CASE REPORT

ENDOTRACHEAL TUBERCULOSIS WITH OBSTRUCTION

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Abstract. Endotracheal involvement of tuberculosis (TB), a type of endobronchial TB, is defined as granulomatous infection of the tracheobronchial tree. We present the case of a 33 year-old female agriculture engineer with endotracheal tuberculosis (ETTB). It was treated successfully with prompt long-course antituberculous medication without complications or need for endotracheal intervention. This unusual case of ETTB, diagnosed promptly by fiberoptic bronchoscopy, and microbiological studies, is presented to emphasize the importance of macroscopic recognition to start anti-TB therapy in cases with significant airway obstruction. This case is important for countries where the various presentations of TB are encountered as well as in countries where TB is not endemic.

Key words: endotracheal tuberculosis, lymphadenitis, obstruction

INTRODUCTION

Endotracheal involvement with tuberculosis (TB), a type of endobronchial TB, is defined as granulomatous infection of the tracheobronchial tree. Ten to 40% of active pulmonary tuberculosis patients have endobronchial involvement. The most probable cause of endobronchial TB is involvement of adjacent lymph nodes with resulting fistulization into the related bronchi. The most common complaints are cough, sputum production, hemoptysis, wheezing, chest pain, fever and dyspnea. Ten to 20% of patients have normal chest X-rays, but this finding does not exclude the diagnosis. The clinical picture can be mistaken for malignancy, thus biopsy of the lesion is required. Granulation tissue with caseating necrosis along with Langhan's type giant cells can be observed and tuberculosis bacilli should be seen on biopsy. The culture should be positive for TB. Diagnosis is frequently delayed with the development of complications, such as tracheal/bronchial erosion or stenosis, atelectasis or bronchiectasis (Davidson, 1985; Smith *et al*, 1987; Kashyap *et al*, 2003; Steinfort *et al*, 2009). Destruction of the lateral aspect of multiple tracheal cartilages due to caseating necrosis has been reported (Park *et al*, 2000). Early diagnosis and treatment are important to prevent complications.

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CASE REPORT

A 33-year old female patient admitted to our department with symptoms of non-

productive, irritating cough, dyspnea and fatigue for 15 days. She was an agriculture engineer with an unremarkable medical, family and work history.

Her physical examination was normal, including chest examination. Laboratory findings, including kidney and liver function tests, erythrocyte sedimentation rate, hemogram and urine tests were all within normal limits. A tuberculin skin test was positive with induration of 16 mm in diameter. One BCG vaccination scar was observed on the left shoulder. A plain posteroanterior chest X-ray showed enlargement of the upper mediastinum and right paratracheal area (Fig 1). High resolution thorax computed tomography (HRCT) revealed milimetric apical subpleural nodules which were irregularly distributed in the parenchymal area and a calcified paratracheal lymph node, 3 x 3 x 2.5 cm in size, which was bulging into the trachea along with narrowing of the superior vena cava (Fig 2).

As the patient had non-productive cough, a sputum sample could not be obtained and fiberoptic bronchoscopy was performed. Two headed gray-yellow tubercular lesions, bulging into the tracheal lumen, causing 30% obstruction of the lower one-third were observed (Fig 3). The bronchial system distal to the carina was otherwise normal.

Forceps biopsy of the lesions and a fine needle aspiration biopsy of the subcarinal lymph nodes showed non-specific inflammation without evidence of TB or malignancy. The specimen was reported as hypocellular in appearance with widespread bronchial epithelial cells, macrophage hystiocytes and rare metaplastic round epithelial cells. Tracheal aspirate and bronchial lavage culture with the BACTEC-PCR method revealed acid-fast bacilli

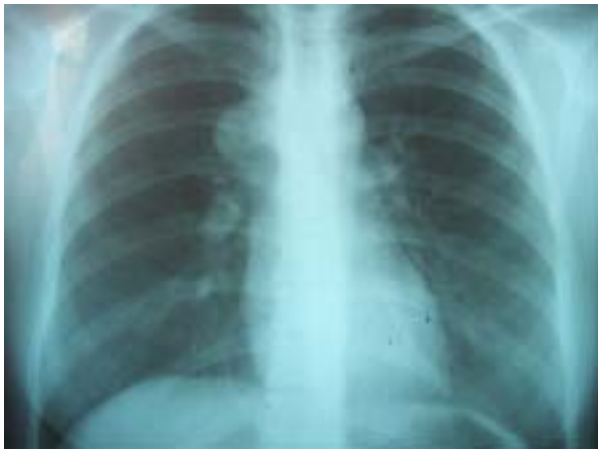

Fig 1–Chest X-ray showing, enlargement of the upper mediastinal and right paratracheal regions.

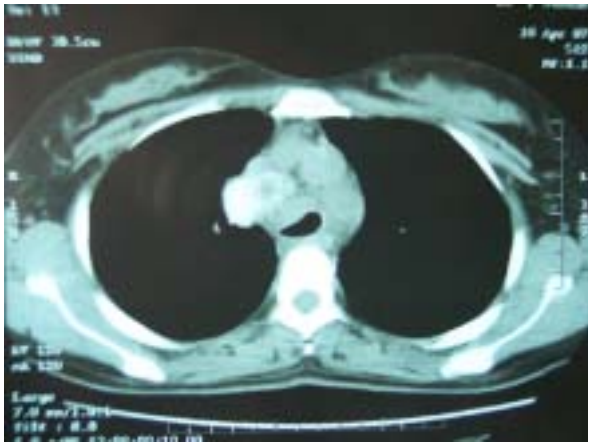

Fig 2–CT showing calcified paratracheal lymph nodes, causing bulging into the trachea along with narrowing of superior vena cava.

(AFB) positivity within a week. This growth was identified as *M. tuberculosis*. Her sputum before and after fiberoptic bronchoscopy remained negative for AFB. After the diagnosis of ETTB, a four-drug initial anti-TB regimen of isoniazid (INH), rifampicin (RF), pyrazinamide (PZ) and ethambutol (EMB) was commenced immediately. The patient could not tolerate the

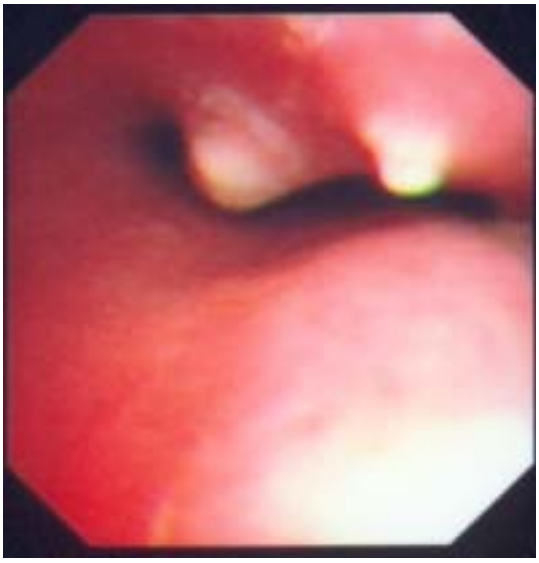

Fig 3–Two headed gray-yellow tubercular lesions protruding into the tracheal lumen.

PZ due to severe nausea and vomiting; it was stopped on the 15th day of treatment. Liver enzymes were in the normal range. The PZ was replaced with streptomycin (SM). After 20 days of SM, severe vestibular symptoms with mild sensory neural hearing loss developed. Thereafter, INH, RF and EMB were given during the initial phase for three months. The total treatment period was expected to be at least 9 months due to PZ exclusion. However, the duration of treatment was extended to 12 months due to the TB lymphadenitis. According to the national TB treatment program, extrapulmonary sites warrant a maintenance phase of 7-10 months. During the 6th month of treatment, significant regression was observed. During the 9th month, almost complete resolution of the parenchymal and tracheal lesions on chest X-ray and CT was observed. Since the patient refused fiberoptic bronchoscopy, tracheal lumen patency was observed by virtual bronchoscopic imaging (Fig 4). Sputum cultures and smears were negative after initiation of therapy. The patient is still undergoing follow-up; a CT scan showed complete resolution of the tracheal lesion and almost complete regression of lymph nodes (Fig 5).

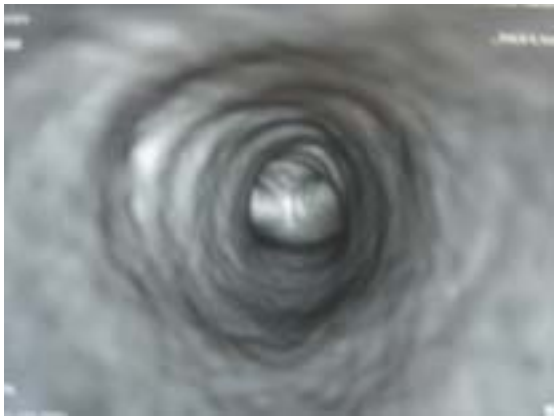

Fig 4–Tracheal lumen patency was observed with virtual bronchoscopic imaging.

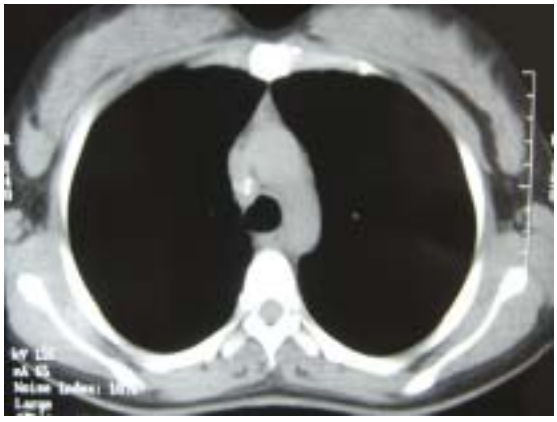

Fig 5–CT scan showing complete resolution of tracheal involvement and significant regression of lymph nodes with residual calcifications.

DISCUSSION

Endobrochial tuberculosis was first described by the English physicien, Rich-

ard Morten, in 1698 (Hudson, 1957). It develops either by mucosal implantation of bacilli originating from cavitary lesions of parenchyma, from infected lymph nodes which erode through the tracheobronchial tree, lymphatic or hematogenous dissemination from parenchymal lesions located in peribronchial regions (Davidson 1985; Smith *et al,* 1987; Kashyap *et al,* 2003; Steinfort *et al,* 2009). In our case, the cause was most likely direct extension of infectious paratracheal lymph nodes into the trachea. Parenchymal lesions may also develop secondary to the peribronchial foci due to bronchial dissemination of the bacilli from eroded tracheal lesions. The clinical presentation of ETTB varies. The most common symptoms are cough, sputum production, hemoptysis, wheezing, chest pain, fever and dyspnea. In patients with active endobronchial disease, bronchorrea can be seen. Chest pain in the sternal or parasternal regions can be experienced due to lymph node involvement or rupture into these areas. Dyspnea is usually secondary to atelectasis of the lungs (Han *et al,* 1992; Lee *et al,* 1992; Chung and Lee 2000; Kashyap *et al,* 2003; Cha *et al,* 2009; Steinfort *et al,* 2009). In our case, irritating cough and dyspnea were the most prominent symptoms, most likely due to invasion into the trachea. Early anti-TB therapy results in fewer structural and functional deficits. Corticosteroid therapy does not influence the outcome in the treatment of endobronchial tuberculosis. Early diagnosis and prompt treatment, prior to the development of fibrosis is important to prevent complications.

An AFB positive smear has been reported in 16-53% of patients with endobronchial involvement, but the percentage is not known for ETTB. A negative smear does not exclude the diagnosis of ETTB; mycobacteria should be sought for using conventional or rapid culture methods. Nucleic acid amplification tests, such as PCR and other methods for amplifying DNA and RNA, may facilitate rapid detection of *M. tuberculosis* respiratory tract specimens. The PCR test is able to detect *M. tuberculosis* in 60% of the smear negative samples but positive on culture with nearly 100% specificity in a short time period (less than a day), compared to the average conventional (2-4 weeks) and radiometric BACTEC techniques (12-14 days), which is supported by earlier studies (Pfyffer *et al,* 1997; Flores *et al,* 2009). Our case supported this recommendation, since she had a negative smear and a negative routine culture, but a positive result with BACTEC and PCR combined.

Chest X-ray findings in ETTB include subcarinal, hilar or perihilar masses, mediastinal enlargement and atelectasis secondary to obstruction. However, 10-20% of ETTB patients have a normal chest X-ray (Lee *et al,* 1992). HRCT revealed additional findings, such as asymmetric nodular branching, lymphadenopathy, tracheal stenosis and an increase in wall thickness. These radiologic findings are not adequate for diagnosis.

Fiberoptic bronchoscopy is the gold standard to evaluate endotracheal obstructive lesions. ETTB can present as active caseous-edematous, hyperemic, fibrostenotic, tumoral, granular or ulcerative lesions (Park *et al,* 2000; Steinfort *et al,* 2009). Granulomatous inflammation can be found with tissue or lymph node needle aspiration biopsy. The macroscopic appearance was also characteristic in our case, with gray-yellow tubercular lesions. For this reason, we can recommend starting anti-TB therapy as soon as fiberoptic bronchoscopy suggests characteristic signs of ETTB. The ETTB lesion in our case caused narrowing of the tracheal diameter

by 30%. The literature report bronchostenosis may develop in 60 to 95% of cases of ETTB, and they may involve the main stem bronchi (Kashyap *et al*, 2003). Bronchial stenosis and strictures are commonly seen in cases of delayed diagnosis but may occur in spite of adequate anti-TB treatment (Chung and Lee, 2000; Park *et al*, 2000).

The development of airway obstruction due to involvement of the trachea is the worst outcome. The place for corticosteroids in bronchostenosis is not clear. Bronchoscopic dilatation is a relatively safe and effective modality for the initial assessment and management of benign tracheobronchial stenosis. In irreversible cases, balloon dilatation or stenting have been recommended (Han *et al*, 1992). Nd:YAG laser therapy and stent placement may be used in conjunction dilatation with bronchoscopic dilatation to treat benign tracheobronchial stenosis in selected cases (Chhajed *et al*, 2001). Since this case was diagnosed early and treated immediately, no complications occurred.

To the best of our knowledge, a case with pure tracheal involvement resulting in distal tracheal obstruction is unique. Marked clinical and radiologic improvement was achieved with the anti-TB drug regimen used, without the need for bronchoscopic intervention. The treatment of ETTB includes combined therapy with INH, RF, EMB and PZ for the first 2-3 months, followed by treatment for 7-9 months with two anti-TB drugs. Due to conditions such as drug resistance and intolerance to therapy to certain drugs, the duration of therapy and the therapeutic regimens could change for tuberculosis management. Therapeutic regimens not including PZ require therapy for a minimum of 9 months. In our case, the duration of therapy extended to 12 months due to intolerance to both PZ and SM, and the fact the TB was extrapulmonary (Anonymous, 2009).

The unusual presentation of ETTB diagnosed by fiberoptic bronchoscopy and microbiological studies emphasizes the importance of macroscopic recognition of ETTB, and the need to start anti-TB therapy in cases with significant airway obstruction. This is especially important in countries where TB is endemic and the various presentations of TB occur as well as in countries where TB is not endemic.

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