Case Report

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TANAFFOS

Non-Occupational Anthracofibrosis Presenting as Massive Hemoptysis: A Case Report

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Anthracofibrosis is considered a benign obstructive lung disease, usually in elderly non-smoking females. The disease is commonly seen in areas where exposure to biomass fuel is common from heating and cooking sources. The common presenting symptoms are cough and breathlessness. It is difficult to differentiate clinically and radiologically from other common lung diseases like COPD, TB, and lung cancer due to non-specific symptoms and radiological findings. The common radiological findings include enlarged calcified mediastinal lymph nodes, mass-like lesions, nodules, and atelectasis. Even on histopathology, it is also difficult to differentiate it from malignant disease like melanoma. Hemoptysis was reported as a symptom of anthracofibrosis in 9-20% of cases, but not as massive or life-threatening hemoptysis. We report a case of bronchial anthracofibrosis presenting as massive hemoptysis. Other possible causes of hemoptysis were ruled out. The patient refused further surgical management. She was treated conservatively and on regular follow-up for the last year without recurrence of hemoptysis and other complications.

Keywords: Anthracofibrosis; Hemoptysis; Biomass fuel; Exposure and Massive

INTRODUCTION

Anthracofibrosis is defined as anthracotic pigmentation and bronchial lumen narrowing. It is commonly observed in patients with coal workers' pneumoconiosis; other known causes include air pollution and exposure to biomass fuel smoke (1,2). It is commonly seen in non-smoking elderly females with a history of exposure to biomass fuel. Cough and breathlessness are common presenting symptoms. Other symptoms are chest pain, sputum production, weight loss, and hemoptysis (1,3). Radiology findings include bronchial narrowing, atelectasis, mass-like lesion, and calcified mediastinal lymph nodes. Therefore, it is difficult to differentiate it from other common lung diseases such as malignancy, COPD, and TB (1-5). Lung function parameters reported were decreased across all parameters, with an obstructive type of lung function abnormality (6). Confirming the diagnosis of anthracofibrosis not only requires bronchoscopic findings but also requires ruling out the differential diagnosis. Clinically, radiologically, and histopathologically, it is difficult to differentiate it from malignant disease such as melanoma (7). The common causes of hemoptysis include bronchiectasis, lung cancer, TB, mycetoma, and pulmonary vascular diseases (8-9). Here, we are presenting a case of anthracofibrosis presenting with massive hemoptysis.

CASE SUMMARIES

A 70-year-old non-smoking female from Leh, Ladakh presented with complaints of dyspnea on exertion and cough with minimal sputum for 6 years. There was also a history of 4 episodes of massive hemoptysis (500-600ml per

episode) and fever for the last one week. There was no history of decreased appetite or weight loss. She was a known case of hypertension and hypothyroidism on regular medications. There was no history of TB in the past. She was a homemaker with a history of exposure to biomass fuel at home for around 40 years. She was referred to our hospital for further evaluation of massive hemoptysis after initial stabilization. Clinical examination revealed a pulse rate of 96/minute, respiratory rate of 22/minute, BP 144/82, and SpO_2 80% on room air. There was no pallor, clubbing, or peripheral lymphadenopathy. Bilateral diffuse rhonchi were present on auscultation of the chest. Baseline investigations were unremarkable with Hb 15.2g/dl, total leucocyte count 5800/µl, and platelet count 272000/µl. Coagulation profile was normal. Chest X-ray showed bilateral prominent bronchovascular markings with hilar prominence. Contrast enhanced computer tomography (CECT) of the chest revealed a few small calcified subcarinal and bilateral hilar lymph nodes, bilateral random nodules, with a few patchy areas of GGOs in the left lower lobe (Figure 1).

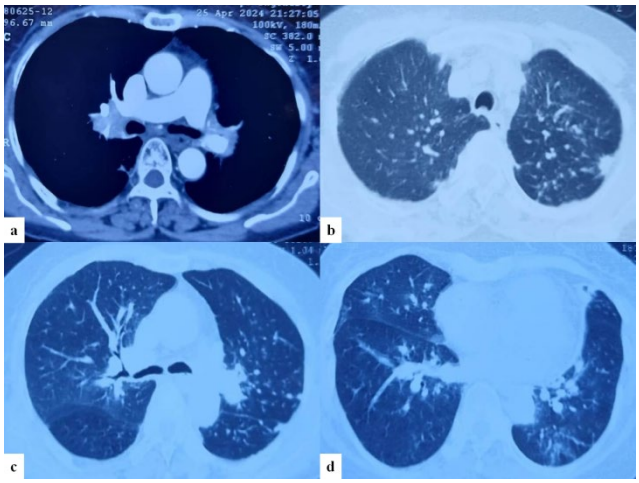


Figure 1. CECT chest, a) mediastinal window showing enlarged subcarinal and hilar lymph nodes with calcification; b, c, d) Parenchymal window showing bilateral nodules in all lobes with left lower lobe ground-glass opacities

Her sputum for acid-fast bacilli (AFB) staining was negative, and *Mycobacterium tuberculosis* (M. tb) was also not detected on GeneXpert. Sputum for pyogenic and fungal culture revealed no growth. Video bronchoscopy showed multiple patches of anthracotic pigmentation present in the bilateral bronchial tree with distortion of the bronchial lumen. The lumen of all lobar bronchi on the left side was narrowed and distorted (anthracofirbrosis); the bronchoscope could not be negotiated further. Moderate bleeding occurred during the procedure, which was controlled by cold saline instillation (Figure 2).

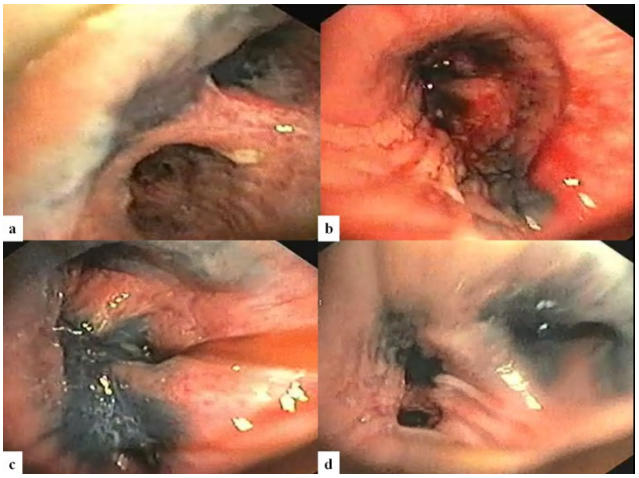


Figure 2. a) Bronchoscopy showing anthracotic pigment deposition at the carina; b) Anthracotic pigments in left main bronchus; c) Multiple anthracotic pigments with distortion and narrowing of all left lobar bronchi; d) Multiple anthracotic pigments with distortion of right lobar bronchus

Bronchial washing for AFB, GeneXpert, pyogenic culture, and fungal culture was negative. Bronchial biopsy from the lesions showed pseudostratified ciliated columnar cells with dense anthracotic pigment deposition in the subepithelium, with no atypical cells or malignant cells (Figure 3). We ruled out all possible causes of hemoptysis and treated her with conservative management. Patient had refused other surgical intervention. She was on regular follow-up for more than a year after massive hemoptysis without recurrence and other complications.

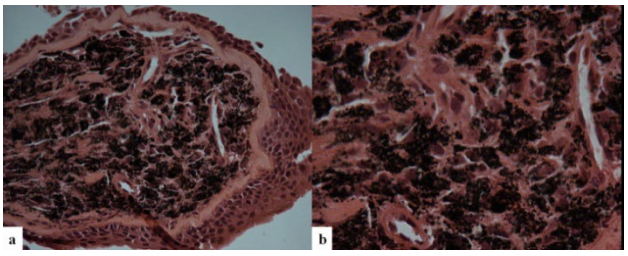


Figure 3. Section showing endobronchial bits lined by pseudostratified ciliated columnar epithelium with dense anthracotic pigment deposit in the epithelium

DISCUSSION

The exact cause of anthracofibrosis remains unknown to date. However, several hypotheses play a role in the development of the disease, including history of exposure to biomass fuel smoke, genetic predisposition, air pollution, and chronic infections like TB (1,10). The disease is mostly reported from developing and underdeveloped countries of Asia, especially from Korea and Iran (1-5,10-11). Kim et al. from South Korea described biomass fuel-induced bronchial anthracofibrosis in 333 patients. They found that most of the patients were elderly, non-smoking females and concluded that the disease clinically appears as a form of obstructive airway disease (3). Sigari and Mohammadi from Iran, in a larger study (n=778) of anthracosis/anthracofibrosis, reported that most of the patients were elderly, with a mean age of 63years. They found that most of the female patients were housewives and most of the male patients were farmers or manual workers (11). In both Iranian and Korean rural homes, biomass fuels such as wood, cow dung, leaves, and crop residues were used for cooking and heating. Family members often lived in a single room, where cooking was also performed. These rooms typically had only a single roof window for ventilation, resulting in inadequate air circulation and potentially prolonged exposure to indoor air pollutants. This may be a reason for the higher incidence of anthracofibrosis in this population. Our case is an elderly, non-smoking lady reported from Ladakh with similar housing conditions and lifestyle.

The common clinical symptoms are breathlessness and cough. The other symptoms of anthracofibrosis are sputum production, fever, hemoptysis, weight loss, and chest pain (1,3,5,11-12). Hemoptysis as a symptom of anthracofibrosis was reported in 2-46% of cases by different studies. However, none of the studies mentioned the grade of hemoptysis (3,5,11-12). This high variability of hemoptysis among different studies might be due to study sample size, disease severity, and type of study. Two larger studies of 300-700 patients reported that hemoptysis as a symptom was seen in 9-20% of cases (3,11). Yun et al., in a study from Korea, found hemoptysis in only four of 167 bronchial anthracofibrosis patients. They also reported that hemoptysis is one symptom used used to differentiate anthracofibrosis from endobronchial lung cancer, with statistical significance (5). Singh et al. from India found that hemoptysis is seen in 46% of the cases in their study. The higher rate of hemoptysis might be due to a small sample size and study inclusion criteria. The study included only 30 cases who underwent bronchoscopy for various indications (12).

The present case is a rare case of anthracofibrosis presenting with massive hemoptysis as the presenting symptom. All the other possible causes were ruled out, and the patient's follow-up also did not reveal recurrence or any other complications even after a year of the massive hemoptysis episode.

The confirmed diagnosis of anthracofibrosis required bronchoscopy. The clinical and radiological findings of the disease are not specific. Due to its non-specific symptoms and radiological findings, the confirmed diagnosis required visualization of anthracotic pigmentation, bronchial narrowing, and ruling out all other possible differential diagnoses. There is a lack on well design studies on the radiology of anthracofibrosis. The common radiological findings are bronchial narrowing, atelectasis, calcified mediastinal lymph nodes, mass-like lesions, collapse, consolidation, nodules, and fibrotic bands (1,3,5,11-14). Mirsadraee et al., in a prospective study with 70 patients and 40 controls, found that lymph node calcification and bronchial calcifications were the most significant radiological findings of anthracosis/ anthracofibrosis. The odds ratio of lymph node calcification was 34 for anthracofibrosis, with 83% sensitivity and 89% specificity. The odds ratio of bronchial calcification was 9.4 for anthracofibrosis, with 77% sensitivity and 72% specificity. They concluded that anthracofibrosis can be accurately diagnosed by identifying lymph node and bronchial calcifications on CT Chest (13). The lung mass, collapse, and mediastinal lymph nodes on CT make it difficult to rule out malignancy in

elderly patients. These findings lead to multiple invasive procedures for confirmatory diagnosis of anthracofibrosis. As in our case, nodular opacities, enlarged mediastinal node with massive hemoptysis are very difficult to differentiate from lung cancer without histopathological confirmation.

There is no definitive consensus on the definition of a massive hemoptysis. But in general, bleeding rates >100ml/hour or total volumes > 500 mL in 24hours are considered massive hemoptysis (8-9,15). Massive hemoptysis is also called life-threatening hemoptysis invariably because of its consequences. So, a life-threatening hemoptysis is defined as any hemoptysis with the following criteria: 1) > 100 mL in 24 hours, 2) leads to abnormal gas exchange or airway obstruction, 3) causes haemodynamic instability in the patient. The cut-off volume of 100 ml/24 hours has been selected because it is the amount of haemoptysis that can threaten the patient's life (15). As per the studies, it is estimated that 5–14% of patients presenting with hemoptysis will have massive/life-threatening hemoptysis. The common causes of massive hemoptysis include infections like TB, fungal/bacterial pneumonia, lung malignancies, bronchiectasis, chronic bronchitis, pulmonary vascular diseases, collagen vascular diseases of the lung, and pulmonary trauma (8,9). To the best of our knowledge, there is no published report of anthracofibrosis causing massive hemoptysis. So, anthracofibrosis can be considered a cause of massive hemoptysis, especially in an endemic area like our case. Anthracofibrosis should be considered in the diagnostic evaluation of patients presenting with massive hemoptysis, particularly in populations with a high burden of the condition, as early recognition may help prevent unnecessary invasive procedures.

CONCLUSION

Anthracofibrosis is a benign obstructive lung disease usually presenting with cough and breathlessness in non-smoking elderly females with a history of biomass fuel exposure. Massive hemoptysis may be the presenting feature of anthracofibrosis. Anthracofibrosis should be considered as a possible cause of massive hemoptysis, especially in high burden population for prevention of unnecessary invasive procedures.

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