Primary Care Respiratory Journal (2008); 17(1): 51-55

Primary Care
RESPIRATORY JOURNAL
www.thepcrj.org

CASE REPORT

Bronchial anthracofibrosis and tuberculosis presenting as a middle lobe syndrome

Jaya Kala, Sandeep Sahay, *Ashok Shah

Department of Respiratory Medicine, Vallabhbhai Patel Chest Institute, University of Delhi, India

Received 18th May 2007; accepted 12th December 2007

Summary

Bronchial anthracofibrosis, a clinical entity described less than a decade ago, is characterised by anthracotic pigmentation of the bronchial mucosa with multifocal bronchial lumen narrowing. The right middle lobe is predominantly involved and is frequently associated with tuberculosis. The condition is generally seen in non-smoking elderly ladies with a longstanding history of wood smoke exposure. A 65 year-old lady presented to us with a one-month history of dry cough. The chest radiograph revealed a middle lobe syndrome which was confirmed on computed tomography (CT) scanning. In addition, narrowing of the right middle lobe bronchus was seen. This raised the suspicion of a malignancy. Fibreoptic bronchoscopy revealed anthracotic pigmentation, and bronchial aspirate showed acid fast bacilli. Culture of the aspirate grew *Mycobacterium tuberculosis*. The patient responded to standard antituberculous treatment.

J Kala *et al. Prim Care Resp J* 2008; **17**(1): 51-55.
doi:10.3132/pcrj.2008.00003

Keywords anthracotic pigmentation, bronchial anthracofibrosis, middle lobe syndrome, pulmonary tuberculosis, wood smoke

Introduction

The term "bronchial anthracofibrosis" is of fairly recent origin and has come to define a clinical entity with bronchial lumen narrowing and anthracotic pigmentation visible on bronchoscopy. First reported from Korea,[1] it was predominantly seen in non-smoking elderly ladies with a longstanding history of exposure to wood smoke used either for cooking or heating. The luminal narrowing and anthracotic pigmentation was most commonly observed in the right middle lobe.[1-3] A very strong association between bronchial anthracofibosis and pulmonary tuberculosis was also recorded.

We report an elderly lady who presented with a right middle lobe syndrome (MLS). Fibreoptic bronchoscopy led to the diagnosis of bronchial anthracofibrosis with pulmonary tuberculosis. We highlight the radiological presentation of this syndrome. To our knowledge, this is the first report from India of bronchial anthracofibrosis associated with tuberculosis presenting as a right MLS.

Case report

A 65 year-old non-diabetic lady, who was human immunodeficiency virus seronegative, was referred to our Institute for evaluation of a "non-resolving pneumonia". She had a one-month history of a predominantly dry cough with diffuse right sided chest pain which worsened on coughing. She had a significant history of exposure to wood smoke – wood fires being the method of cooking – since childhood. Physical examination revealed an elderly lady in no acute distress.

A review of the chest radiograph revealed an ill-defined opacity abutting the right cardiac border with loss of cardiac silhouette. The presence of the silhouette sign suggested a MLS (Figure 1). This was confirmed with a right lateral view which showed a wedge-shaped density extending from the hilum anteriorly and inferiorly along with loss of volume (Figure 2). Computed tomography (CT) scanning of the thorax at presentation showed pretracheal calcified lymph nodes and consolidation of the middle lobe with air

* Corresponding author: Professor Ashok Shah, Department of Respiratory Medicine, Vallabhbhai Patel Chest Institute, University of Delhi P.O. Box 2101, Delhi 110 007, India. Tel: 91 – 11 – 2543 3783 Fax: 91 – 11 – 2766 7420 E-mail: ashokshah99@yahoo.com

Figure 1. Chest radiograph (postero-anterior) done on presentation showing the presence of the silhouette sign suggestive of a middle lobe syndrome.

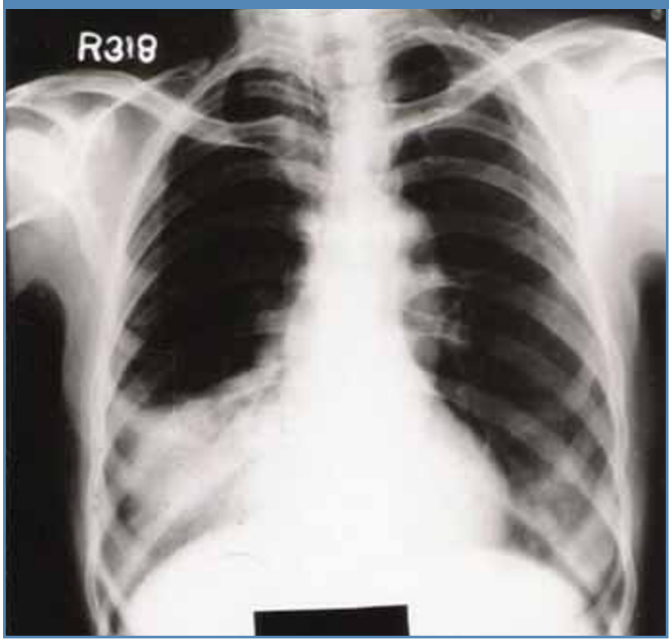


Figure 2. Chest roentgenogram (lateral view) showing a wedge-shaped density extending from the hilum anteriorly and inferiorly confirming the middle lobe syndrome.

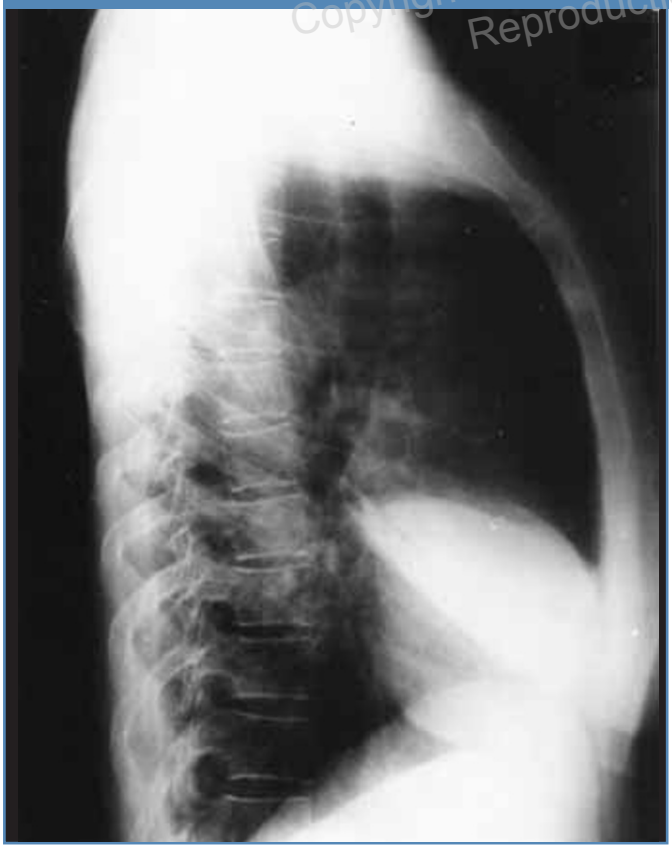


Figure 3. Contrast enhanced computed tomography of the thorax showing the multifocal nature of bronchial stenosis. Consolidation of the right middle lobe with air bronchograms was also seen.

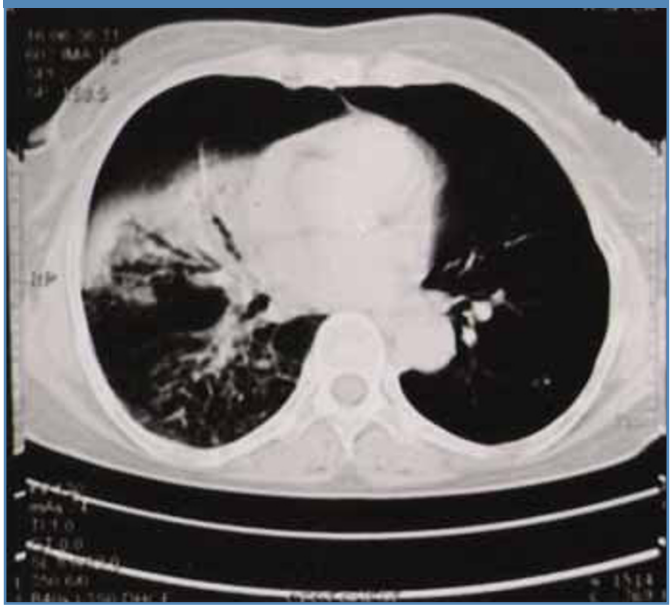
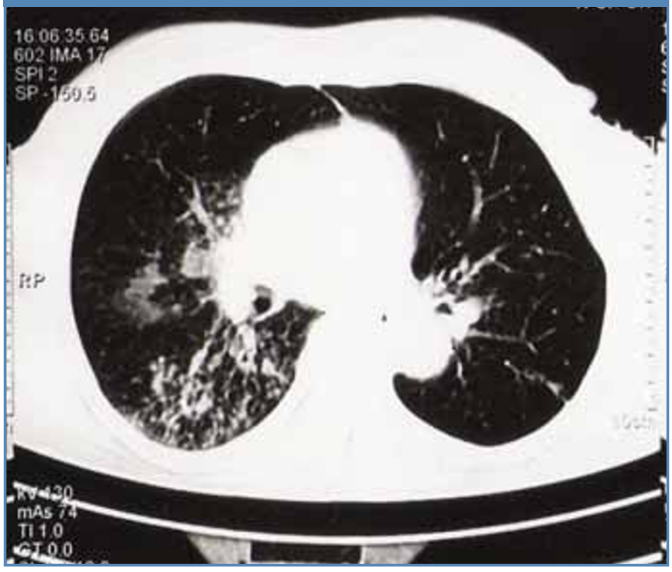


Figure 4. Contrast enhanced computed tomography of the thorax showing bronchial wall thickening due to peribronchial cuffing of soft tissue. Luminal narrowing is also seen. Nodular infiltrates in the right lower lobe were also seen.



bronchograms. Multifocal stenosis of the right middle lobe bronchus was a prominent feature (Figure 3). Peribronchial cuffing of soft tissues indicating bronchial wall thickening, a narrowed lumen and nodular infiltrates in the right lower lobe were also visualised (Figure 4).

Several stains for acid fast bacilli (AFB) were negative as was the Mantoux test. Fibreoptic bronchoscopy visualised a narrowed but patent right middle lobe bronchus with patchy areas of bluish-black mucosal hyperpigmentation. Bronchial aspirate was positive for AFB, and culture grew *Mycobacterium tuberculosis*. Culture of post-bronchoscopy sputum also yielded *M. tuberculosis*. A transbronchial lung biopsy obtained from the right middle lobe revealed a chronic granulomatous inflammation with epitheliod cell granulomas. There was significant bleeding after biopsy. A diagnosis of MLS due to bronchial anthracofibrosis associated with pulmonary tuberculosis was made. Within a few days of initiating

Table 1. Reported patients with bronchial anthracofibrosis: clinical & roentgenological details.

Parameters	Chung *et al*, 1998[1]	Kim *et al*, 2000[2]	Long *et al*, 2005[3]	Our patient
Number of patients	28	54	2	1
Gender (M/F)	8 / 20	16 / 38	0 / 2	0 / 1
Median age (years)	64	67	67	65
Non smoker	20 / 28 (71.42%)	46 / 54 (85%)	2 / 2	yes
Lobes on bronchoscopy Single Isolated middle lobe	 6 / 28 (21.42%) 3 / 28 (10.71%)	 26 / 54 (48%) 15 / 54 (27.7%)	 1 / 2 1 / 2	 0 0
Multiple Middle lobe with other lobes	22 / 28 (78.5%) 21 / 28 (75%)	28 / 54 (52%) 34 / 54 (63%)	1 / 2 1 / 2	Present Present
Right middle lobe with anthracofibrosis with tuberculosis	13 / 21 (61%)	No information available	2 / 2	Present
Active tuberculosis	17 / 28 (75%)	32 / 54 (59.3%)	2 / 2	Present
Chest radiograph	Consolidation with reticulonodular shadows 9 / 28 (32%) Consolidation 5 / 28 (17.8%) Segmental / sub-segmental atelectasis 2 / 28 (7%) Others 12 / 28 (42.8%)	No information available	Consolidation with volume loss 1 / 2 Multiple intrathoracic lymph nodes 1 / 2	Consolidation with volume loss
CT Thorax - Parenchyma	Peribronchial cuffing 24 / 28 (85.7%) Nodules 23 / 28 (82%) Consolidation 13 / 28 (46.4%) Atelectasis 10 / 28 (36%)	Bronchial narrowing or atelectasis 51 / 54 (94%) Bronchial narrowing without atelectasis 4 / 54 (7.4%) Atelectasis with bronchial narrowing 39 / 54 (72.2%)	Bronchial narrowing with right middle lobe consolidation 2 / 2	Peri-bronchial cuffing Consolidation Bronchial narrowing
Lymph nodes	Calcified lymph nodes surrounding bronchi 8 / 28 (28.6%) Non-calcified lymph nodes 4 / 28 (14.3%)	Mediastinal lymph nodes 31 / 54 (57.4%) Mediastinal and hilar lymph nodes 17 / 54 (31.5%) Hilar lymph nodes 3 / 54 (5.6%) Calcification 29 / 54 (57%)	Multiple intrathoracic nodes with calcification 1 / 2	Pre tracheal calcified lymph nodes

antituberculous therapy she experienced symptomatic relief. A chest radiograph done after two months showed considerable resolution, in that the middle lobe parenchymal opacity had cleared to a large extent with reduction in the density. The patient was then lost to follow-up.

Discussion

Bronchial anthracofibrosis was first described by Chung and colleagues from Korea.[1] They identified a group of 28 patients, with a median age of 64 years, who had definite narrowing or obliteration of the lobar or secondary bronchi with anthracotic pigmentation in the surrounding mucosa on fibreoptic bronchoscopy. All 28 patients, 20 of whom were females, had a significant history of wood smoke exposure. Apart from one, all were non-smokers. The right middle lobe was involved in 21 patients, three of whom had an isolated MLS. In addition, 17 had active tuberculosis and three more had a remarkable radiological improvement with antituberculous treatment.[1]

This newly-recognised clinical entity was further confirmed when Kim and colleagues described the CT findings in 54 more Korean patients with bronchial anthracofibrosis who had a similar clinical profile and right middle lobe involvement associated with tuberculosis.[2] Our patient too was an elderly lady who had longstanding exposure to wood smoke and who presented with a MLS. Fibreoptic bronchoscopy confirmed the presence of anthracofibrosis and tuberculosis. Long *et al* also described two Indian female patients who had emigrated to Canada and had had similar presentations[3] – both were diagnosed as anthracofibrosis with active tuberculosis and involvement of the right middle lobe. The clinical details and radiological features of patients in the three reports[1-3] are summarised in Table 1.

Radiological techniques appear to play a vital role in the assessment of bronchial anthracofibrosis and associated tuberculosis. Chest radiographs usually show areas of consolidation with or without reticulonodular shadows, segmental or subsegmental atelectasis, or reticular shadows. CT findings include bronchial narrowing or obstruction, along with thickening of the wall or peribronchial cuffing. Low attenuation endobronchial nodules with calcified or noncalcified lymphadenitis compressing the bronchi are important features.[2] The multifocal nature of bronchial narrowing, often seen on CT, helps to exclude the possibility of a malignancy.[1] The right middle lobe was most frequently involved.[1-3] The CT was repeated after treatment with antituberculous drugs in the two patients described by Long *et al*.[3] The exudative parenchymal lesions had cleared in both patients. In the first patient airway stenosis too had responded with treatment. However, in the second patient, there was only partial response to the adenopathy-related bronchial disease and no improvement in the right middle lobe bronchostenosis.

Learning points for clinicians

- Pulmonary tuberculosis can rarely present as an isolated middle lobe syndrome
- Anthracotic pigmentation, on fibreoptic bronchoscopy, is generally seen in patients with occupational exposure
- Bronchial anthracofibrosis is characterised by anthracotic pigmentation of the bronchial mucosa with multifocal bronchial lumen narrowing
- The right middle lobe is predominantly involved and is frequently associated with tuberculosis
- Non-smoking elderly ladies with a long-standing history of wood smoke exposure are at risk

On fibreoptic bronchoscopy, the striking features are bronchial narrowing and anthracotic pigmentation along with a significant bleeding tendency after bronchial biopsy[1], as was seen in our patient. Stenosis in bronchial anthracofibrosis was thought to be due to an exaggerated immunological response to tuberculous antigen in lymphatics or lung parenchyma.[1-3] It was postulated that exposure to domestic wood smoke, through altered macrophage function, could possibly predispose such patients to tuberculosis.[3]

Pulmonary tuberculosis presenting as a MLS in an elderly subject is not a common clinical presentation in India. The term "middle lobe syndrome" was coined by Graham *et al* when they reported 12 cases of middle lobe atelectasis of non-tuberculous origin subsequent to bronchial compression caused by enlarged lymph nodes.[4] However, it was Brock and colleagues who first drew attention to MLS and hypothesised that the strategic location of the middle lobe bronchus in relation to the intermediate bronchus was responsible for the middle lobe atelectasis.[5] Culiner postulated that anatomical isolation and ineffective collateral ventilation were important factors.[6] In a review of 933 patients with MLS, tuberculosis was recorded in only 9%.[7]

The term "hut lung" was coined by Grobbelaar and Bateman to describe a clinical entity which is a domestically acquired form of pneumoconiosis also caused by wood smoke exposure at home in rural African women.[8] These women, mostly symptomless, also had anthracosis. The radiological picture ranged from diffuse nodulation resembling miliary tuberculosis to extensive fibrosis resembling progressive massive fibrosis. Some patients also had coarse irregular nodules. In contrast to our patient, none

of these patients had evidence of active tuberculosis. The term "hut lung" appears to have evolved to encompass non-infectious, non-malignant respiratory conditions caused by chronic exposure to high levels of biomass smoke in poorly ventilated houses.[9]

In an elderly subject, the combination of bronchial narrowing along with distal atelectasis in association with mediastinal lymphadenopathy suggests the possibility of a malignancy. Given a history of prolonged wood smoke exposure, it is important for the clinician and/or radiologist to recognise that bronchial anthracofibrosis with associated tuberculosis can also present similarly.

Conflict of interest declaration

There were no conflicts of interest for the authors during the preparation of this manuscript.

References

1. Chung MP, Lee KS, Han J *et al*. Bronchial stenosis due to anthracofibrosis. *Chest* 1998;**113**:344-50.
2. Kim HY, Im JG, Goo JM *et al*. Bronchial anthracofibrosis (inflammatory bronchial stenosis with anthracotic pigmentation): CT findings. *Am J Radiol* 2000;**174**:523-7.
3. Long R, Wong E, Barrie J. Bronchial anthracofibrosis and tuberculosis: CT features before and after treatment. *Am J Radiol* 2005;**184**:S33-S36.
4. Graham EA, Burford TH, Mayer JH. Middle lobe syndrome. *Postgrad Med* 1948;**4**:29-34.
5. Brock RC. Post-tuberculous bronchostenosis and bronchiectasis of the middle lobe. *Thorax* 1950;**5**:5-39.
6. Culiner MM. The right middle lobe syndrome, a non-obstructive complex. *Dis Chest* 1966;**50**:57-66.
7. Wagner RB, Johnston MR. Middle lobe syndrome. *Ann Thorac Surg* 1983; **35**:679-86.
8. Grobbelaar JP, Bateman ED. Hut lung: a domestically acquired pneumoconiosis of mixed aetiology in rural women. *Thorax* 1991;**46**:334–40.
9. Diaz JV, Koff J, Gotway MB, *et al*. A case of wood-smoke-related pulmonary disease. *Environ Health Perspect* 2006;**114**:759-62.