Case Report

Spontaneous Tracheal Rupture Associated with Acquired Tracheobronchomalacia

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We report here a very rare case of pneumomediastinum due to spontaneous tracheal rupture with tracheobronchomalacia. The patient was a 74-year-old woman who had suffered nocturnal dyspnea due to productive cough for five days prior to admission and had been treated with corticosteroids for five years at another hospital after being diagnosed with bronchial asthma. Computed tomographic scanning of the chest demonstrated over 1 cm longitudinal small air collections behind the upper trachea. Crescent-type tracheobronchomalacia was diagnosed by emergency bronchoscopy. At the right side of the upper trachea, a 1-cm laceration was revealed. Fibrin glue (Bolheal, Kaketsuken, Kumamoto, Japan) was sprayed on the laceration through an instrument of our design for endoscopic gluing and she was intubated for three days. Furthermore, treatment including administration of antibiotics, an antitussive agent, and a mucolytic agent, in addition to pulmonary physical therapy involving pursed lip breathing exercises and smoking cessation improved her complaints one month after admission. (Ann Thorac Cardiovasc Surg 2003; 9: 394–6)

Key words: acquired tracheobronchomalacia, pneumomediastinum, tracheal rupture

Introduction

We report here an extremely rare case of spontaneous tracheal rupture that, interestingly, was observed in a patient suffering from bronchial asthma and tracheobronchomalacea, and discuss the clinical features and treatment.

Case Report

A 74-year-old woman who had been smoking 20 cigarettes a day for 42 years was admitted to the hospital with nocturnal dyspnea due to productive cough that began five days before admission. She had been suffering from

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bronchial asthma for the past five years and had been treated with corticosteroids (predonisolone 5 mg/day). She did not present with any operation history. She was 158 cm tall and weighed 60 kg. Blood pressure upon admission was 150/70 mmHg, PaCO2 was 50 torr, PaO2 was 82 torr, and O2 saturation was 84% according to arterial blood gas analysis. Sibilant rhonchi over the upper fields of the chest was found by chest examination. Results from pulmonary function study were within normal limits, but a notch of a forced expiratory volume per second (FEV1) had been found by spirometry performed four months earlier at another hospital. Chest computed tomography (CT) revealed over 1 cm longitudinal small air collections behind the upper trachea (arrowheads in Fig. 1). There were no significant findings in the lung field and other mediastinum. Emergency bronchoscopy demonstrated an abnormal collapse of the trachea and main bronchi (Fig. 2), and crescent-type tracheobronchomalacia was diagnosed. The narrowing caliber was above 90% on coughing. At the right side of the edge of the membrane portion of the middle trachea, an approximately 1 cm longitudinal laceration was revealed. Therefore the air collections finding was judged as a result from a tear of the

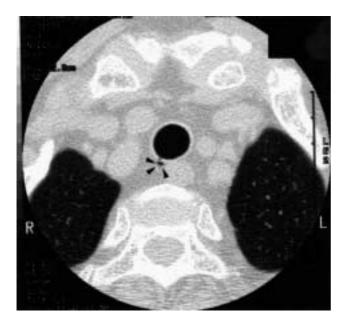


Fig. 1. Chest computed tomography revealed over 1 cm longitudinal small air collections behind the upper trachea (arrowheads).

tracheal membrane. Fibrin glue (Bolheal, Kaketsuken, Kumamoto, Japan) was sprayed into and on the laceration using an instrument of our design for endoscopic gluing and she was intubated with the cuff inflated distal to the tear for three days as a conservative treatment. Her sputum bacterial cultures grew Haemophilus influenzae

and Klebsiella pneumoniae. A fungal culture of the bronchial aspirate was weak positive. Furthermore, antibiotics, an antitussive agent, and a mucolytic agent were administered. After extubation of the tracheal tube, pulmonary physical therapy involving pursed lip breathing exercises and smoking cessation improved her complaints one month after admission. CT scan confirmed that the air collections had disappeared and the tear had changed to a scar when she was discharged from our hospital.

Discussion

This is a very rare case of spontaneous tracheal rupture of a patient with tracheobronchomalacia. To our knowledge, there have been no reports of a case like this one in the past. All cases of tracheal rupture reported so far were iatrogenic, like overdistention of a tube balloon and a balloon herniation of an intubated patient, or post-traumatic thoracic injury. The rupture was located at the edge of the membranous portion, therefore we assume that the rupture was caused by intratracheal high pressure due to severe cough, and the weakness of the tracheal tissue due to tracheobronchomalacia and continuous corticosteroid treatment. Nocturnal dyspnea due to productive cough, the chief complaint of this case, was most likely caused by acute tracheal rupture and the induced tracheal inflammation with tracheobronchomalacia.

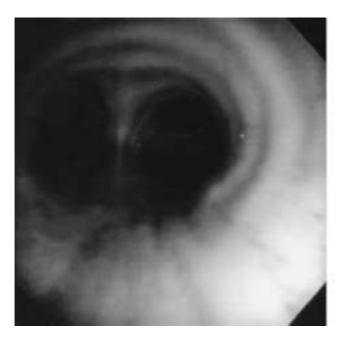




Fig. 2. Emergency bronchoscopy demonstrated an abnormal collapse of the trachea and main bronchi (right, inspiration; left, on coughing).

On the other hand, weakness of the membranous trachea often seen in woman and the elderly, and chronic obstructive pulmonary disease or steroid therapy following inflammation are also plausible explanations for this case. The patient had a history of corticosteroid treatment because of bronchial asthma for five years, thus it is possible that the medication led to and accerlated the weakening of tracheobroncheal tissue in addition to age-related regressive changes. In many cases, tracheobronchomalacia is diagnosed as chronic bronchitis, and dyspnea is treated as asthma.2) Early examinations are important for diagnosing and selecting suitable treatments for these symptoms. As for spirometry findings, a low FEV₁/ a forced inspiratory volume per second (FIV₁) and notch in FEV1 are suggestive of tracheobronchomalacia and an indication for bronchoscopy.³⁾

Tracheal rupture is very difficult to diagnose at the first examination. As an initial examination, CT is very sensitive for detecting tracheal injury such as pneumothorax and pneumomediastinum. However, it is essential to utilize a flexible or rigid bronchoscope to obtain early confirmation. The CT of the present case revealed only localized air collections, therefore tracheal rupture would not have been detected without performing flexible bronchoscopy. It is very important not to hesitate to perform bronchoscopic examination as early as possible if some abnormalities of tracheobronchial trees are suspected.

The therapy for tracheal rupture is still controversial, mainly because the selection between early surgical repair or conservative treatment is difficult. Gabor et al.⁵⁾ who had experienced 31 patients with tracheobronchial ruptures concluded that conveniently localized short lacerations can be treated with antibiotics and intubation with the cuff inflated distal to the laceration, especially if the case did not involve the whole thickness of the tracheal

wall but the majority of the cases of tracheobronchial ruptures had indications for prompt surgical repair. In the present case, the length of the tracheal longitudinal laceration was very short, and severe urgent clinical signs such as hemoptysis, asphyxiating dyspnea, subcutaneous emphysema, uni- or bilateral pneumothorax or severe pneumomediastinum containing deep cervical air were not observed. Moreover she was elderly, therefore we carried out a conservative treatment at first. If the pneumomediastinum had worsened after extubation, operative repair must have been performed. Fortunately, however she recovered uneventfully by undergoing only conservative treatment.

In conclusion, spontaneous tracheal rupture must be carefully considered as a significant complication of tracheobronchomalacia or corticosteroid treatment. If it is clinically suspected, CT and bronchoscopic study must be performed as early as possible.

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