RESPIRATORY MEDICINE (1997) 91, 107–110

Airway stenosis after tracheo-bronchial tuberculosis

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Primary endobronchial localization of tuberculosis without change on chest X-ray is a rare clinical entity, and bronchoscopic examination is most appropriate to reveal such an occurrence. A 38-year-old man and a 52-year-old women underwent fibre-optic bronchoscopy many months after the onset of cough with poor sputum and dyspnoea on exercise, chest X-ray being normal. In both cases, a widespread granulomatous involvement of the tracheo-bronchial tree was found and cultures of bronchial wash grew *Mycobacterium tuberculosis*. Patients recovered after 6 months of combined anti-tuberculous and steroid therapy; the granulomatous lesions disappeared but stenoses were found in the trachea and/or main bronchi. In one case, CO_2 laser therapy was performed with no improvement.

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Introduction

Tracheo-bronchial tuberculosis without parenchymal involvement on chest X-ray is a rare clinical entity. Pathogenesis is unclear but it is thought that it is the result of direct implantation of inhaled *Mycobacterium tuberculosis* into the bronchial wall. Due to normal chest X-rays, the diagnosis may be difficult and delayed. Fibre-optic bronchoscopy is the most suitable diagnostic tool for visualizing such lesions. Clinical course is variable. In spite of anti-tuberculous therapy, bronchial stenotic sequelae have been reported and steroid administration has not showed significant advantage (1–9). These complications occur particularly if there is an associated lymphadenitis (3). The present case reports describe two cases of tracheo-bronchial tuberculosis developing in multiple stenoses.

Received 26 July 1995 and accepted in revised form 7 April 1996.

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Case Reports

CASE 1: 38-YEAR-OLD MAN, MANAGER, NON-SMOKER

Fifteen months before admission to the authors' hospital (10 October 1990), the patient began to complain of cough with poor sputum, occasional mild fever and dyspnoea on exercise. Antibiotic administration improved symptoms for a short time. Bronchial asthma was suspected and he underwent skin tests which were positive for *Olea europeae*, *Parietaria officinalis* and *Cupressus*. Subcutaneous desensitization was tried and bronchodilators were administered without improvement. Symptoms were worsened on admission, and inspiratory and expiratory sounds were detected during physical examination. Chest X-ray was normal. Fibre-optic bronchoscopy showed granulomatous inflammation in the lower part of the trachea running to the carina and left main bronchus, with severe lumen reduction and abundant yellow sputum. Acid-fast bacilli were found in the bronchial wash and *M. tuberculosis* grew in cultures.

Rifampicin ($10\ mg\ kg^{-1}$), ethambutol ($20\ mg\ kg^{-1}$) and isoniazid ($5\ mg\ kg^{-1}$) were

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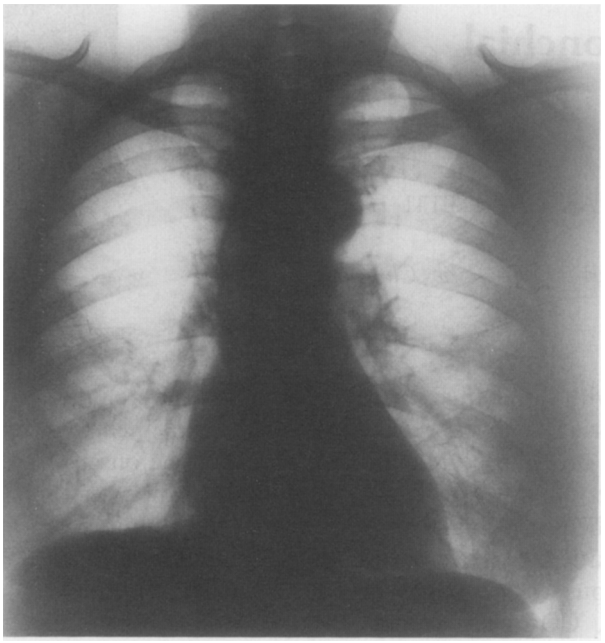

PLATE 1. Normal view of chest X-ray (Case 2).

administered orally for 6 months, and betamethasone 1·5 mg im was administered once a day for 2 months, promptly producing considerable improvement of the endobronchial lesions. After 2 yr, the subject complained of dyspnoea on exercise; fibre-optic bronchoscopy and bronchography showed a severe stenosis of the left main bronchus, located 2 cm below the carina. The patient underwent CO_2 laser therapy with no significant results.

CASE 2: 52-YEAR-OLD WOMAN, TEACHER, NON-SMOKER

In the past year, the subject complained of cough, yellow sputum, pharyngodinia, dyspnoea on exercise, chest pain and intermittent dysphonia without fever. Chest X-ray and skin prick tests gave no positive results. β-adrenergic drugs, steroids by aerosol and oral antibiotics were administered at irregular intervals, improving symptoms sufficiently to keep on teaching in the primary school. In the past 3 months, she had frequent haemoptysis and dysphonia was worsened. In May 1993, sputum smears were positive for acid-fast bacilli and she was admitted to the hospital for evaluation. Physical examination and other laboratory tests were normal. Chest radiogram did not show parenchymal lesions or mediastinal enlargement (Plate 1). Fibre-optic bronchoscopy (6 May 1993) showed widespread inflammation and granulomatous lesions in the trachea, especially in the middle and lower segments where the lumen was reduced and distorted. Lesions went as far as the carina and right main bronchus [Plate 2(a)], thereby interfering with examination. White caseous exudate was present in the examined tracheo-bronchial tree. Rare small nodules were also present in the mucosa of the upper left bronchus and in the lingula. Brushing and bronchial wash were positive for acid-fast bacilli, and

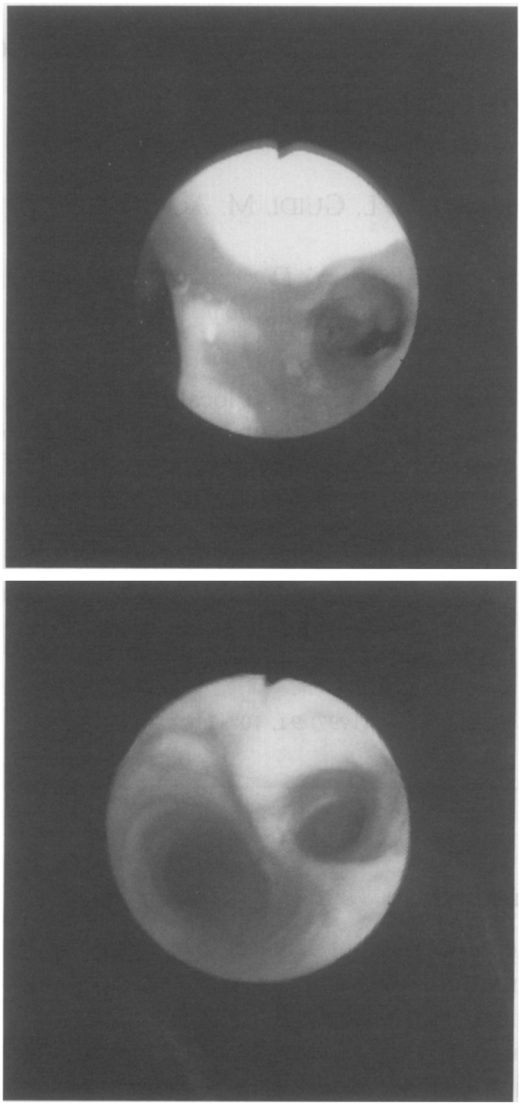

PLATE 2. Endoscopic view of the right main bronchus (a) before and (b) after therapy. The first check showed a widespread inflammation and granulomatous lesions; the second examination showed lesions resulting in stenosis of the airways.

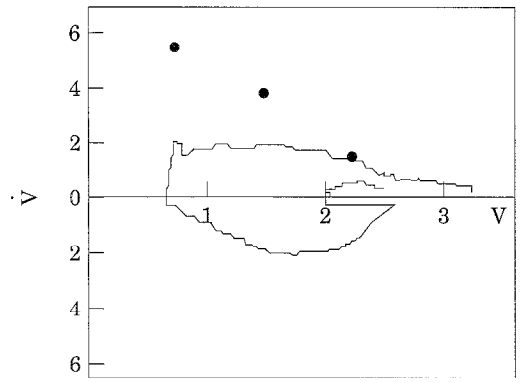

FIG. 1. Flow–volume curve showing an impairment of peak expiratory flow (PEF) and 25–75% maximum expiratory flow ($MEF_{25-75\%}$).

cultures (Lowenstein-Jensen) grew streptomycin-resistant *M. tuberculosis*; biopsy specimens showed caseating granulomas which are typical in tuberculosis. Rifampicin (10 mg kg^{-1}), isoniazid (5 mg kg^{-1}), ethambutol (20 mg kg^{-1}) and betamethasone 0·5 mg were administered for 6 months, improving symptoms. In July 1994, the endoscopic examination showed resolution of granulomatous inflammation, but a severe tracheal stenosis was present 7 cm below the vocal chords (tracheal diameter 7–8 mm). Consequently, the introduction of the instrument caused acute dyspnoea. Right main bronchus ostium was 3–4 mm in diameter [Plate 2(b)] and prevented investigation from being completed. No lesion was found on the left. Bronchial wash was negative for acid-fast bacilli. Lung volumes were normal but the flow–volume curve (Fig. 1) showed a great impairment of peak expiratory flow and maximum expiratory flow at 25–75% FVC (PEF=41%, MEF_{75}=38%, MEF_{50}=34%, MEF_{25}=42%). At the present time, the patient only complains of dyspnoea on exercise.

Discussion

Within the respiratory system, tuberculosis lesions may be located in the parenchyma and/or bronchi. Endobronchial tuberculosis without change in chest X-ray is a rare clinical entity, and the diagnosis may be difficult and delayed (10,11). Lee *et al.* (11) found 10 from 121 tuberculous patients to have chest radiogram without abnormalities. Symptoms are non-specific and are underestimated by patients and general practitioners. For a long time, dry cough may be the first, and sometimes only, symptom followed by dyspnoea, localized wheezing, haemoptysis, hoarseness (due to laryngitis), anorexia, weight loss, chest pain and occasionally fever in the advanced stages (7–10). In addition, sputum smears are often negative for acid-fast bacilli (1,11,12). Ip *et al.* (1) found negative sputum smears in 85% of patients. In these cases, in the absence of radiographic abnormalities, other diseases are diagnosed as bronchial asthma or cancer (13–15). Often after haemoptysis, patients belatedly undergo fibre-optic bronchoscopy, which reveals endobronchial lesions; the correct diagnosis is made by examination of the biological specimens. A common lesion of endobronchial tuberculosis is mucosal inflammation in early stages; submucosal granulomas, ulcerative lesions in the bronchial walls and polypoid mass mimicking bronchial carcinoma are frequent in the advanced disease. Today, bronchial findings after anti-tuberculous chemotherapy can develop in '*restitutio ad integrum*' or cicatricial bronchostenosis (2–4) only revealed with bronchoscopy. Flow–volume curves show abnormalities only when the tracheal diameter is less than 50% (16). Steroid administration could help clinical course and prevent bronchostenosis, but this result is often doubtful (7,8). Three months after 9 months of anti-tuberculous treatment without steroids, Kim *et al.* (3) pursued a fibre-optic bronchoscopy follow-up in 29 patients with endobronchial lesions as a complication of pulmonary tuberculosis. They reported an improvement of bronchial lesions in 53·6%, stationary response in 35·7% and progressive bronchostenosis in 10·7% of patients. Chan and Pang (7) and Williams *et al.* (14) believe that a combined steroid therapy can prevent a hypersensitivity reaction to tuberculoprotein, improving the clinical course, but they are unlikely to be helpful when extensive fibrosis is present. Ip *et al.* (1) and Van den Brande *et al.* (4) thought that steroid therapy had no influence in the outcome of tuberculous endobronchitis.

The present patients complained of dry cough followed by dyspnoea and dysphonia in the absence of parenchymal lesions for a long time. In both patients, the first suspected diagnosis was bronchial asthma, and symptoms improved with non-specific therapy, so they could attend

to daily activities and even work until a few days before admission. Only haemoptysis and persistent dyspnoea indicated the need to employ diagnostic procedures identifying a tuberculous infection. They showed a widespread involvement in the trachea and main bronchi. Due to the severe endoscopic findings, the antituberculous drugs associated with steroids were administered in full doses for many months and with good tolerance in both patients, but tracheo-bronchial lesions resulted in severe upper airway impairment.

In conclusion, tracheo-bronchial tuberculosis without chest X-ray abnormalities is a rare occurrence and the airways involvement may be severe, especially when diagnosis is delayed. Fibre-optic bronchoscopy is the only means able to reveal endobronchial lesions. Antituberculous therapy is effective, but the outcome of the disease is frequently a stenosis of the upper airways, not withstanding steroid therapy.

References

1. Ip MSM, So SY, Lam WK, Mok CK. Endobronchial tuberculosis revisited. *Chest* 1986; **89:** 727–730.
2. Tse CY, Natkunam R. Serious sequelae of delayed diagnosis of the endobronchial tuberculosis. *Tubercle* 1988; **69:** 213–216.
3. Kim YK, Kim HT, Lee KS, Uh ST, Kung YT, Park CS. Serial fibreoptic bronchoscopic observations of the endobronchial tuberculosis before and early after antituberculosis chemotherapy. *Chest* 1993; **103:** 673–677.
4. Van den Brande PM, Van de Mierop F, Verbeken EK, Demedts M. Clinical spectrum of the endobronchial tuberculosis in elderly patients. *Arch Intern Med* 1990; **150:** 2105–2108.
5. Smith LS, Schillaci RF, Sarlin RF. Endobronchial tuberculosis. Serial fibroptic bronchoscopy and natural history. *Chest* 1987; **91:** 644–647.
6. Hoheisel G, Chan BK, Chan CH, Chan KS, Teschler H, Costabel U. Endobronchial tuberculosis: diagnostic features and therapeutic outcome. *Respir Med* 1994; **88:** 593–597.
7. Chan HS, Pang JA. Effect of corticosteroids on deterioration of endobronchial tuberculosis during chemotherapy. *Chest* 1989; **96:** 1195–1196.
8. Alzeer AH, FitzGerald JM. Corticosteroids and tuberculosis: risks and use as adjunct therapy. *Tuberc Lung Dis* 1993; **74:** 6–11.
9. Sanderotz T, Viskum K. Corticosteroids and tuberculosis. *Respir Med* 1994; **88:** 561–565.
10. Pierson DJ, Lakshminarayan S, Petty TL. Endobronchial tuberculosis. *Chest* 1973; **4:** 537–539.
11. Lee JH, Park SS, Lee DH, Shin DH, Yang Sc, Yoo BM. Endobronchial tuberculosis. Clinical and bronchoscopic features in 121 cases. *Chest* 1992; **102:** 990–994.
12. Volckaert A, Roels P, Van der Niepen P, Schandevyl W. Endobronchial tuberculosis: report of two cases. *Eur J Respir Dis* 1987; **70:** 99–101.
13. Van den Brande P, Lambrechts M, Tak G, Demedts M. Endobronchial tuberculosis mimicking lung cancer in elderly patients. *Respir Med* 1991; **85:** 107–109.
14. Williams DJ, York EL, Norbert EJ, Sproule BJ. Endobronchial tuberculosis presenting as asthma. *Chest* 1988; **93:** 836–838.
15. Matthews JI, Matarese SL, Carpenter JL. Endobronchial tuberculosis simulating lung cancer. *Chest* 1984; **86:** 642–644.
16. Maggi G, Ardissone F, Cavallo A, Oliaro A, Scappaticci E, Giobbe E. Tracheal stenosis. A study of 100 cases. *Int Surg* 1990; **75:** 225–230.