Under the bronchoscope, a lump 3 cm below the glottis could be seen obstructing the airway and causing 95% of the airway stenosis, with a large amount of white viscous secretion on the surface (Fig. 1).The abnormal secretions were sucked out by a bronchoscope, then the needle-shaped electric knife was introduced through the forceps of the bronchoscope to cut the left tracheal lump repeatedly, then the cryosurgery probe was introduced to freeze the cut site, and the argon knife was used to stop the bleeding. When the airway stenosis decreased to 40% after treatment, bronchoscopy was continued to access the left and right main bronchi; the lump was found to invade the tracheal carina and the left and right main bronchi (Fig. 2). The bronchoscopy biopsy forceps were used to repeatedly cut the lump in these areas until the airway lumen was completely patent. The operation was completed, and the biopsy tissues were examined pathologically. Postoperatively, patients' vital signs were stable, dyspnea was relieved, and a small amount of hemoptysis that was found at the beginning, disappeared in 2 days, and the pathological return was amyloidosis (Fig. 3A and B).The patient was discharged on March 5, 2018.

The patient underwent the second interventional therapeutic bronchoscopy on March 27, 2018. The patient was transferred to the operating room, the No. 4 laryngeal mask was inserted after intravenous induction anesthesia, and the bronchoscope was inserted through the laryngeal mask. Bronchoscopic examination showed that there was approximately 40% widespread stenosis from 3 cm below glottis till the tracheal carina (Fig. 4A). The argon nozzle (leside spray nozzle or ring) was introduced through the forceps of the bronchoscope for repeated electrocautery. Thereafter, biopsy forceps were used to reduce the tracheal stenosis to 20% (Fig. 4B), and bronchoscopy was continued to the left and right main bronchi. Amyloid substances can be seen as protuberances in the left and right main bronchi, especially in the left main bronchus. Biopsy forceps were repeatedly used to extract the tumor, and argon knife was used to stop bleeding, and the lesions in trachea were cauterized by argon gas from the nozzle on the side. Compared with the first treatment, inflammatory exudates were significantly reduced, there were reduced inflammatory secretions in the lungs, and the secretions were aspirated. The patient had no hemoptysis after treatment, showed stable vital signs and was discharged on the third day after treatment. After discharge, the patient did not use oral or intravenous hormones and other drugs, clinical symptoms were stable, experienced occasional coughing and a small amount of white sputum, no hoarseness, no yellow sputum, no hemoptysis, and could walk one block and three stairs with no wheezing. The patient underwent two follow-up bronchoscopy examinations on July 3, 2018 and January 30, 2019 (Figs. 5 and 6, respectively), with no recurrence of the disease.

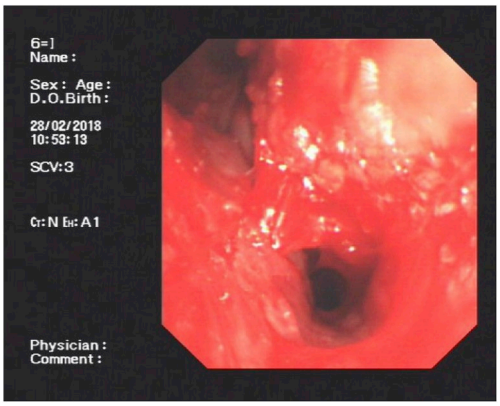

Fig. 2. On February 28, 2018, bronchoscopic examination revealed that the tumors invaded the tracheal carina and left and right main bronchi.

3. Discussion

Amyloidosis is a disease that causes structural and functional changes in many tissues and organs in the whole body because insoluble proteins are deposited between cells in the form of abnormal fibrous structures. The cause of the disease is unknown, with men having a higher incidence than women, but women get affected at an early age with a more severe disease course [2]. Although amyloidosis is a benign disease, there is still no cure, and has high recurrence and mortality rates [3]. Tracheobronchial amyloidosis is a rare type of amyloidosis, which can be divided into two subtypes, focal and diffuse, according to the scope of the lesion. The diffuse subtype is more serious and has a poor prognosis [4]. The case we reported herein, was that of diffuse tracheobronchial amyloidosis, the patient had made repeated visits for cough, yellow sputum, and repeated wheezing seven years before, but no definitive diagnosis was made. The reasons for the delay in diagnosis were analyzed; foremost, the rare incidence of amyloidosis in the respiratory tract [5]; and secondly, the diverse and atypical clinical manifestations of this type of amyloidosis led to the difficulty in clinical diagnosis of this disease. Tracheobronchial amyloidosis is often manifested as progressive cough, wheezing, dyspnea, recurrent pneumonia, and hemoptysis, and the clinical manifestations vary according to the location of the disease. For example, central airway involvement is mainly manifested as airway obstruction; moderate airway involvement is mainly manifested as lung collapse and repeated infection; and small terminal airway involvement is manifested as recurrent pneumonia and bronchiectasis. Due to the above reasons, the clinical misdiagnosis rate of tracheobronchial amyloidosis is high. Therefore, through

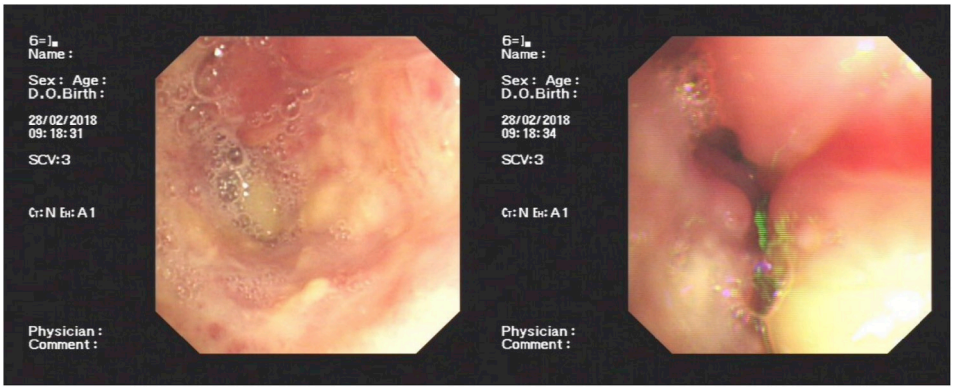

Fig. 1. On February 28, 2018, bronchoscopic examination revealed that the lumen was blocked by a lump 3cm below the glottis, resulting in 95% of the lumen stenosis and a large amount of secretions on the surface.

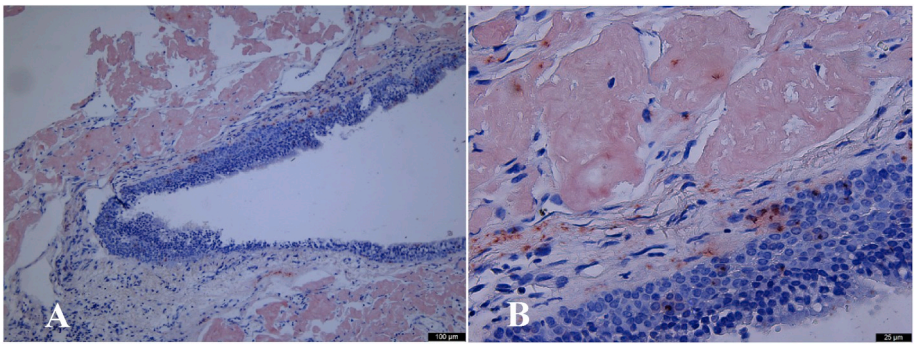

Fig. 3. (A) Biopsy of bronchial lesion showed amyloidosis (Congo red, ×100).(B) Biopsy of bronchial lesion showed amyloidosis (Congo red, ×400). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

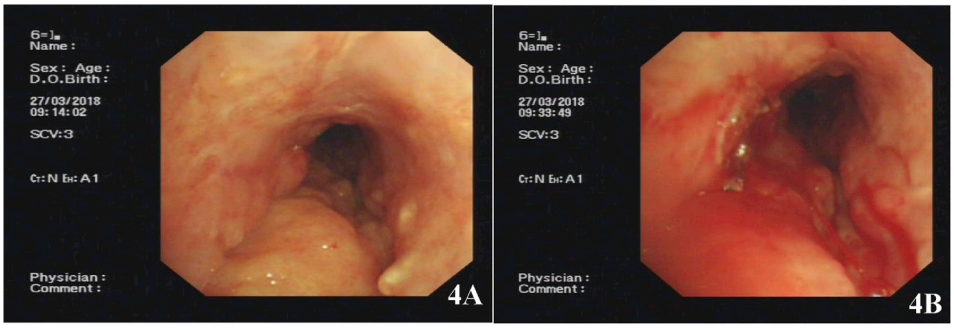

Fig. 4. (A) On March 27, 2018, before the second bronchoscopy treatment, bronchoscopic examination revealed that there was approximately 40% widespread stenosis from 3 cm below glottis till tracheal carina.(B) On March 27, 2018, after the second bronchoscopy treatment, bronchoscopic examination revealed that there was approximately 20% widespread stenosis from 3 cm below glottis till tracheal carina.

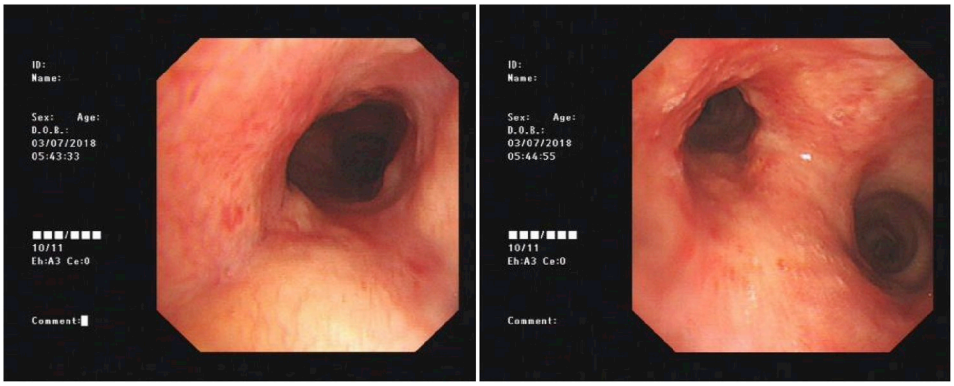

Fig. 5. Follow-up bronchoscopy images on July 3, 2018.

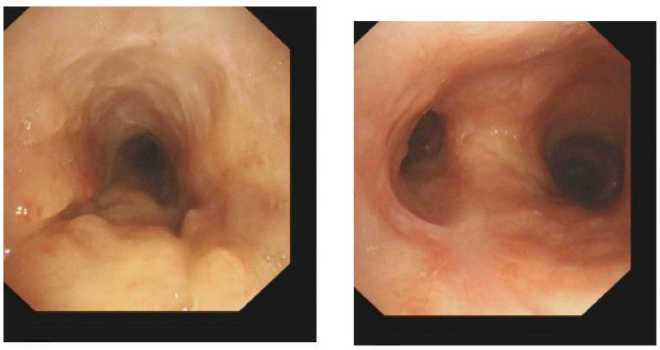

Fig. 6. Follow-up bronchoscopy images on January 30, 2019.

summarizing the diagnosis and treatment of this case, it can help clinicians to improve their understanding of tracheobronchial amyloidosis.

In recent years, with the development of bronchoscopy interventional therapy, a new effective treatment method has been provided to patients with central airway stenosis, especially severe airway stenosis with obvious dyspnea. Currently, the commonly used interventional treatment methods of bronchoscopy in clinic mainly include ablative technique and mechanical dilatation technique, and the treatment scope mainly includes central airway stenosis caused by benign and malignant diseases. In connective tissue diseases, lesions that can cause central airway stenosis include Wegener's granulomas, recurrent polychondritis, and systemic amyloidosis. For the central airway stenosis caused by these three diseases, bronchoscopy intervention therapy has some differences in specific methods employed in each of these diseases: currently, the commonly used treatment method for Wegener's

granuloma is bronchoscopic laser freezing resection combined with local hormone therapy [6]. However, as the lesion has relatively serious vascular and inflammatory necrosis, it may easily create a larger scar on the treatment site, leading to more severe stenosis. Moreover, it may easily cause serious edema of the airway mucosa, which can aggravate dyspnea or asphyxia. Central airway stenosis caused by recurrent polychondritis is currently treated by tracheotomy or stent placement. Although stent placement can rapidly relieve airway stenosis, there are many short-term and long-term complications, such as sputum retention, recurrent lung infection, and granulation tissue hyperplastic restenosis [7].Currently, the main interventional therapy for central airway stenosis caused by amyloidosis is cryoablation. Recent studies have suggested that in cryotherapy [8], if the lesion area resected by cryotherapy is large, the avulsion of lesion tissue may easily occur, which may lead to massive bleeding, and in the course of treatment, it may cause edema of the lesion tissue which can exacerbate airway stenosis, making breathing more difficult, and can even lead to asphyxiation and death.

We report a case that was treated using interventional therapy combined with bronchoscopic argon and cryotherapy under laryngeal mask anesthesia, the lesion tissue which caused over 95% obstruction of the lumen in the airway was treated twice by this technique. The reason we chose this combination method first, was because argon knife (argon plasma coagulation) therapy involves the argon ion beam produced by argon under the action of high frequency electric current. It uses its thermal effect to coagulate the pathological tissue. Because of the inertia of argon, it does not carbonize and vaporize the tissue in the treatment process, which is conducive to tissue repair. The treatment ensures both hemostatic effect and safety. Consequently, a frozen probe was introduced to cut the lesion site; this method can absorb the surrounding heat energy in a moment when the liquid CO_2 gas expands rapidly, and produces a cryogenic effect on the front end of the frozen probe, killing tissue cells, similar to mechanical excision of lesions [9]. Considering that it is easy to bleed when the tissue is avulsed after freezing cutting, argon knife is used timely to stop bleeding, which is conducive to the thorough removal of the lesion tissue, and to ensure that the damage to the surrounding tissues is very small during hemostasis. After the second treatment, the patient's condition was completely controlled. Two follow-up bronchoscopy examinations were conducted within one year of the treatment, and the airway was unobstructed without any signs of recurrence. Furthermore, the patient did not consume oral hormone and other amyloidosis drugs after treatment, and has achieved clinical cure.

After evaluating the therapeutic effect in this patient, we believe that argon knife combined with cryotherapy is a safe and effective method to relieve severe central airway stenosis caused by amyloidosis, with complete resection of lesion tissues, less intraoperative bleeding, less damage to normal tissues, and less recurrence after surgery. This method of treatment not only has the reliable curative effect, but also has the reliable security. We referred to a great deal of literature, and found no reports of patients with severe central airway stenosis caused by amyloidosis treated with argon knife combined with CO_2 cryoablation under laryngeal mask anesthesia. Therefore, our approach is innovative, it is a novel technique that is worth promoting, which will benefit patients with this disease, and it may be helpful to clinicians who are engaged in bronchoscopy intervention. However, this kind of treatment is difficult to perform, requires high clinical skills of anesthesiologists, and skilled and experienced respiratory interventional therapy physicians to operate.

4. Conclusion

Due to the rare occurrence of tracheobronchial amyloidosis and its atypical clinical symptoms, it is prone to be misdiagnosed. Therefore, pulmonary CT should be performed repeatedly and bronchoscopy should be performed at the earliest.

Bronchoscope guided argon knife combined with cryotherapy can achieve good efficacy and high safety for severe central airway stenosis.

Authors contributions

Lixue Jiang contributed to the conception of the study; Che Chunli and Dong Fushi performed the clinical operation and acquired the data. All authors have made significant contributions to the manuscript and have reviewed it before submission.

All authors have confirmed that the manuscript is not under consideration for review at any other Journal. All authors have read and approved the final manuscript.

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Declaration of competing interest

None of the authors have a financial relationship with a commercialentity that has an interest in the subject of the manuscript. No financial support was used for this case report.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2019.100948.

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