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## Spontaneous Repetitive Tracheal Rupture

### Spontan Tekrarlayıcı Trakea Yırtılması

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**ABSTRACT** Tracheal ruptures are one of the life-threatening situations in critical care unit and can be formed after trauma, intubation, tracheotomy, bronchoscopy or spontaneously. Significant risk factors for tracheal rupture include urgent intubation, use of stilet, high cuff pressure, recurrent intubation attempts, double lumen endotracheal tubes, very thick endotracheal tubes, female gender, short height, older age, tracheomalacia, tracheal stenosis, congenital tracheal abnormalities and chronic steroid therapy. Among these risk factors, corticosteroids can cause rupture by inhibiting collagen synthesis and reducing connective tissue stability. In this case report, a case with spontaneous recurrent tracheal rupture was defined and the underlying mechanisms and therapeutic approach to this complication that could be seen in patients using chronic steroids were discussed.

**Keywords:** Tracheal rupture, corticosteroid, treatment

**ÖZ** Trakea yırtılması, yoğun bakım ünitelerinde hayatı tehdit eden bir durum olup, travma, entübasyon, trakeotomi, bronkoskopi sonrası veya kendiliğinden oluşabilir. Acil entübasyon, stilet kullanımı, kaf basıncı yüksekliği, tekrarlayan entübasyon girişimleri, çift lümenli endotrakeal tüpler, çok kalın endotrakeal tüpler, kadın cinsiyet, kısa boy, ileri yaş, trakeomalazi, trakea stenozu, konjenital trakea anomalileri ve kronik steroid tedavisi trakea yırtılması için önemli risk faktörleridir. Bu risk faktörlerinden kortikosteroidler, kollajen sentezini inhibe ederek ve bağ doku stabilitesini azaltarak yırtılmaya neden olabilmektedir. Bu olgu sunumunda, kendiliğinden tekrarlayan trakea yırtılması olan bir olgu tanımlanmış ve kronik steroid kullanan hastalarda görülebilecek bu komplikasyonla ilgili altta yatan mekanizmalar ve terapötik yaklaşım tartışılmaya çalışılmıştır.

**Anahtar Kelimeler:** Trakea rüptürü, kortikosteroid, tedavi

## Introduction

Tracheal ruptures are one of the rare emergency condition in critical care patients (1). Tracheal ruptures can be formed by cervical/thoracic trauma or iatrogenic (intubation, tracheotomy, bronchoscopy, stent operations, esophagectomy, positive end expiratory pressure) or spontaneously (2,3).

Spontaneous tracheal ruptures are reported by different authors in children and adults which had lung diseases, persistent cough, vomiting and chronic steroid usage (1,4-9).

Corticosteroids have been widely used in the treatment of chronic obstructive lung disease (COPD) because of the anti-inflammatory effects on the airways (10). The corticosteroids had many side effects including truncal obesity, acne, hyperpigmentation, hirsutism, striae, hypokalemia, myopathy,

glucose intolerance, pancreatitis, mood alterations, insomnia, psychosis, headache, intracranial hypertension, hypertension, osteoporosis, adrenal insufficiency, gastrointestinal diseases, hyperlipidemia, weakness of connective tissues and myopathy (11,12).

The corticosteroids decreased the hydroxyproline production, thus an inhibition on fibroblast growth and collagen synthesis which leads abnormalities on structure of the connective tissues. Also, long term corticosteroid administration induces myopathy with different pathways (11,12).

In the literature repetitive spontaneous tracheal rupture was not reported. The authors report a case in which the repetitive spontaneous tracheal rupture in a patient with COPD and bronchiectasis.

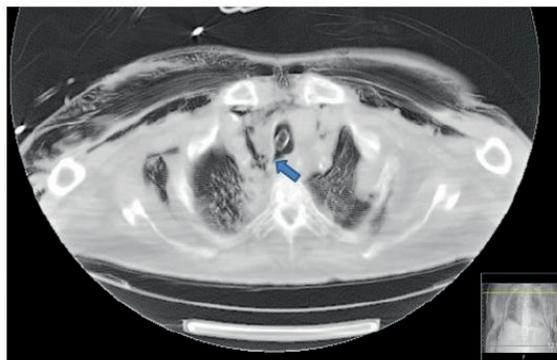
## Case Report

A 84-year-old, 80 kg, man with COPD and bronchiectasis was admitted to our clinic with progression of dyspnea and cough for last one week. He has a history of COPD and bronchiectasis for about fifty years. He had methylprednisolone usage for about twenty years. Examination revealed cyanosis and tachypnoea. Chest examination revealed bilateral crepitation and rhoncus. In the chest graphy bilateral diffuse infiltrations and bronchiectasis was seen. No pneumomediastinum and pneumothorax was detected. Because of severe hypoxemia endotracheal intubation was done by using a 7.5 mm internal diameter polyvinyl made endotracheal tube without any complications. After intubation the pressure of the cuff was set to 20-30 cmH<sub>2</sub>O by using an endotracheal tube cuff manometer (Cufflator™, Posey Company, USA). Afterwards mechanical ventilation therapy was started with 50% fraction of inspired oxygen, 500 mL tidal volume and 3 cmH<sub>2</sub>O ekspirasyon sonu pozitif basınç (PEEP). Intravenous dexmedetomidine hydrochloride (Precedex®, Meditera) with a dose of 0.2-1.4 mcg/kg/hr was started for harmony with the mechanical ventilation. Iv piperacillin and tazobactam sodium and methylprednisolone (1 mg/kg) was given to patient. On the second day the sedation was stopped, on the same day suddenly subcutaneous emphysema of the neck and right side of the chest was detected without any change on the arterial blood gases and vital signs. The PEEP decreased to 0 cmH<sub>2</sub>O. An urgent chest radiography confirmed subcutaneous emphysema, pneumomediastinum and pneumothorax in the right lung. In the thorax computed tomography (CT) subcutaneous emphysema, pneumomediastinum and a rupture on the right posterolateral tracheal wall in 4 cm above carina was detected (Figure 1). Skin incision was done for decompression of the subcutaneous emphysema and tube thoracostomy was used for the pneumothorax. Sedation was started again. On the third day after a sudden repetitive increase on subcutaneous emphysema another CT scan was done and 1 cm above the level of previous rupture another rupture on the posterior tracheal wall was detected (Figure 2). The same treatment was continued. On the 8. day the patient was died due to septic shock.

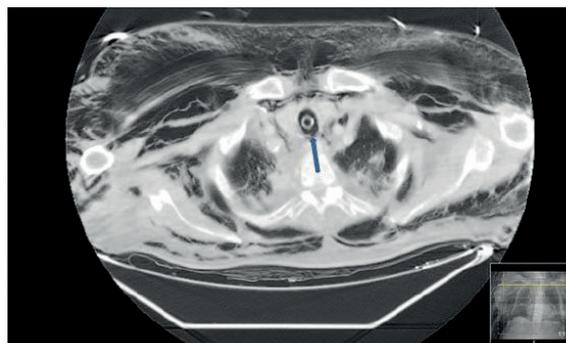
## Discussion

In this case report, we describe repetitive tracheal rupture in a patient with COPD and bronchiectasis.

The tracheal rupture was generally secondary to trauma or iatrogenic. The major reason of iatrogenic tracheal



**Figure 1.** Arrow: the rupture on the right posterolateral tracheal wall



**Figure 2.** Second rupture on the posterior tracheal wall 1 cm above the level of previous rupture

rupture was endotracheal intubation. The incidence of tracheobronchial rupture after intubation is very rare and reported as 0.01% (2). The risk factors for post-intubation tracheal rupture were emergency intubation conditions, stylet use, over inflation cuff pressure, repeated intubation attempts, double-lumen endotracheal tubes, too thick endotracheal tubes, female gender, short height, older age, tracheomalacia, tracheal stenosis, congenital tracheal anomalies and chronic treatment with steroids (2,7-13). In our patient no open or close trauma, congenital tracheal anomalies and tracheal stenosis history was detected and also intubation was uneventful. Intubation was done in only one attempt in optimal conditions by using a 7.5 mm internal diameter endotracheal tube. No pneumomediastinum and pneumothorax was detected in the first chest graphy.

In the literature only a few case reports were reported about spontaneous tracheal rupture. Kumar et al. (1) reported spontaneous tracheal rupture in a patient with interstitial lung disease. After chronic cough attack; subcutaneous emphysema and pneumothorax were detected. In another case report the authors reported spontaneous tracheal rupture after severe coughing in a 3-year-old boy (4). Both of the authors concluded that sudden increase in intratracheal pressure caused by severe coughing may cause tracheal

rupture. Stevens et al. (7) reported spontaneous tracheal rupture in a 14 years old girl due to vomiting. The authors concluded that this situation was due to violent vomiting because of diabetic ketoacidosis. Our patient had cough before intensive care unit (ICU) but in the first chest graphy no pneumomediastinum and pneumothorax was detected.

Farooqui et al. (3) reported the harmful effects of PEEP on tracheal injuries, but in our case we used only 3 cmH<sub>2</sub>O PEEP thus we thought that the PEEP had no effect on tracheal injury.

In our case, no iatrogenic trauma, cough, vomiting and high PEEP were detected, only had a history of chronic steroid usage so we think that this situation may be due to the chronic steroid usage. The corticosteroids decreased the hydroxyproline production, inhibited fibroblast growth and collagen synthesis (10). The corticosteroids had also adverse effects on the repair process of the airway epithelial cells. Although the molecular mechanisms remains unclear studies reported the depression of migration and proliferation of airway epithelial cells by steroids (10). Another harmful effect of steroids was myopathy. In skeletal muscle, steroids decreased the protein synthesis and increased protein destruction (11,12). As a result of muscle wasting difficulty ventilation, fatigue and decreased quality of life were occurred (12). Different studies reported the harmful effects of steroids on connective tissue, airway epithelial cells and muscle fibers (10-12).

In the literature a few steroid-dependent tracheal rupture was found (8,9) but no repetitive tracheal ruptures was detected. Zettl et al. (8) reported a 82 year old woman with a tracheal rupture following intubation. The patient had a history of chronic steroid usage for chronic polyarthritis. The authors detected the older age and chronic steroid usage as two main risk factors for this patient. In another case report (9) the authors reported a 67 years old woman with a spontaneous posterior tracheal wall rupture following a cough. The patient had a history of chronic steroid usage and the authors concluded that connective tissue weakness due to long term corticosteroid therapy and postoperative severe cough was the reason for this situation. In our patient we think that chronic steroid usage made a structural slimming on tracheal epithelial tissue and repetitive tracheal ruptures were occurred.

The most common signs of tracheal rupture were subcutaneous emphysema, pneumothorax and pneumomediastinum (4-6). Massive subcutaneous

emphysema compresses thoracic outlet, caused airflow obstruction and perfusion abnormalities to the head. Also subcutaneous emphysema caused chest wall rigidity and leads to high airway pressure (13,14). In our case, the subcutaneous emphysema was tracking from eyelids to the middle of sternum.

Urgent interventions of tracheal rupture should include X-ray graphy, fibreoptic bronchoscopy and thorax CT (1,2). Determining the correct place of the laceration will help for future therapy options. In our case we used chest X-ray and finally thorax CT for detecting the rupture.

A conservative therapy was used for a rupture length of less than two cm, partial tracheal wall rupture, rupture located in the cranial 2/3 of the trachea, patients with poor biological condition and high operatory risk. Cervicotomy or cervical mediastinotomy, thoracotomy, endoscopic techniques, skin incisions, subcutaneous drains and tracheostomy were used for surgery (1,2,14-16). In our patient we only used skin incisions and tube thoracostomy.

A delay in detecting tracheal rupture caused mediastinitis, impaired pulmonary functions and airway stenosis (1,2,14-17).

## Conclusion

In conclusion, we think that this was the first report of spontaneous repetitive tracheal rupture. Patients with sudden occurred or increased subcutaneous emphysema, pneumothorax or pneumomediastinum, and a history of chronic steroid usage repetitive tracheal injuries must be remembered and patients should be treated appropriately.

## Ethics

**Informed Consent:** Patient consent was obtained.

**Peer-review:** Externally peer-reviewed.

## Authorship Contributions

Surgical and Medical Practices: M.T.İ., D.M., N.T., Ş.Ç.U., A.Ş.U., H.B., F.Y., Concept: M.T.İ., D.M., Design: M.T.İ., D.M., Data Collection or Processing: M.T.İ., D.M., Analysis or Interpretation: M.T.İ., D.M., Literature Search: M.T.İ., D.M., Ş.T., Writing: M.T.İ., D.M.

**Conflict of Interest:** No conflict of interest was declared by the authors.

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