THORACIC/ CARDIAC

Radiological spectrum of anthracofibrosis: A series of 40 patients with computed tomography, bronchoscopy, and biopsy

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Abstract

Introduction: Anthracofibrosis is a lesser known clinical entity. Patients present with chronic symptoms of cough and breathlessness with a history of biofuel/wood fire smoke exposure. There are distinct computed tomography (CT) imaging features of anthracofibrosis that can differentiate it from more common conditions such as tuberculosis (TB) and bronchogenic carcinoma. Findings include multifocal noncontiguous stenosis of bronchial tree, calcified enlarged mediastinal or hilar nodes, and secondary lung parenchymal changes. However, in TB, bronchostenosis usually involves a single lobar bronchus in a contiguous manner with trachea and/or major bronchi also being affected. In this study, we highlight the imaging characteristics of anthracofibrosis. Context: The CT findings of anthracofibrosis closely mimic TB and bronchogenic carcinoma, hence we highlight the key imaging features of anthracofibrosis. Aims and Objectives: To identify and describe the CT imaging features of anthracofibrosis and correlate it with bronchoscopic findings. Setting and Design: Retrospective study. Materials and Methods: Retrospectively, 40 patients were selected who were diagnosed with anthracofibrosis on bronchoscopy and biopsy. However, CT scan records of only 14 patients were available for review. Two radiologists reviewed the scans independently. Results: Most common CT finding was multisegmental noncontiguous bronchostenosis seen in 93% patients mostly involving the right middle lobe. 85% of the cases showed lymph node enlargement involving hilar, peribronchial, and mediastinal nodes. The nodes were calcified in 91.7% of the cases, with 58% showing pressure effect on adjacent bronchi due to nodal enlargement. The next common findings were peribronchial cuffing and bronchial obstruction seen in 57 and 28% of the cases, respectively. Conclusion: The key imaging features of anthracofibrosis on CT are multifocal involvement of bronchi with smooth peribronchial thickening and enlarged calcified lymph nodes.

Key words: Anthracofibrosis; anthracosis; bronchostenosis; calcified lymph nodes

Introduction

Anthracofibrosis is a disease characterized by deposition of anthracotic pigments in the bronchial wall with associated

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bronchostenosis.^[1,2] The computed tomography (CT) appearance is the narrowing of bronchi with peribronchial soft tissue and peribronchial lymph node enlargement,

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peripheral segmental atelectasis of the lung, and parenchymal changes secondary to bronchial narrowing. Endobronchial tuberculosis (TB) and bronchogenic carcinoma are commonly encountered causes of bronchial stenosis mimicking anthracofibrosis on imaging,^[2] hence it necessitates bronchoscopy and biopsy to clinch the diagnosis. Awareness of anthracofibrosis being poor, radiologists rarely report it as a primary diagnosis, especially in a country like India where majority of patients are diagnosed as having lung TB. This study highlights the CT imaging characteristics of anthracofibrosis and its correlation with bronchoscopy.

Materials and Methods

This is a retrospective study in which we obtained records of patients who underwent bronchoscopy and biopsy from 2009 to 2015. Forty such reported cases were of anthracofibrosis. These patients had presented to the pulmonology outpatient department of our hospital with chest complaints.

The detailed clinical history of presenting symptoms, their duration, history of smoking, exposure to wood fire smoke (Chula/Tandoor) during domestic cooking or heating (as in bukhari), occupational history including working in stone and coal industries, area of residence, and past history of TB were obtained.

The CT scans of 14 patients were available and were conducted on a 64-slice MDCT Philips Brilliance scanner in the Department of Radiology. High resolution and routine sequence in lung and mediastinal window were studied in detail, and multiplanar and maximum intensity projection reconstructions done at the workstation. Two radiologists recorded the CT findings and a consensus was achieved. The CT scans were performed and analyzed in detail [Table 1] for bronchial narrowing/stenosis, bronchial obstruction, peribronchial soft tissue thickening, atelectasis/collapse, hilar/ mediastinal/peribronchial lymph nodes. Additional findings such as fibro-nodular opacity, consolidation, bronchiectasis, interstitial reticular opacities, fibrosis, air trapping, or mosaic attenuation in lungs were also recorded. Lung parenchymal changes, such as atelectasis and bronchiectasis occurring secondary to bronchostenosis, their distribution, and associated pleural abnormalities were also noted.

Bronchostenosis [Figure 1A] was defined as peribronchial wall thickening with soft tissue attenuation at both sides

	Intraparenchymal soft tissue cuffing			Atelectasis	Mediastinal/ hilar LN	Pressure effect on adjacent bronchi	Consolidation	Bronchoscopy	Cytology Biopsy
1	-	+ MS	-	+ US	+	+ US		MS + AP	No malignancy/No granuloma/AFE Antracotic pigments – EBB, TBNA
2	-	+ MS	-	-	+	+ US	+ US	MS + AP	No malignancy/No granuloma/AFE Antracotic pigments – EBB, TBNA
3	-	+ MS	+ US	+ US	+	+ US	+ US	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments – EBB, BAL
4	-	+ MS	+ US	+ US	+	-	+ US	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments – BAL, TBNA
5	+ US	+ US	+ US	-	+	-	+ US	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments – EBB, TBNA
6	+ US	+ US	+ US	+ US	-	-	-	US + AP	No malignancy/No granuloma/AFB Antracotic pigments – EBB
7	+ US	+ US	-	+ US	+	-	-	US +AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL, EBB
8	+ MS	+ MS	-	-	+	-	-	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL, EBB
9	+ MS	+ MS	-	-	-	-	+	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL, EBB
10	-	-	-	-	+	-	-	AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL
11	-	+ MS	-	+ US	+	+ US	-	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL
12	+ MS	+ MS	-	-	+	+ US	-	MS + AP	No malignancy/No granuloma/AFB Antracotic pigments –EBB
13	+ MS	+ MS	-	+ US	+	-		MS + AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL
14	+ US	+ US	-	+ MS	+	+ US	-	US + AP	No malignancy/No granuloma/AFB Antracotic pigments –BAL

of the bronchus on axial images. Visible noncalcified and calcified mediastinal [Figure 2A] and peribronchial lymph nodes were recorded, and those with a diameter of more than 10 mm in short axis were regarded as lymphadenopathies. Intraparenchymal soft tissue cuffing [Figure 2C] was regarded as the increased bronchial wall thickness from a segmental level onwards. Visible noncalcified and calcified mediastinal [Figure 2A] and peribronchial lymph nodes were recorded, and those with a diameter of more than 10 mm in short axis were regarded as lymphadenopathies.

Bronchoscopy was conducted in all patients [Figures 2D and E, 3]. Bronchial lavage (BAL) and/or endobronchial biopsy (EBB) or trans bronchial nodal aspiration (TBNA) were obtained. Bronchoscopy revealed scattered anthracotic pigmentation in bronchial mucosa. The pigmentation was usually seen beyond proximal main stem bronchi or seen in multiple segments, some of which were with stenosis. The mucosa was swollen in some cases with partial obstruction. There were multisegmental irregular, distorted, and stenotic bronchi [Figure 3] involving both sides of the lungs. Predilection for the right middle lobe (RML) bronchus stenosis was noted often during bronchoscopy. Negotiation through severely stenosed segments was sometimes quite difficult and not achievable.

Bronchoscopic biopsy was performed in few cases and was associated with brisk and significant bleeding when samples were obtained from severely anthracotic segments. Hence, subsequently, a segment of bronchus showing stenosis with lesser severity of pigmentation was chosen for biopsy. Biopsy revealed inflammation with mononuclear infiltrates

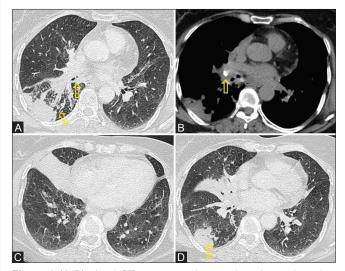


Figure 1 (A-D): Axial CT sections in lung and mediastinal window reveal (A) right lower lobe bronchial narrowing and obstruction with distal subsegmental atelectasis, tree in bud nodules and consolidation seen in right lower lobe. (B) Calcified lymphnode and peribronchial soft tissue causing narrowing of the bronchi at the hila. (C) Right middle lobe atelectasis with bilateral mosaic attenuation in lung fields. (D) Consolidation in right lower lobe

showing pigmentation in macrophages and bronchial epithelium in EBB and lymphocytes in TBNA [Figure 2F]. BAL was obtained for cytology and microbiology. Active TB was excluded by biopsy, acid fast bacilli staining, and culture of BAL fluid in all cases.

Results

Out of the 40 cases diagnosed on bronchoscopy and biopsy as anthracofibrosis, CT was available for 14 cases. The age of these 14 patients ranged from 59 to 89 years (mean = 71 years). 8 patients were males, 6 were females. Presenting complaints were predominantly cough, dyspnea, phlegm, and wheezing without any constitutional symptoms of TB (weight loss, fever, and loss of appetite). None of the patients had active TB at the time of presentation; however, 3 patients had a positive history of TB and gave a history of intake of antitubercular treatment for 6 months.

4 out of 6 female patients elicited the history of chronic exposure to wood fire smoke. 10 out of 14 patients hailed from Afghanistan where wood fire is commonly used for cooking. History of smoking was elicited in 3 out of 14 patients and none of the patients had an occupational exposure to coal, wood dust, asbestos, or silica.

Pulmonary function test (PFT) was also conducted in all cases. Most patients had mild obstructive pattern without significant reversibility. Mixed obstructive and restrictive pattern was seen in a few cases.

The CT findings in our 14 patients were noted. CT features with bronchoscopy and histopathological findings are summarized in Table 1. Bronchial stenosis [Figures 1, 2, 4, and 5] was the most common CT finding

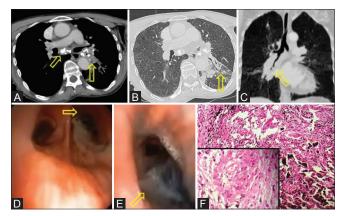


Figure 2 (A-F): (A and B) Axial sections show enlarged calcified lymph nodes causing multifocal bronchial narrowing and peribronchial soft tissue cuffing with lung consolidation (long arrow). (C) MIP images show circumferential peribronchial soft tissue thickening encasing right bronchus intermedius. Few fibrotic opacities with diffuse emphysematous changes are also seen in the lung fields. (D and E) Deposition of black pigments on mucosa with bronchial narrowing. (F) Histopathology slides show chronic non specific inflammation, fibrosis with mononuclear cell infiltrate with anthracotic pigments

seen in 13 patients (93%). It was multisegmental [Figure 2] in 9 cases and unisegmental [Figure 4] in 4 cases, with RML bronchus [Figure 2C] being the most commonly involved. The stenosis is moderate to severe with complete bronchial obstruction [Figure 4C] seen in 4 cases (28%). Peribronchial soft tissue cuffing [Figures 2C, 5] was another finding seen in 8 cases (57%). This is seen along the distal segmental bronchi rather than within the central main stem bronchi or trachea. Lymph node enlargement was the second most common finding seen in 12 patients (85%) involving the hilar [Figure 6B], peribronchial, and mediastinal region. Calcification within the lymph nodes [Figures 1B, 2A and 5] was seen in 11 out of 12 cases (91.7%). Pressure effect of the enlarged lymph nodes [Figures 1B, 2 and 5] on adjacent bronchi causing architectural distortion was seen in 7 out

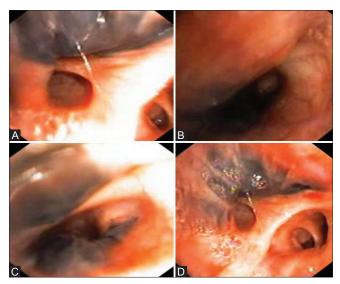


Figure 3 (A-D): Bronchoscopy findings in Anthracofibrosis. (A) Bronchoscopy reveals dense anthracotic deposits occluding left B1 – apicoposterior bronchus. (B and C) Bronchoscopy at level before division of right middle medial and lateral B4, B5 bronchi shows stenosed medial and lateral segments due to anthracotic deposits. (D) Distorted stenosed left B1,B2, and open left B3 (Left Apicoposterior and anterior) segmental bronchi

of 12 cases (58%) of nodal enlargement. It was seen that the causative factor for the bronchial stenosis was hence either peribronchial soft tissue cuffing and/or pressure effect of enlarged lymph nodes on the adjacent bronchi.

Subsegmental atelectasis [Figures 6D, 4B] was seen in 8 out of 14 (57%) patients, commonly involving the RML. Other findings observed on CT were bronchiectasis [Figure 4A and B], interstitial fibrosis [Figure 2C], mosaic lung attenuation [Figures 1C, 6A], fibro-nodular opacities [Figure 6C], consolidation [Figures 1D, 4D], emphysema [Figure 2C], and parenchymal calcification indicating sequelae to previously healed infection. Pleural disease, including thickening and effusion, was rare.

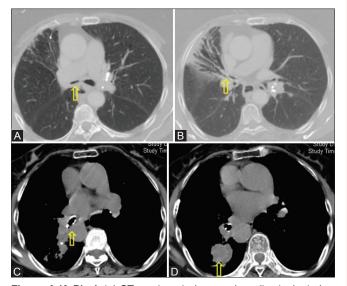


Figure 4 (A-D): Axial CT sections in lung and mediastinal window showing bronchial narrowing and secondary lung changes in different patients: (A) Right mainstem bronchus narrowing, (B) bronchiectasis in right middle lobe with distal atelectasis and fibrosis, (C) bronchial narrowing, obstruction with bronchial calcification and (D) soft tissue mass like consolidation in right lower lobe

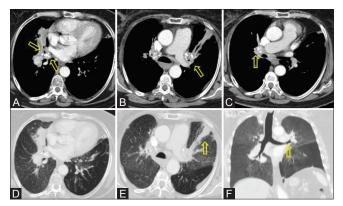


Figure 5 (A-F): CT Axial sections in mediastinal and lung window show: Multifocal bronchial narrowing in right middle lobe (A and D), left upper lobe (B) with enlarged calcified lymph nodes (C) and subsegmental atelectasis (E). MIP coronal reconstruction in lung window shows peribronchial soft tissue thickening (F)

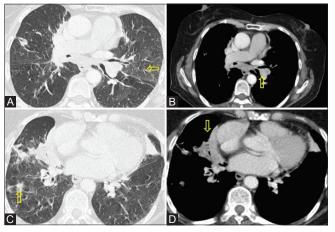


Figure 6 (A-D): Contrast CT scan through the chest in lung and mediastinal window show various spectra of findings in patients with anthracofibrosis – (A) mosaic attenuation, (B) hilar lymphadenopathy, (C) fibronodular opacities and (D) parenchymal atelectasis

Bronchoscopy revealed scattered anthracotic pigmentation in bronchial mucosa [Figures 2D and E, 3]. The pigmentation is seen beyond proximal main stem bronchi in multiple segments with associated stenosis. There were multisegmental and unisegmental irregular, distorted, and stenotic bronchi [Figure 3] involving both sides of lungs. Predilection for RML bronchus stenosis was noted.

Biopsy [Figure 2F] revealed inflammation with mononuclear infiltrates showing pigmentation in macrophages and bronchial epithelium in EBB and lymphocytes in TBNA. 3 patients gave a history of pulmonary TB. None of the cases had a positive culture or acid fast bacilli.

Discussion

Simple anthracosis (anthrac: coal, osis: condition) is a black discoloration of mucosal surface of the bronchi and often incidentally detected during bronchoscopy and is nonobstructing on the airway, hence does not qualify to be a pathological entity. However, when associated with luminal narrowing and fibrosis, it is titled as anthracofibrosis and the patient presents symptomatically.

Anthracofibrosis is an airway disease occurring due to airway inflammation with bronchial narrowing and is diagnosed on bronchoscopy. It results from repeated exposure to air pollution, wood smoke, and dust.^[1,2] The pathogenesis of anthracofibrosis is reasoned to be due to exposure and deposition of carbon causing impairment of ciliary action, followed by an exaggerated immunological and inflammatory reaction, which results in peribronchial inflammation, fibrosis, and bronchostenosis.^[2] In our study, patients of anthracofibrosis have no history of exposure to mining or coal dust/silica.^[3,4] Also, none of the patients gave a history of occupational exposure to mining or a related industry. However, 3 patients gave a history of smoking in whom features of chronic obstructive pulmonary disease such as emphysema was seen. These cases are less and there was no significant association of smoking with bronchostenosis.

In our study, anthracofibrosis occurred predominantly in elderly patients with a mean age of 71 years and no gender predilection. The significant radiological findings observed in our case series were multilobar bronchostenosis in majority of cases (93% cases) with preferential involvement of RML (7 of the 14 cases – 50%) and left upper lobe (in 46% cases). Similar predilection of the segments in RML has been reported in literature.^[3,5] Stenosis (partial or complete) of RML orifice was also a common observation on bronchoscopy.

Peribronchial soft tissue thickening causing bronchostenosis was seen in 57% cases and has also been reported as a common finding in literature.^[6-8]

Enlarged and calcified lymph nodes were the second most important finding and seen in all except two cases (85.7%). Calcification in nodes and irregular peribronchial soft tissue in perihilar region causing architectural distortion and bronchostenosis are commonly encountered in cases of anthracofibrosis.^[5,6] These lymph nodes cause extrinsic compression without eroding into the bronchi^[4,9] and are associated with peribronchial and hilar soft tissue.^[10] Similar high incidence of lymph node enlargement and bronchial pressure has been reported by other authors.^[4]

All these CT features are neither specific nor exclusively found in anthracofibrosis and close differentials are endobronchial TB and bronchogenic carcinoma. The peribronchial lymph node enlargement has many differentials such as TB, carcinoma, lymphoma, and sarcoidosis. In TB bronchostenosis is either secondary to an exaggerated immunologic response to tuberculous antigens in the lymphatics, or from bacilli originating in upstream cavities, or to extrinsic compression from proximal intrathoracic lymph nodes. Bronchoscopy with microbiological and cytological examination of the bronchial lavage or TBNA material gives a definitive diagnosis and can be considered on a case-to-case basis by the treating pulmonologist. It should be noted that in cases of anthracofibrosis the nodes are usually fluorodeoxyglucose-avid on positron emission tomography and further confuse the diagnosis and thus, often reported to be suspicious of malignancy. In such cases TBNA of nodes show macrophages laden with black anthracotic pigments and areas of necrosis with absence of malignant cells or acid fast bacilli or granulomas, which help to exclude malignancy or TB in these hot nodes. On rare occasions, the draining nodes can show granulomatous reaction in response to the pigment laden macrophages migrating to the lymph nodes but unlike tuberculosis, areas of caseation are not seen.

Park *et al.*^[11] reported that bronchostenosis in anthracofibrosis tends to be multifocal, noncontiguous, and involves segmental or lobar bronchi in both lungs. The main stem bronchus and trachea are preserved. They stated that in TB, bronchostenosis involves single lobar bronchus in a contiguous manner and involvement of trachea and major bronchi is more common. Tuberculosis may also show additional radiological findings of cavitation, tree in bud appearance, or centrilobular nodules and military spread. TB nodes can show low attenuation center and peripheral rim enhancement.^[10,11] Kunal *et al.*, in a short series in India, have also highlighted the importance of recognizing this entity and that tuberculosis is an association and not a causative factor.^[12]

To add to the quandary, some of the previous studies^[5,6] have shown a debatable, however, close association of TB with anthracofibrosis.^[2,4] They also speculated that preexisting bronchial injury by TB and course of tubercular

Table 2: CT differentiating features of Anthracofibrosis from Tuberculosis, Malignancy and Sarcoidosis									
CT feature	Anthracofibrosis	Tuberculosis	Malignancy	Sarcoidosis					
Bronchostenosis location	Multisegmental	Unisegmental/ occasionally multisegmental	Unisegmental	Rare					
Bronchostenosis pattern	Smooth	Irregular	Irregular	smooth					
Contiguity of involvement	Noncontiguous	Contiguous	No Contiguity	No Contiguity					
Peribronchial soft tissue	Common	Less common	Peribronchial mass	Peribronchial smooth nodules					
Trachea and Major bronchi	Less common	Common	Any	Less common					
Lobar & segmental bronchi	Common	Less common	Any	Common					
Nodes	Bulky, calcified	Necrotic, confluent and calcified.	Noncalcified, necrotic	Calcfied					
Pressure effect of nodes	Common	Not common	Not common	Not common					
Lung Parenchymal changes	Atelectasis, mosaic attenuation, nodules, consolidation secondary to bronchostenosis	Cavitation, consolidation, infiltration and tree in bud nodules	Parenchymal mass with collapse consolidation	Parenchymal nodules, interstitial changes					

drug treatment could be responsible for development of anthracotic pigment in the fibrosed tissue.

TB was excluded in all patients in the present case series by cytology and culture of the bronchoscopic aspirate. 3 patients had a past history of TB; however, no active TB lesion in the form of caseating epitheloid cell granulomas or Langhans-type giant cell was seen on the biopsy of these patients. Pleural involvement was also uncommon in our series. Hence findings in our study suggest anthracotic deposits even in the absence of tuberculosis can produce peribronchial soft tissue thickening and enlarged calcified lymph nodes, leading to bronchial narrowing.

Malignancy is another important quintessential differential in cases presenting with bronchial narrowing, distal atelectasis, and mediastinal lymphadenopathy. Multisegmental atelectasis, smooth bronchial narrowing, and presence of bulky nodal disease with calcification favor anthracofibrosis. Bronchial carcinoma results in focal and unilobar stenosis which is irregular. Sarcoidosis, another differential, presents with lymph node calcification and lung parenchymal changes.

Table 2 illustrates and summarizes the CT findings in anthracofibrosis, endobronchial tuberculosis, lung carcinoma, and sarcoidosis.

Bronchoscopy is the gold standard in diagnosing anthracofibrosis, and presence of anthracotic pigmentation in association with multisegmental bronchostenosis on bronchoscopy alone may suffice for definite diagnosis. The role of bronchoscopy has been discussed in many previous studies.^[5,6] The differential of pigmented patches on bronchoscopy is malignant melanoma. The increasing use of endobronchial ultrasound (EBUS) TBNA has resulted in higher detection rate of anthracosis in mediastinal/hilar nodes. In such cases, the aspirate is found to be blackish, or the anthracotic pigmented macrophages are seen on cytology. Bronchoscopic biopsy should be undertaken in selective cases when a strong suspicion of malignancy/tuberculosis is present as biopsy of these anthracotic areas that often leads to profuse bleeding and inadequate diagnostic yield on cytology. The choice of mucosal biopsy site should preferably be from a nonanthracotic/nonfibrosed segment of bronchus, which is not totally stenosed.^[12,13] Here the findings of CT are helpful and TBNA may be a safer option with a less bleeding risk.

Our study had a number of limitations. Firstly, it was a retrospective study with a small sample size. Secondly, radiologists were not blinded to bronchoscopy and histopathological results, hence there was a bias.

Conclusion

In conclusion, anthracofibrosis is a distinct entity with widespread but nonspecific CT abnormalities, which may mimic malignancy and endobronchial TB, all of which have an entirely different management and treatment protocol. The awareness of this clinical condition is low among clinicians and radiologists, prevalence of tuberculosis being high hence anthracofibrosis is rarely suggested on imaging, and to our knowledge, not many studies or a large case series have been conducted in India. Our study suggests multifocal involvement of bronchi with smooth peribronchial thickening, enlarged calcific lymph nodes causing pressure effect, and architectural distortion with bronchoscopic correlation (which is gold standard) will enable us to clinch the diagnosis and help us differentiate it from other common conditions avoiding unnecessary invasive diagnostic and therapeutic procedures.

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Conflicts of interest There are no conflicts of interest.

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