



Delayed Right Main Bronchus Stenosis With Involvement of the Tracheal Carina After Blunt Chest Trauma: Successful Treatment With Tracheobronchial Anastomosis

Estenosis tardía del bronquio principal derecho con afectación de la carina traqueal tras traumatismo torácico cerrado: un tratamiento exitoso mediante anastomosis traqueobronquial

Dear Editor,

Blunt chest trauma is a rare cause of acquired benign airway stenosis. Only 0.4–1.5% of these events induce airway injury, which mostly occurs in the right main bronchus and within 2 cm of the tracheal carina.¹ Half of all airway injuries are not identified during the first 24–48 h, because normal ventilation may be present despite the airway injury and because of non-specific symptoms and other injuries, such as head trauma, abdominal injuries, and multiple extremity fractures.¹ If airway injuries are not recognized early, an aberrant airway healing process can lead to the formation of granulation tissue and collagen deposition and, ultimately, to delayed tracheobronchial stenosis.² Although delayed tracheobronchial stenosis after blunt chest trauma is treated with bronchoscopic intervention (e.g., balloon dilation, laser ablation, and airway stents), surgical intervention, or a combination of these methods, no treatment consensus or guidelines have been established for this condition.^{2,3} The management of delayed tracheobronchial stenosis caused by blunt chest trauma highly depends on patient status, severity of symptoms, location and extent of stenosis, degree of distal pulmonary parenchymal destruction, and local expertise.² Here, we describe the case of a patient with delayed right main bronchus stenosis involving the tracheal carina after blunt chest trauma who was successfully treated with tracheobronchial anastomosis.

A 34-year old male current smoker had been involved in a motor vehicle accident at the age of 21. He suffered bilateral pneumothorax, which was treated only with chest drainage tubes. The patient developed pneumonia every 12 years starting at the age of 24. During the pneumonia episodes, he complained of wheezing and dyspnea. The patient did not have any lower respiratory tract infections during childhood. Recently, he started having recurrent obstructive pneumonia every 12 months, which led him to visit the Department of Respiratory Medicine of the local hospital. Chest computed tomography (CT) showed ground glass opacity and

consolidation in the right middle and lower lobes, and bronchial mucous retention with stenosis of the right main bronchus, the length of which was less than 1 cm (Fig. 1A, B). Flexible fiber-optic bronchoscopy (FOB) showed a fibrotic lesion causing almost complete stenosis of the proximal right main bronchus, with the exception of a pinhole located at the lateral posterior aspect of the stenosis (Fig. 1D). His forced vital capacity (FVC), forced expiratory volume in 1 s (FEV₁), FEV₁/FVC ratio, expiratory reserve volume (ERV), and residual volume (RV)/total lung capacity (TLC) ratio, were 92.4% (4.23 L), 61.4% (2.47 L), 58.39%, 76.8% (1.29 L), and 32.13%, respectively (Fig. 1F).

The patient was referred to our hospital for treatment of the right main bronchus stenosis. We selected surgical intervention (supplemental video). Under general single-lung ventilation with a double-lumen tube, we performed a right posterolateral thoracotomy and found that the lung had adhered to the chest wall and that the middle and lower lobes showed consolidation. We opened the right main bronchus at the proximal side; however, the stenosis remained, and we resected half of the tracheal carina and the lower part of the trachea. At the distal side, the main bronchus was removed to the smallest extent possible, which was approximately 2 cm. Anastomosis was performed with continuous suture for the mediastinal side and interrupted suture for the other part using 4-0 polydioxanone suture, and pedicled pericardial fat tissue was used as a cover. The patient was discharged on postoperative day 12 without any postoperative complications. Pathological analysis of the resected specimen showed the presence of granulation tissue with a focal fibrosis lesion and no evidence of malignancy.

Follow-up chest CT (Fig. 1C) and flexible FOB (Fig. 1E) performed 3 months postoperatively showed successful healing of the anastomosis with luminal patency. A follow-up pulmonary function test revealed improvement of the obstructive ventilatory defect based on FVC, FEV₁, FEV₁/FVC ratio, ERV, and RV/TLC ratio values of 101.3% (4.66 L), 93.3% (3.77 L), 80.9%, 98.8% (1.67 L), and 27.6%, respectively (Fig. 1G). The patient did not develop pneumonia for 10 months after the operation.

If stenosis involves the tracheal carina, surgical intervention may be associated with an increased risk because of technical difficulties and a high level of postoperative complications, such as anastomotic dehiscence and restenosis.^{4,5} Two previous studies showed that delayed airway stenosis with involvement of the tracheal carina after blunt chest trauma was successfully treated with repeated balloon dilation and stent placement.^{6,7} Thus, it

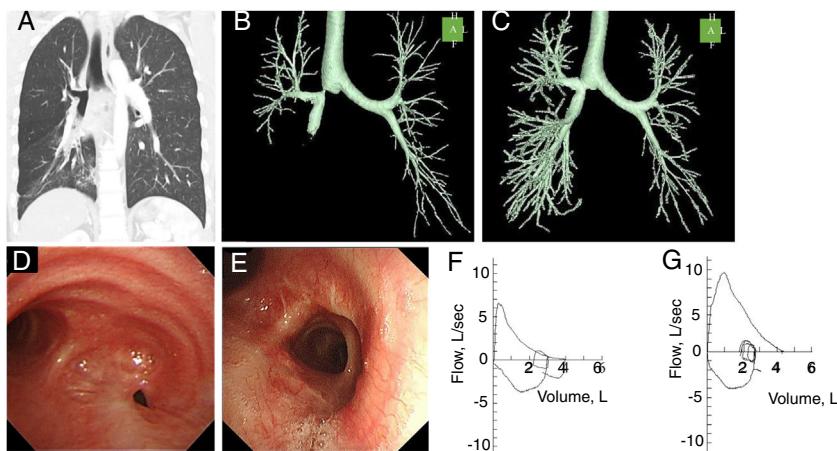


Fig. 1. Coronal chest CT image (A) and three-dimensional chest CT image (B) acquired before the operation and three-dimensional chest CT image acquired 3 months after the operation (C). Flexible fiber-optic bronchoscopic images before the operation (D) and 3 months after the operation (E). Flow-volume loops before the operation (F) and 3 months after the operation (G).

was considered that we should perform a bronchoscopic intervention; however, we decided on surgical intervention for the following reasons. First, we considered that we could perform tension-free anastomosis, because the stenotic lesion affected the main bronchus exclusively. Second, the stenosis did not induce the formation of a right lung abscess. Third, bronchoscopic treatments might require repeated interventions because of migration and formation of granulation and because of the recurrence of stenosis after stent removal.⁸ Fourth, the patient was young and his preoperative condition was good. In the present case, surgical intervention was an acceptable method for managing stenosis involving the carina. Previous studies reported no perioperative mortality by surgical intervention for benign airway stenosis.^{9,10} Therefore, surgical intervention should be considered in the future under the same condition.

The median time from initial injury to diagnosis of delayed tracheobronchial stenosis is 6 months.² Kiser et al. reported an absence of association between time to the diagnosis of delayed tracheobronchial stenosis and the rate of successful surgical repair.¹¹ Perioperative complications related to surgical intervention for tracheobronchial stenosis includes empyema, rebleeding, stenosis, and fistula.^{2,12} The current patient was diagnosed 13 years after the initial motor vehicle accident and experienced no perioperative complications. The long-term complications or long-term follow-up outcomes are unknown¹²; therefore, we should carefully investigate the future disease profiles of this case.

Here, we successfully treated a case of delayed airway stenosis involving the tracheal carina with surgical intervention. As the treatment consensus or guidelines for delayed airway stenosis after blunt chest trauma have not been established, a very careful clinical and radiological evaluation is needed to prevent peri- and post-operative complications before deciding on whether to adapt this technique.

Financial conflicts

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Conflict of interest

Satoshi Hamada reports grants from Teijin Pharma, outside the submitted work.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.arbres.2020.09.008.

References

- Shemmeri E, Vallières E. Blunt tracheobronchial trauma. *Thorac Surg Clin*. 2018;28:429–34.
- Glazer ES, Meyerson SL. Delayed presentation and treatment of tracheobronchial injuries due to blunt trauma. *J Surg Educ*. 2008;65:302–8.
- Ernst A, Feller-Kopman D, Becker HD, Mehta AC. Central airway obstruction. *Am J Respir Crit Care Med*. 2004;169:1278–97.
- Weder W, Inci I. Carinal resection and sleeve pneumonectomy. *J Thorac Dis*. 2016;8:S882–8.
- Wright CD, Grillo HC, Wain JC, Wong DR, Donahue DM, Gassert HA, et al. Anastomotic complications after tracheal resection: prognostic factors and management. *J Thorac Cardiovasc Surg*. 2004;128:731–9.
- DiBardino DM, Bramley K, Haas A. A 67-year-old woman with a “shrunken” left lung and recurrent pneumonia. *Chest*. 2018;154:e49–53.
- Roh JY, Kim I, Eom JS, Lee G, Ahn HY, Lee MK. Successful stenting for bronchial stenosis resulting from blunt airway trauma. *Intern Med*. 2018;57:3277–80.
- Murgo SD, Egressy K, Laxmanan B, Doblaré G, Ortiz-Comino R, Hogarth DK. Central airway obstruction: benign strictures, tracheobronchomalacia, and malignancy-related obstruction. *Chest*. 2016;150:426–41.
- Tsukioka T, Takahama M, Nakajima R, Kimura M, Inoue H, Yamamoto R. Efficacy of surgical airway plasty for Benign Airway Stenosis. *Ann Thorac Cardiovasc Surg*. 2016;22:27–31.
- Li J, Wang W, Jiang L, Yin W, Liu J, Shao W, et al. Video-assisted thoracic surgery resection and reconstruction of carina and trachea for malignant or benign disease in 12 patients: three centers' experience in China. *Ann Thorac Surg*. 2016;102:295–303.
- Kiser AC, O'Brien SM, Dettberbeck FC. Blunt tracheobronchial injuries: treatment and outcomes. *Ann Thorac Surg*. 2001;71:2059–65.
- van Roozendaal LM, van Gool MH, Sprooten RTM, Maesen BAE, Poeze M, Hulsewé KWE, et al. Surgical treatment of bronchial rupture in blunt chest trauma: a review of literature. *J Thorac Dis*. 2018;10:5576–83.

Satoshi Hamada ^{a,b,*}, Akihiro Ohsumi ^c, Tatsuya Goto ^c,
Masatsugu Hamaji ^c, Hiroshi Date ^c, Toyohiro Hirai ^d

^a Department of Advanced Medicine for Respiratory Failure, Graduate School of Medicine, Kyoto University, Kyoto, Japan

^b Department of Respiratory Medicine, Ayabe City Hospital, Kyoto, Japan

^c Department of Thoracic Surgery, Graduate School of Medicine, Kyoto University, Kyoto, Japan

^d Department of Respiratory Medicine, Graduate School of Medicine, Kyoto University, Kyoto, Japan

* Corresponding author.

E-mail address: sh1124@kuhp.kyoto-u.ac.jp (S. Hamada).

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A Case of Disseminated Tuberculosis in a Child with Crohn's Disease After Treatment with Azathioprine, Adalimumab and Ustekinumab



Un caso de tuberculosis diseminada en un niño con enfermedad de Crohn tras tratamiento con azatioprina, adalimumab y ustekinumab

We report the case of an 11-year-old girl with refractory Crohn's disease treated with ustekinumab, hospitalized for disseminated tuberculosis. This is the first case of disseminated tuberculosis in a child under anti-IL12 - IL23 treatment described in literature.

L. was diagnosed with Crohn's disease at the age of 7. She was first treated with oral corticosteroids for 9 months, thenaza-

thioprine. Because of persistent symptoms, anti-tumor necrosis factor agents (first infliximab during 3 years, and then adalimumab for 6 months) were added to azathioprine. The child had received BCG vaccination when aged 2 months. The tuberculin skin test was negative, and IGRAs (interferon gamma release assays = QuantiFERON-TB-Gold Cellestis) results were indeterminate before starting anti TNF agents. Abdominal pain, diarrhea, with evening rise fever were persistent despite anti TNF agents. One month after adalimumab was dropped when the child was eleven; ustekinumab treatment was started together with azathioprine, after a collective decision of pediatric and adult gastroenterologists (Fig. 1). No chest X-ray was performed before starting ustekinumab. It was started after 15 days of oral corticosteroids (budesonide 9 mg per day). Fifteen days after the first injection of ustekinumab