Case report

Laryngeal involvement in a patient with active postprimary tuberculosis: Case report of a rare extrapulmonary manifestation☆✩✩,✩✩★

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A B S T R A C T

A 43-year-old woman was found to have active post-primary tuberculosis and a lateral neck radiograph showing a thickened epiglottis. Bronchoscopy-guided biopsies of the epiglottis and lung were acid fast bacilli stain positive. Histopathology from both showed multiple caseating granulomas. The patient’s condition improved with RIPE therapy. This case illustrates the importance for physicians to be aware of possible laryngeal involvement in tuberculosis and that it can present even without evidence of active or latent tuberculosis.

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Introduction

Laryngeal tuberculosis (TB) is a rare manifestation of extrapulmonary TB. However, with the global rise in incidence of TB, the importance of being aware of the possibility of laryngeal TB cannot be understated. The clinical presentation of laryngeal TB is largely nonspecific, which can include dysphonia, weight loss, fever, cough, and odynophagia, and thus, can be mistaken for laryngeal carcinoma. Given its vague presentation and contagious nature of TB, prompt diagnosis should be made to avoid delay of treatment.

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Case summary

A 43-year-old woman without significant past medical history presented to the emergency department with progressively worsening productive cough, fever, and sore throat for 2 months. She also reported hoarseness and unintentional weight loss of 20 pounds during this time. She denied night sweats, hemoptysis, chills, chest pain, and dyspnea. The patient emigrated from Brazil about a year ago and is currently living with her family. She denied any recent travel or exposure to tuberculosis (TB). No past history of pneumonia or TB. She is a nonsmoker. Physical examination was notable for a temperature of 39.3 C, bilateral rhonchi, mild wheezes, and 2 enlarged (1 cm) lymph nodes, that were nontender, mobile, and located on the right posterior cervical chain. No pharyngeal exudates or erythema were appreciated. WBC was 13,200/mcL and platelet was 706,000/mcL.

Sputum gram stain showed gram negative rods and gram positive cocci in chains. Chest radiography revealed multifocal pneumonia and right upper lung cavitation (Fig. 1A). Computed tomography (CT) thorax with contrast revealed a large cavitary lesion in the left upper lobe, as well as smaller cavitary lesions, multiple patchy consolidations and ground glass opacities in bilateral lungs (Fig. 1B-E). The CT scan findings were concerning for active post primary tuberculosis. Lateral neck radiograph revealed a lobulated thickened epiglottis, concerning for epiglottis involvement as well (Fig. 2). On bronchoscopy, the epiglottis appeared thickened and irregular with pale mucosa and thick purulent secretions in the bilateral mainstem bronchi. Bronchoscopy guided biopsies of the epiglottis and lung were acid fast bacilli stain positive but periodic acid-Schiff and Grocott’s methenamine silver stains

Fig. 1 – (A) Chest radiograph showing bilateral multifocal pneumonia, with large left upper lobe cavity. (B, C, D) CT scan coronal, sagittal and axial lung window showing large left lung cavitary lesion, multiple smaller cavitary lesions, multifocal patchy consolidations. (E) CT contrast enhanced scan axial mediastinal window showing cavitary lesions, consolidation. Note marked lack of subcutaneous fat representing cachexia.

Fig. 2 – Lateral soft tissue neck radiograph showing a lobulated, enlarged epiglottis (white arrow).
negative (Fig. 3). The histopathology from both epiglottis and lung revealed multiple caseating granulomas (Fig. 4). Patient was started on RIFE therapy and was discharged several weeks later after clearance from the Department of Public Health. On follow up at 7 weeks, she had no complaints, cough had resolved and she gained some weight. Also, the chest was clear on auscultation.

**Discussion**

Laryngeal TB is a rare and often overlooked manifestation of extrapulmonary TB, with its current incidence estimated to be less than 1% of all TB cases [1]. In the early 1900s, laryngeal TB had been one of the most common diseases of the larynx [2], but its incidence dropped dramatically after the 1950s with the introduction of anti-TB drugs, improvements in living standards, and preventative measures [3]. However, there has been a rise in incidence in the last several decades that could be attributed to rises in TB, HIV, and immunosuppression (related to diseases or therapy) [3,4]. Thus, it is important for physicians to be conscious of laryngeal TB and its presentation so that diagnosis and treatment are not delayed.

In terms of clinical presentation, it appears that the major symptoms of laryngeal TB have changed. Bailey et al. [5] reported that constitutional symptoms such as fever, weight loss, night sweat, fatigue, and hemoptysis had been the major symptoms in the past due to progression from pulmonary TB. Lim et al. [6] found that constitutional symptoms are becoming rarer and that hoarseness, odynophagia, and dysphagia are now the major symptoms of laryngeal TB.

However, in Benwill and Sarria’s review of 106 reported laryngeal TB cases in the US, they found that dysphonia (hoarseness) was the most common symptom (96%), followed by weight loss (47%), cough (38%), dysphagia (26%), and odynophagia (25%) [7]. This is somewhat consistent with our case, where laryngeal TB presented with dysphonia and odynophagia, but also with systemic symptoms of fever and weight loss.

Multiple methods have been used to confirm the diagnosis of laryngeal TB. In Benwill and Sarria’s review, they found that sputum culture provided the diagnosis in 70% of cases and laryngeal tissue histopathology in 41% [7]. They suggest that sputum acid-fast bacilli smears and cultures should be obtained in all patients with suspected laryngeal TB. Chest radiographs should also be obtained as most cases of laryngeal TB are secondary to pulmonary TB. Their review found
evidence of pulmonary TB in 86% of cases. In cases where cultures and/or radiographs are negative and patient presents with unexplained odynophagia and weight loss, laryngoscopy with tissue biopsy may be necessary to confirm laryngeal TB or exclude other pathologies, with the main diagnostic clue being laryngeal carcinoma. The presence of caseating granulomas or isolation of TB in tissue culture would make laryngeal TB definitive. Some imaging findings that suggest laryngeal TB over laryngeal carcinoma include a bilateral diffuse pattern of involvement vs local; intact laryngeal architecture; and focal low-attenuation areas in the lesion, suggestive of caseating necrosis [8].

It is important to note that while laryngeal TB is most commonly due to progression from pulmonary TB, it can also manifest without lung involvement. In a retrospective review of 22 cases of laryngeal TB from January 1990 to December 1998 in Asan Medical Center in Seoul, Korea, Shin et al. [9] reported laryngeal TB in 9 patients with normal chest radiographs (41%). In a retrospective review of 60 cases of laryngeal TB at Severance Hospital in Seoul, Korea from 1994 to October 2004, Lim et al [6] found laryngeal TB in patients with normal lung status in 12 cases (20%). Of those cases, sputum test or PCR was positive in 3 cases. So, they found primary laryngeal TB in 9 (15%) of all cases. These studies highlight the importance of keeping laryngeal TB on the differential even when radiographs suggest otherwise.

RIPE therapy is the first-line treatment for laryngeal TB and has shown to lead to excellent responses and resolution of laryngeal lesions in 4-8 weeks [10]. Prompt diagnosis and treatment are crucial as untreated laryngeal TB could lead to irreversible laryngeal stenosis or cricoarytenoid fixation [8].

In conclusion, with the rise in incidence of TB and along with laryngeal TB, it is important for physicians to be aware of its varying clinical presentations. Physicians must also recognize that laryngeal TB can present without evidence of active or latent TB.

Patient Consent

Informed consent was obtained from the patient.

References

[1] Uslu C, Oysu C, Uklumen B. Tuberculosis of the epiglottis: a case report. Eur Arch Otorhinolaryngol 2008;265(5):599–601.
[2] Auerbach O. Laryngeal tuberculosis. Arch Otolaryngol 1946;44:191–201.
[3] Rizzo P8, Da mosto MC, Clari M, Scotton PG, Vaglia A, Marchiori C. Laryngeal tuberculosis: an often forgotten diagnosis. Int J Infect Dis 2003;7(2):129–31.
[4] Cleary KR, Batsakis JG. Mycobacterial disease of the head and neck: current perspective. Ann Otol Rhinol Laryngol 1995;104(10 Pt 1):830–3.
[5] Bailey CM, Windle-Taylor PC. Tuberculous laryngitis: a series of 37 patients. The Laryngoscope 1981;91(1):93–100.
[6] Lim JY, Kim KM, Choi EC, Kim YH, Kim HS, Choi HS. Current clinical propensity of laryngeal tuberculosis: review of 60 cases. Eur Arch Oto Rhino Laryngol 2006;263(9):838–42.
[7] Benwill JL, Sarria JC. Laryngeal tuberculosis in the United States of America: a forgotten disease. Scand J Infect Dis 2014;46(4):241–9.
[8] Moon WK, Han MH, Chang KH, Kim HJ, Im JC, Yeon KM, et al. Laryngeal tuberculosis: CT findings. AJR Am J Roentgenol 1996;166(2):445–9.
[9] Shin JE, Nam SY, Yoo SJ, Kim SY. Changing trends in clinical manifestations of laryngeal tuberculosis. Laryngoscope 2000;110(11):1950–3.
[10] Richter B, Fradis M, Köhler G, Ridder GJ. Epiglottic tuberculosis: differential diagnosis and treatment. Case report and review of the literature. Ann Otol Rhinol Laryngol 2001;110(2):197–201.