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Case report

A rare case of fibrostenotic endobronchial tuberculosis of trachea

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HIGHLIGHTS

- This is the only case of ETBT reported in the US.
- How to diagnose a unusual case of tracheal obstruction.
- How to treat a case of fibrostenotic endobronchial tuberculosis by repeated balloon dilation, and follow up.

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ABSTRACT

Endobronchial tuberculosis (EBTB) is a sequelae of pulmonary tuberculosis (TB) that extends to the endobronchial or endotracheal wall causing inflammation, edema, ulceration, granulation or fibrosis of mucosa and submucosa. This case depicts a 20 year old foreign-born woman with a history of active pulmonary TB on anti-TB chemotherapy, who presented with worsening stridor, dyspnea, cough and weight loss. The disease state was diagnosed with multiple modalities including, spirometry, CT scan of the neck, and bronchoscopy. The biopsies of the tracheal web revealed fibrotic tissue without any granulomas or malignancy establishing the diagnosis of EBTB. Serial balloon dilations and anti-neoplastic therapy with Mitomycin C was used to accomplish sufficient airway patency to relieve her symptoms. ETBT is a rare consequence of TB, which although has a low incidence in the United States, so physicians should have a high clinical suspicion based on the need for prompt intervention.

1. Introduction

Tuberculosis (TB) is relatively a rare disease in the United States with the rate of reported cases of 3.0 cases per 100,000 persons in 2013, which represents a decline of 4.3% compared to 2012. According to Centers of Disease Control and Prevention, 65% of the reported TB cases occurred among foreign-borne persons. Although there have been many case reports and studies in Japan, Korea and India of endobronchial tuberculosis (EBTB), the prevalence of EBTB in the United States is extremely low and no reported cases were discovered upon the literature review. We report a case of EBTB in a young female with active pulmonary TB on antituberculosis chemotherapy who presented with shortness of breath and stridor.

2. Case report

A 20 year old Vietnamese female presented to the emergency department (ED) with shortness of breath, cough, hoarseness, and significant weight loss. She was recently diagnosed with active pulmonary TB four months ago, and was started on anti-TB therapy, including Rifampin, Pyridoxine, Pyrzinamide, Isonaizid and Ethumbutal. Despite being complaint with the therapy, she frequently presented to ED with dyspnea and was treated with bronchodilators and a short course of steroids for the presumptive diagnosis of reactive airway disease secondary to anxiety ("anxiety asthma").

On her current presentation, she was afebrile with stable blood pressure of 129/84 mmHg, pulse of 102 beats per min, respiratory rate of 18 breaths per min with 96% saturation on room air. Her physical examination was remarkable for tachycardia. The lung examination revealed labored breathing with the use of accessory muscles, biphasic stridor, diminished breath sounds and mild expiratory wheezes in all lung fields. Laboratory evaluation was unremarkable with normal white blood cell count. Chest X-ray showed focal rounded airspace opacity in the left upper lobe

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consistent with the previous evaluations. CT-scan of neck revealed previous cavitating lesions in the left apex but no suspicious neck abnormality, mass, collection or adenopathy in the neck soft tissues (Fig. 1). Initially in the ED, she was treated with intravenous methylprednisolone and was placed on albuterol 10 mg/h continuous nebulizer treatments.

She was admitted for an upper airway obstruction secondary to laryngeal edema or vocal cord dysfunction. She was placed on bronchodilator nebulizing treatments as needed and humidified oxygen along with a dose of racemic epinephrine with minimal improvement in her symptoms. Consequently, a direct laryngoscopy was done that was negative for any focal spasm, polyps, edema or vocal cord malfunction. A trial of lorazepam was given to rule out psychogenic stridor caused by emotional stress, that failed to relieve her symptoms. Bronchoscopy revealed 90% occlusion of the trachea with a small pin hole opening and tracheal web approximately 3 cm below the vocal cords (Fig. 3). CT-scan performed at the admission was re-evaluated, which showed the tracheal stenosis that was missed at the initial reading (Fig. 1). Cardiothoracic surgery was consulted at this point for balloon dilation. The biopsies of the tracheal web showed fibrosis but no granulomas or malignant tissue. Following the procedure, she was discharged home two days later in a stable condition with outpatient otorhinolaryngology and cardiothoracic surgery follow up appointments.

Three months later, she presented to the emergency department with similar symptoms associated with fever and chills. Along with antibiotics, a repeat balloon dilation was performed for tracheal stenosis, and she was discharged upon resolution of her symptoms after two days.

Three months after repeat tracheal balloon dilation, she was noticed to have labored breathing associated with chest tightness on her regular otorhinolaryngology follow up visit. The decision was made to perform suspension micro-laryngoscopy with CO_2 laser incision of the tracheal stenosis. Also Mitomycin C, an antineoplastic agent, was applied to the area to prevent re-stenosis. She was discharged home in a stable condition after observation and did not have a recurrence of the symptoms again. The biopsy of the tracheal web revealed the same findings. The patient has not returned to the hospital for repeat dilations in twelve months.

3. Discussion

In a large prospective study done outside the United States, Jung et al. reported that among the patients with active pulmonary tuberculosis, 50% or more have EBTB [4]. However, our literature search we were unable to find any cases documented in the United States Due to the rarity of EBTB, and the paucity of the reported cases in the United States, EBTB cases are commonly misdiagnosed as one of the complications of tuberculosis as obstructive pneumonia, obstructive airway disease, such as bronchial asthma [5], or lung cancer [6]. Furthermore, the diagnosis is frequently delayed due to low suspicion of EBTB, since the tracheal stenosis in adult population in the United States is commonly due to trauma (i.e intubation), malignancy, lymphadenopathy, or granulation tissue. Lee et al. reported that the incidence of tracheobronchial stenosis is 68% in the first 4–6 months and may rise further if the course of disease is elongated. Also, Jung et al. concluded that the main predictors of concomitant EBTB are female gender and symptom duration of more than 4 weeks. Although the reasons for the female predominance are unclear, some studies have pointed that since females have thinner tracheobronchial lumens, they have weaker expectoration capacity than males, which may make females prone to direct implantation of *Mycobacterium tuberculosis* within the expectorated phlegm from pulmonary focus [16]. Furthermore, Jung et al. reported that the severe bronchostenosis (luminal narrowing of one to two thirds) is more frequent in females than males (37.3% vs 20.7%; $p = 0.009$). These findings are consistent with our case.

The clinical presentation of EBTB is nondistinctive as the common chief complaints include cough, expectoration, hemoptysis, wheezing, fever and felling short of breath [3]. Our patient presented with dyspnea and stridor, which could be explained by the site of involvement of EBTB. The previous studies have shown that the most common sites for EBTB are right upper lobe and right main bronchus [9], but our patient had a rare presentation of tracheal fibrostenosis, which explains the unique presentation of stridor. Systemic symptoms related to TB such as anorexia, weight loss, and night sweats might not be prominent in EBTB [10]. Pulmonary examination usually shows diminished breath sounds, rhonchi and localized moist rales [3]. The white blood cell count is usually normal as seen in our patient; though lymphocytes might be increased. Some studies have suggested that C-reactive protein is significantly increased and is related to the severity of the disease [11]. Previous case reports and studies have shown that about 10–20% of patients with EBTB have normal chest radiological findings [3]. CT scan may reveal the extent of tracheobronchial lesions, and the location of stenosis, although limited to the expert's ability and experience as seen in our case. This again reinforces the value of maintaining a high index of suspicion for EBTB in a patient with history of pulmonary tuberculosis presenting with obstructive signs and symptoms.

Based to the different classifications of EBTB, patients can have an active sub-type or a fibrotic disease. Both the active form and the fibrostenotic EBTB have markedly different CT findings. Patients with active disease show irregular bronchial luminal narrowing with wall thickening, and enlarged adjacent mediastinal lymph nodes due to hyperplastic changes and inflammatory edema. In contrast, the fibrostenotic subtype of EBTB shows smooth narrowing of the tracheal or bronchial lumen with minimal wall thickening. They also do not have the enlarged adjacent lymph nodes, as seen in our patient. Since the clinical presentation is often misdiagnosed as obstructive phenomenon, previous studies have suggested spirometry to evaluate patient's lung function. The flow-volume loop shows (fixed) blunting of the inspiratory and

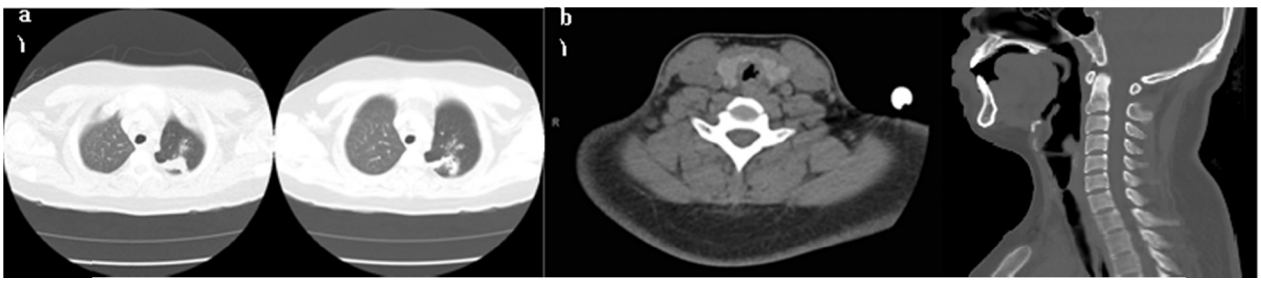

Fig. 1. a) CT-scan of chest showing micro-cavitations in the left upper and lower lobes. b) CT scan of neck showing tracheal narrowing.