

Life Threatening Stridor Due to Primary Laryngeal Tuberculosis

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Abstract

Tuberculosis is a potentially serious infectious disease. In otorhinolaryngology practice, lymph node infection is the most common site of predilection followed by the larynx. Laryngeal tuberculosis (LTB) typically presents with dysphagia, odynophagia and hoarseness. We reported a case of primary LTB presenting with acute upper airway obstruction and respiratory distress mimicking acute supraglottitis which requires emergency tracheostomy. Therefore, in acute upper airway obstruction, the appropriate initial investigation should be done to rule out TB to make sure early treatment can be given and it may prevent complications of disease to the patient.

Keywords

Extra-Pulmonary, Granulomatous, Infection, Malignancy, Upper Airway

1. Introduction

Mycobacterium tuberculosis infection commonly involves the respiratory system, however, extra pulmonary involvement has become more common in the present years. In otorhinolaryngology practice, lymph node infection is the most common site of predilection followed by the larynx [1]. Dysphagia, odynophagia and hoarseness are the typical presentation of Laryngeal Tuberculosis (LTB) [2]. Laryngeal involvement commonly affects the interarytenoid region, the arytenoid cartilages, the posterior surface of the true vocal cords and the laryngeal surface of the epiglottis [3]. Primary laryngeal tuberculosis (PLTB) without pulmonary involvement is rare and merely constitutes < 1% of extrapulmonary tuberculosis cases [4]. It is postulated that PLTB occurs as a result of the direct invasion of the larynx by inhaled mycobacteria, rather than ascending infection

from the lower airways [5]. However, we reported patient presented with atypical LTB symptoms, acute respiratory distress which mimicked acute supraglottitis symptoms and denied any TB symptoms and contact. We suggested although in acute respiratory distress presentation, TB investigation should be carried out if the patient does not response to supraglottitis treatment. Therefore, early treatment can be given and may prevent complications of disease to the patient, spreading of disease in the community and improve quality of life.

2. Case Report

A 71-year-old gentleman, with underlying Chronic Obstructive Pulmonary Disease, Diabetes and Hypertension, was brought to the Emergency department with complaints of shortness of breath and noisy breathing for 1 day associated with dysphagia and hoarseness for 4 months prior to presentation. He also had a loss of weight and loss of appetite. He denied fever, tuberculosis contacts or prolonged cough. Examination showed the presence of loud inspiratory stridor with tracheal tug and sternal recession suggestive of respiratory distress. He had a hoarse voice with a main component of strain and an overall dysphonia of grade 3. Auscultation of the lungs showed marked reduced air entry bilaterally with transmitted sounds. Bedside flexible nasopharyngolaryngoscopy revealed diffuse supraglottic edema with bilateral immobile vocal folds at paramedian position as shown in **Figure 1**. Blood investigations revealed a raised white cell count which was neutrophil predominant. He underwent emergency tracheostomy under local anesthesia followed by direct laryngoscopy and biopsy under general anesthesia. Intraoperative findings showed diffuse mucosal edema involving the epiglottis, bilateral true and false cords, subglottis and post cricoid region. Multiple punch biopsies were taken from the edematous mucosa and were sent for histopathological and culture studies. In the interim, he was treated as acute supraglottitis with intravenous ceftriaxone for one week, after which no symptomatic or endoscopic improvement was seen.

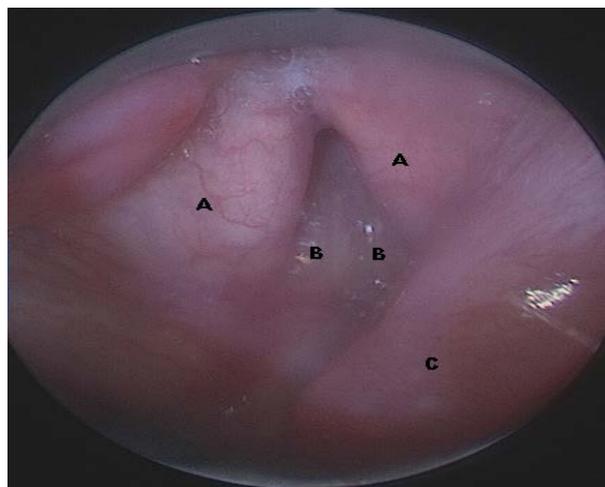


Figure 1. Direct laryngoscope view, on day 1 presentation. Noted diffuse supraglottic edema mainly at bilateral false cord (A); Bilateral true cord edema (B); Arytenoid (C).

Sputum specimens were negative for acid-fast bacilli and the chest radiograph did not show any consolidations suggestive of pulmonary tuberculosis. Mantoux test was positive at 20 mm (>15 mm) and erythrocyte sedimentation rate was raised at 40 mm/H. Histopathological examination of the initial biopsy showed no evidence of malignancy or granulomatosis. A Computer Tomography scan (CT scan) of the neck and thorax revealed soft tissue edema at the supraglottic region causing significant narrowing of the airway, with no signs of an underlying mass as shown in **Figure 2**. A repeated biopsy of the supraglottic tissue was attempted and revealed a positive result for acid-fast bacilli direct smear.

Specimen	Result
Sputum AFB	Negative
ESR	40 mm/H
1 st tissue for AFB	Negative
1 st HPE	No evidence of malignancy or granulomatosis
Repeated (2 nd) tissue for AFB	Positive
Repeated (2 nd) HPE	No evidence of malignancy or granulomatosis

Following a diagnosis of primary tuberculous laryngitis anti-tuberculosis medication was started for him, it was Rifampicin (150 mg) + Isoniazid (75 mg) + Ethambutol (275 mg) + Pyrazinamide (400 mg). After 2 months of intensive therapy, he was Rifampicin (150 mg) + Isoniazid (75 mg) for 4 months for maintenance phase. Following 6 months of anti-tuberculous pharmacotherapy, partial resolution of endoscopic signs was seen (residual supraglottic oedema with persistent bilateral vocal fold immobility) as shown in **Figure 3**, therefore, the patient was commenced on Rifampicin (150 mg) + Isoniazid (75 mg) for another 3 months.



Figure 2. CT scan neck axial cut showed soft tissue edema at the glottis or supraglottic region causing narrowing of the airway (A).

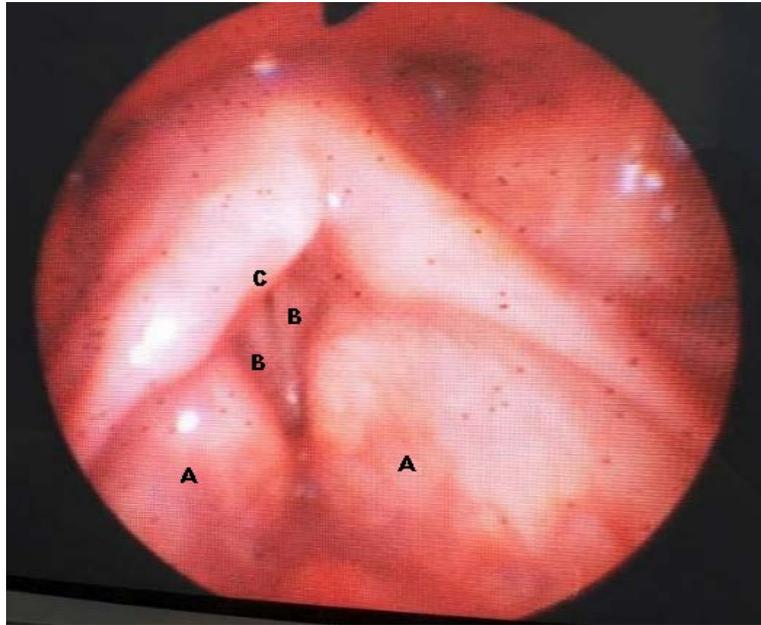


Figure 3. Flexible laryngoscope view on 6 months on anti-TB treatment. False cord not edematous (A); both vocal cords at paramedian position (B) and slit like airway (C).

1 year post completion of pharmacotherapy and remained well since with a pattern airway. He was decannulated. His last follow up was in September 2022 where he remained asymptomatic of upper airway obstruction.

3. Discussion

Tuberculosis (TB) is a chronic disease caused by *Mycobacterium tuberculosis* and has affected mankind for more than 4000 years [6]. In Malaysia, TB is a serious health concern with an annual increase in cases [7]. Data from the World Health Organization (WHO), The Global Tuberculosis Report 2022 showed Pulmonary Tuberculosis (PTB) infection is the most common type of infection by *Mycobacterium tuberculosis* that affects the lung [8]. Extrapulmonary TB (EPTB) is defined as TB infecting organs other than the lung. Approximately 15% of EPTB is from of head and neck. Out of this, 35.6% presented with TB lymphadenitis, followed by 27.4% with TB laryngitis, 13.7% with TB or pharyngitis, 12.3% with TB sialadenitis, 4.1% with TB sinusitis and TB ear, and the least is 2.7% with TB skin [9].

Laryngeal tuberculosis (LTB) commonly occurs secondary to PTB via direct spread [10]. In the absence of PTB, laryngeal TB is believed to be caused by hematogenous or lymphatic spread [11]. Primary Laryngeal tuberculosis (LTB) is a rare type of extrapulmonary tuberculosis, which constitutes < 1% of all types of tuberculosis cases [12]. It is commonly affecting males at a mean age of 4th to 6th decades of life [7].

Frequently, LTB presented with insidious dysphagia, odynophagia and hoarseness [11]. It may also be associated with constitutional symptoms such as night sweats or loss of weight and appetite. In the present case, the patient developed

acute upper airway obstruction which required an emergency tracheostomy. At present, there are only a few reported cases of primary laryngeal tuberculosis causing upper airway obstruction which accounts for 0.5% - 1% of the population in developing countries [13].

Pathognomonic sign of LTB is edema, ulcerative or nodular exophytic lesion appearance [14] and commonly affects the true cord, followed by false cord and epiglottis. It affects more at the posterior part of the larynx. This is based on the theory of mucociliary clearance, where the infected mucus is brought up to the larynx from the trachea, then deposited and infects the posterior part of the laryngeal mucosa [15]. LTB clinical appearance may also resemble a supraglottic carcinoma, therefore it is important to obtain a biopsy from the laryngeal lesion for histopathologic analysis [16] [17]. However, only 34% of HPE results of PLTB demonstrated chronic granulomatous inflammation with caseous necrosis, which is pathognomonic in TB [5].

The diagnostic test for TB is based on a positive mycobacterial smear or culture result. However, in view of the fastidious characteristics of the bacteria, tissue culture may not be successful. Therefore, another supportive test should also be performed example; Mantoux test, sputum for acid-fast bacilli smear (AFB) and culture, histopathological examination of the tissue and radiological examination. Poor response to the initial treatment in this case led us to do another investigation. As a result, repeated blood and tissue samples were sent for testing, along with requests for histopathology analysis and mycobacterium direct smear, the results of which were positive [18].

Based on a study by Kurokawa *et al.* there were various laryngoscopic findings for tuberculous laryngitis. They categorized the endoscopic findings into five different types of lesions: perichondritic, ulcerative, granulomatous, polypoid, and nonspecific inflammatory. Laryngeal tuberculosis may be manifested as a single lesion or multiple lesions [19]. Primary TB of the larynx can cause fibrotic changes in the lamina propria of the vocal fold [3]. Based on Salik *et al.*, progressive scarring from inflammation can cause significant progressive scarring in the glottis. This can lead to vocal cord fixation and paralysis but also can lead to glottic stenosis [20]. But for TB larynx which presents as secondary to pulmonary TB, the recurrent laryngeal nerve could be involved due to tuberculosis in numerous ways. The left is commonly involved due to its long intrathoracic course. Study done by Rajasekanran V *et al.*, the left recurrent laryngeal nerve (RLN) was involved due to compression by mediastinal nodes and scarring. The right recurrent laryngeal nerve could be involved in right upper lobe fibrosis or rarely due to right mediastinal adenopathy [21].

The radiologic features of TB may mimic those of many other diseases and are not specific, thus a high degree of clinical suspicion is required when interpreting the imaging manifestation. The radiological findings of laryngeal tuberculosis depend on the stage and lesion extension. In the infiltrative stage, there may be focal thickening. In the ulcerative stage, the ulceration is normally not deep and rarely reaches the paraglottic spaces and the cartilage. Perichondritis is

sometimes seen (epiglottis, arytenoids), but calcifications are not common and the para-laryngeal fat spaces are usually spared. The last stage is characterized by sclerosis. Various radiological findings that have been described previously include edema alone, an ulcero-infiltrative mass, infiltrative and pseudo-tumoral appearance (66%); sub-glottic laryngitis (isolated swelling of the aryepiglottic fold or even massive cartilaginous ulceration and, sometimes, chondritis or perichondritis); diffuse form; and tuberculoma (enormous ventricular vegetation with a large base that elevates the ventricular strip) [22] [23] [24]. However, all these features should be interpreted carefully with clinical correlation.

The main treatment for LTB is still an anti-tuberculosis regime [7]. The first-line anti-tuberculosis drug consists of ethambutol, isoniazid, rifampicin and pyrazinamide. For patients with Extra pulmonary tuberculosis, a 6- to 9-month regimen (2 months of isoniazid, rifampicin, pyrazinamide and ethambutol followed by 4 - 7 months of isoniazid and rifampicin) is recommended [18].

Currently, in Malaysia, we have combined anti-tuberculosis drugs in a single pill which is called Akurit-4. Despite single tablet regimens that strive for higher compliance, the incidence of multidrug-resistant tuberculosis is rising. Multidrug-resistant tuberculosis (MDR-TB) is defined as resistance to at least isoniazid and rifampicin, and has a relevant epidemiological impact, with 480,000 cases and 190,000 deaths notified in 2014. 10% of them meet the criteria for extensively drug-resistant with additional resistance to any fluoroquinolone, and to at least one injectable second-line drug (capreomycin, kanamycin or amikacin) [25] [26]. Malaysian National TB Information System (TBIS) between 2009 and 2019 reported that the incidence of MDR-TB among patients with TB infections in Malaysia was 0.34% [27]. Therefore, public education on prevention, early diagnosis, and treatment with good drug compliance is essential to avoid these disastrous complications and mortality.

4. Conclusion

Tuberculosis of the larynx may present with acute stridor, mimicking acute supraglottitis. Appropriate initial investigation to rule out TB should be done, as well as proper infectious control measure should be taken. Early treatment may prevent complication of disease to the patient, spreading of disease in the community and improve quality of life.

Consents

The informed consent from the patient was obtained by the patient.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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