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RESEARCH ARTICLE

"LADAKH LUNG" A NON-OCCUPATIONAL PNEUMOCONIOSIS; SPECTRUM OF HIGH RESOLUTION COMPUTED TOMOGRAPHY FINDINGS

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ABSTRACT

Background: Ladakh is one of the world's highest inhabited regions and has a unique environmental features due to presence of both free silica and biomass fuel which is responsible for lung disease. Cases of non-occupational pneumoconiosis has been reported from Ladakh. **Objectives:** To establish the spectrum of High Resolution CT findings in Symptomatic patients with Non-Occupational Pneumoconiosis from Ladakh region of J&K. **Methods:** This is an observational study that was carried out over a period of 18 months in the Department of Radio diagnosis, Department of Pulmonary Medicine and Department of Medicine of Govt. Medical College Srinagar after approval from Institutional Ethical Committee. **Results**: The most common finding among study patients was nodules in 88% of patients, mediastinal lymphadenopathy in 72% followed by bronchial thickening in 60% , linear fibrotic bands in 58%, Calcified mediastinal lymphadenopathy in 54%, ground glass opacity in 48% , multifocal bronchial narrowing in 40%, emphysema in 40%, bronchiectasis in 36%, lung cavitation in 32%, segmental/lobar collapse in 32%, reticulation in 30%, mosaic attenuation 26%, consolidation in 20%, progressive massive fibrosis in 20%, pleural thickening in 20%, honeycombing in 12%,pleural calcification in 4% of patients. **Conclusion**: To conclude clinicians and radiologists have less awareness of non-occupational pneumoconiosis and thus the diagnosis of non-occupational pneumoconiosis is rarely suggested on imaging and instead common diagnosis like tuberculosis are suggested, So our study may help in clinching diagnosis. **Advances in Knowledge:** Only few case reports and case series on patients from Ladakh region with non occupational pneumoconiosis has been reported. This is the first study describing HRCT features. Since tuberculosis is endemic in India, occupational pneumoconiosis was being confused and treated as pulmonary tuberculosis. Our study suggests that certain radiological features can differentiate between tuberculosis from non occupational pneumoconiosis.

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INTRODUCTION

Ladakh is one of the world's highest inhabited regions. [(1)] People usually prefer indoors in semi-closed dwellings and are constantly exposed to smoke. During summers, dust storms occur in most parts of Ladakh and the affected villages are covered by a thick blanket of fine dust for several days or even months. Chemical analysis of this dust has shown that the major constituents of this dust are oxides of silica. [(2)] Ladakh has therefore unique environmental features due to presence of both free silica and biomass fuel. Exposure to dust from dust storms and exposure to soot are responsible for lung disease. Cases of non-occupational pneumoconiosis has been reported from Ladakh. [(1,3,4,5)]Silicosis is caused by the inhalation of fine particles of crystalline silicon dioxide. [(6)] HRCT may show poorly defined centrilobular nodular opacities, bilateral ground-glass opacities and patchy areas of consolidation [(7,8)] or septal thickening (crazy paving) [(9)] . Calcification within areas of consolidation is a common finding. [(8)] Hilar lymph node enlargement may be present. Classic silicosis has two forms: simple and complicated. [(10)] Simple form of classic silicosis is characterized by the presence of multiple small nodules, 2-5 mm in diameter, accompanied by calcifications. [(11)] The distribution may be diffuse, though upper lobe with posterior zone predominance is characteristic. HRCT shows perilymphatic distribution with nodules being observed in centrilobular, paraseptal and subpleural regions. Eggshell pattern of calcification of lymph nodes is common. [(12,13)]Complicated form of classic silicosis, also known as progressive massive fibrosis, develops through confluence of individual silicotic nodules. Coal workers' pneumoconiosis or Anthracosis is seen in coal workers and is caused by inhalation and deposition of black pigments of which carbon is a major constituent. [(15,16)] Air pollution, biomass smoke, and cigarette

smoke are also known to cause deposition of black pigment leading to environmental anthracosis. Environmental anthracosis usually leads to anthracofibrosis (anthracosis with bronchial narrowing) and obstructive airway disease. Both anthracosis and silicosis have been reported due to non-occupational cause.[1,17] As with silicosis, CWP is considered to occur in simple and complicated forms. The CT features of simple CWP include diffuse, small, 1-5 mm sized nodules, most numerous in the upper lung zone. They typically show a perilymphatic distribution but sometimes centrilobular predominance may be observed. These nodules have less distinct margins with granular appearance than those of silicosis and are smaller. [18,19] Lymph node calcification occurs less frequently. In patients with complicated CWP, conglomerate masses develop. Although their radiographic appearance is similar to masses seen in silicosis, their histology is different.

The chest radiograph is the first diagnostic tool for evaluation of pneumoconiosis. However the findings are nonspecific and sensitivity is low, missing as many as 10 to 15 percent of cases with histopathologically documented disease. High-resolution CT (HRCT) is superior to chest radiography in the detection of parenchymal abnormalities being more accurate in providing differential diagnosis and is free from considerable inter-observer variation in its interpretation. Thin-section CT has replaced chest radiography in evaluation of non-occupational pneumoconiosis because of its higher sensitivity for early lung disease and greater accuracy in characterising the pattern of disease. [20,21] It is indicated as investigation for thorough evaluation of positive cases screened by chest radiograph. Furthermore, HRCT detects pulmonary involvement in pneumoconiosis earlier than conventional radiographs and hence early intervention may be offered. To overcome the concern of radiation exposure, a low dose technique and acquisition of limited number of slices can be introduced while using CT for imaging.

MATERIALS AND METHODS

Study Design: This was an observational study that was carried out over a period of 18 months in the Department of Radio diagnosis, Department of Pulmonary Medicine and Department of Medicine of Govt. Medical College Srinagar from November 2018 after approval from Institutional Ethical Committee.

Inclusion Criteria: All adult symptomatic patients hailing from Ladakh who were referred from Department Of Pulmonary medicine as well as Department of Medicine for undergoing HRCT lung were included in the study after obtaining proper consent. A written and informed consent was taken from all patients.

Exclusion Criteria: Patients with contraindications to CT Scan (e.g pregnancy) and age <18 years were excluded.

Methodology

A detailed clinical history fallowed by proper thorough clinical examination was done. Chest Radiograph (PA View Full Inspiration) and High Resolution CT was performed as recommended by the chest specialist. HRCT was performed under following protocol; Scans were obtained using 256 slice dual source DECT (Somatom Defination FLASH, Siemens). Volumetric acquisition was done and patients were examined in supine position in full inspiration. Examination parameters were 80kV and 140kV, 130mAs, 0.5mm rotation time, detector collimation of 1mm. Scans were reconstructed with high special frequency bone algorithm and 512x512 matrix. All the HRCT images were reviewed on the synchronized work station by a senior experienced consultant Radiologist.

Statistical Analysis: The recorded clinical, laboratory and Radiological data was entered and tabulated in MS Excel software. All the stored data was then mopped up to find the spectrum of HRCT findings of these patients. Continuous variables were presented as mean and standard deviation. Categorical variables were presented as frequency and percentage.

RESULTS AND OBSERVATIONS

Table 1. Chest radiograph findings

Findings	Number	Percentage
Nodular opacities	28	56
Reticulations	6	12
Pleural Effusion	1	2

The most common finding was nodular opacity in 56% of study patients followed by consolidation in 20% of study patients, reticulations in 12 % and pleural effusion in 2%.

CT Findings	Number	Percentage
Nodules	44	88
Mediastinal lymphadenopathy	36	72
Bronchial thickening	30	60
Linear Fibrotic bands	29	58
Calcified Mediastinal lymphadenopathy	27	54
Ground glass opacity	24	48
Multifocal Bronchial narrowing	20	40
Emphysema	20	40
Bronchiectasis	18	36
Lung Cavitation	16	32
Collapse	16	32
Reticulation	15	30
Mosaic attenuation	13	26
Consolidation	10	20
Progressive Massive Fibrosis	10	20
Pleural Thickening	10	20
Honeycombing	6	12
Pleural calcification	2	4

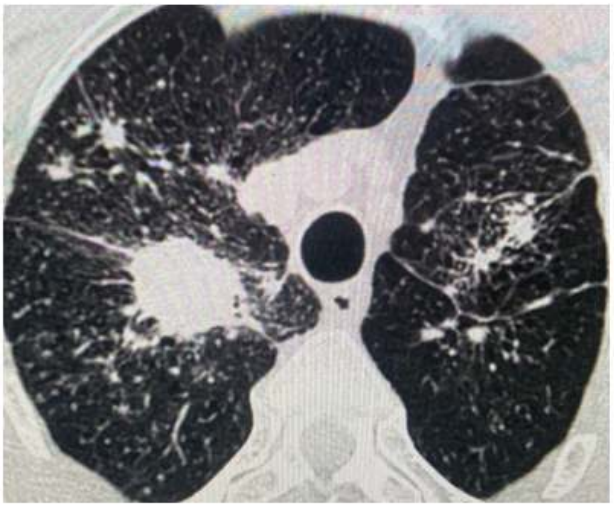
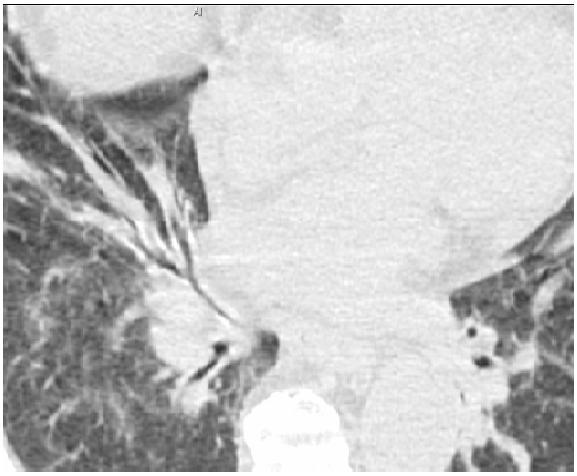


Figure 2A, 2B. Axial images of HRCT Chest showing progressive massive fibrosis with background emphysematous changes



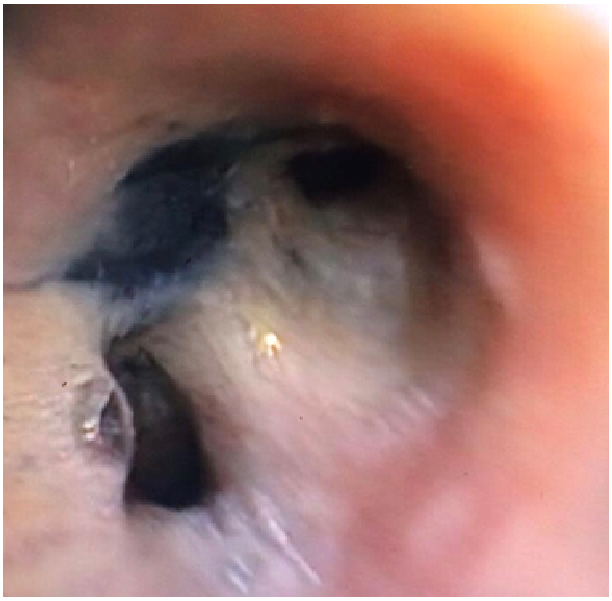


Figure 2A and 3B. Zoom in view of axial cut of HRCT showing bronchial wall thickening and narrowing of RML bronchus (white arrow). Bronchoscopy reveals blackish mucosa of bronchi

DISCUSSION

Pneumoconiosis are a broad group of lung diseases that result from inhalation of dust particles which may occur with or without fibrosis. It may be Occupational or Non-Occupational. Literature is scarce on non-occupational pneumoconiosis. Sepke [(22)] reported silicosis from street dust in 1961. Afterwards, Farina and Gambini [(23)] reported a rare case of silicosis from inhalation of desert sand. The first major study of environmental exposure to silica dust in India was reported by Saiyed *et al* [(1)] in 1991.An epidemiological survey was carried out by them to investigate the occurrence of non-occupational pneumoconiosis in central Ladakh, where there are no mines and industries. The causative agents were suggested to be dust storms in spring and indoor air pollution. Norboo *et al* [(21)] also reported silicosis in Himalayan villages of Chuchot, Shamma and St ok in 1991 . We studied 50 symptomatic patients from Ladakh region who were referred to us for High Resolution CT over a period of 18 months. Our study comprised of 30 men and 20 women with a mean age of 59.96 years (47.69-72.23 years). None of the patients had occupational exposure to dust. The characteristic CT findings were Nodular Opacities, Mediastinal Lymphadenopathy, Bronchial Thickening, Linear Fibrotic Bands, Ground Glass Opacity, Bronchial Narrowing, Emphysema, Bronchiectasis and Progressive Massive Fibrosis.

Nodules: Nodules were the most common radiological finding present in 88% of patients. Out of these, 48% had perilymphatic distribution, 28% had random distribution and 24% had centrilobular distribution. Upper lobe involvement was most common 54. 5%. This finding is consistent with a study done by Antao *et al*. [(16)] wherein nodules were seen using HRCT in 68.3% of 41 workers with silica exposure, including 20 of 22 with abnormal radiographs consistent with silicosis and 4 of 15 who had normal chest radiographs. In this study, subpleural nodules were seen in 21 of the 23 (91.3%) workers with evidence of pneumoconiosis on HRCT. Studies by Lopes AJ *et al* [(24)] , Ozmen *et al* [(25)] , Akira *et al*. [(26)] and Bhawna *et al* [(27)] were in concordance with our study. All of the above-mentioned studies found nodules as the most common HRCT finding as was seen in our study. This could be attributed to the fact that Ladakh has high environmental levels of silica [(2)] and thus had imaging findings consistent with chronic silicosis.

Progressive Massive Fibrosis: Progressive massive fibrosis was seen in 20% of study patients. Progressive massive fibrosis was predominantly bilateral as it was seen in 9(90%) out of 10 study patients with progressive massive fibrosis. Lopes AJ *et al* [(24)] studied 44 patients having silicosis and observed this finding in 33 patients (75%). Upper lobar involvement was predominantly seen as it was present in 7 out of 10 study patients. Our study was in concordance with study by Kahkouee *et al*. [(29)] and Pilaniya V *et al*. [(30)] Adenocarcinoma was confirmed in two of these patients. In our study malignancy was not seen in any patient possibly due to small number of patients and different selection criteria.

Bronchial Thickening: Bronchial thickening was seen in 30(60%) of patients. Chung *et al*. [(20)] had reported peribronchial cuffing on CT chest in 24 of 28 patients with BAF while a study from Iran by Kahkouee S *et al*. [(29)] documented it in 37 of 58 (63.1%) patients Linear Fibrotic Bands:

Linear fibrotic bands were seen in 58% of our study patients. These appear as linear opacities about 1–3 mm in thickness. Kahkouee *et al*. [29] observed this finding in 53% of patients and may represent pleuroparenchymal fibrosis. 11 of the 24 (45.8%) patients with BAF studied by Pilaniya V *et al*. [30] had parenchymal bands on HRCT.

Multifocal Bronchial Narrowing: Multifocal bronchial narrowing involves segmental or lobar bronchi with multiple stenotic segments. In our study multifocal bronchial narrowing was noted in 40%. A study in 58 patients with BAF conducted by Kahkouee *et al*. [29] reported multifocal bronchial stenosis in 23 (39.7%) with right middle lobebronchus being most commonly affected. Pigmentary deposits in BAF are usually encountered at the branching points within the tracheobronchial tree and are followed by inflammatory response and bronchial stenosis [29,32].

Collapse: In our study, 38% patients out of 50 patients had collapse. This included sub segmental collapse, segmental collapse and lobar collapse. 10 patients had collapse secondary to narrowing of bronchi and 1 patient had collapse secondary to extrinsic compression by enlarged calcified hilar node. In a study by Anandamoyee Dhar *et al*. [28] in patients with BAF sub segmental atelectasis was seen in 8 out of 14 (57%) patients, commonly involving the RML. In our study RML collapse was the most common lobe involved noted 9 (47.4%) out of 19 patients, same observations were made by Anandamoyee Dhar *et al*. [28] It is imperative for the pulmonologists/radiologists to exclude malignancy and/or pulmonary tuberculosis in those patients who present with a visible collapse on imaging. Consolidation, bronchiectasis and Emphysema were the other findings noted our study patients. Emphysema**:** Emphysema both paraseptal and centrilobular was present in 40% of study patients. Silicosis is known to cause emphysema. **Bhawna *et al.*** [27] studied CT scans of 150 industrial workers with pneumoconiosis and found emphysema in 27% of workers. Tuberculosis was observed in 28% of our study patients in both active and chronic form.

Mediastinal Lymphadenopathy: Mediastinal Lymphadenopathy was seen in 36(72 %) of patients. Lymph nodes were calcified in 27(54%) of patients. Antao *et al* [16] studied CT scans of 41 stone carvers and found enlarged lymph nodes in 23 stone carvers (56%),17 of whom had evidence of silicosis. 27 workers (65%) showed lymph node calcification. Anandamoyee Dhar *et al*. [28] retrospectively studied 14 CT scans of patients with BAF and found enlarged and calcified lymph nodes in 85.7% patients. These observations are almost similar to the results in our studies. Of 36 patients with mediastinal LAP 54% patients had calcified LAP. 74% patients with calcified LAP had amorphous calcification, 22.2% patients had peripheral calcification, 11.1% had dense calcification and 3% patient had fine stippled calcification.

Pleural Thickening: In our study pleural thickening was noted in 10 (20%) patients. Pleural thickening was noted in bilateral upper lobes anterolaterally. 4% of our study patients had pleural calcification. Hiroaki Arakawa *et al* [35] studied pleural disease on images from 110 patients with autopsy proven silicosis with silica exposure who had been followed radiologically for a mean of nearly 15 years. At CT, pleural thickening was visible in 64 (58%) patients.

This is contrary to our study where 10% patients had pleural thickening. This might be explained by the fact that majority of the patients in above mentioned study had severe and long-term exposure to silica resulting in complicated silicosis where as in our study majority of patients had limited environmental exposure to silica with majority of patients having features of simple silicosis.

CONCLUSION

Non-Occupational Pneumoconiosis is a rare form of pneumoconiosis where people have environmental exposure to dust and smoke typically seen in high altitude regions and in deserts. It can be diagnosed on the basis of spectrum of HRCT findings as described in this study. The HRCT findings of non-occupational pneumoconiosis is non-specific and resembles that of occupational pneumoconiosis.Sometimes these findings mimic pulmonary tuberculosis which has different management. Clinicians and radiologists have less awareness of non-occupational pneumoconiosis and thus the diagnosis of non-occupational pneumoconiosis is rarely suggested on imaging and instead common diagnosis like tuberculosis are suggested. This study will be helpful in suggesting the diagnosis of non-occupational pneumoconiosis in suitable clinical and environmental setting and thus reducing unnecessary invasive diagnostic procedures and treatment modalities.

Limitations of Study: This was a hospital-based study and only symptomatic patients were included in the study. Further studies are needed to establish imaging spectrum in asymptomatic patients and in patients with mild symptoms

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