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Case Report

Bronchiolitis Obliterans in a Coffee Processing Unit Worker from Wayanad: Report of a Rare Case

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Abstract

Background: Bronchiolitis obliterans is a rare condition characterized by fibrosis of terminal and respiratory bronchioles. It usually leads to progressive decline in lung function and has variable outcomes. There are different causes for bronchiolitis obliterans. It can be caused by exposure to inhaled toxins such as diacetyl. Diacetyl is released as a natural byproduct during coffee roasting and grinding. Diacetyl production took place in a completely closed system at high temperature. Exposure to this chemical leads to airway epithelial injury. Methods: Clinical details of a 51 year old patient who worked earlier in coffee processing unit and has features of bronchiolitis obliterans is presented and discussed. Conclusion: A rare case of bronchiolitis obliterans in a coffee processing unit worker is presented here. The cause of bronchiolitis is postulated to be due to exposure to diacetyl from the processing unit.

Keywords: *Diacetyl; Bronchiolitis Obliterans; Popcorn Lung*

Introduction

Bronchiolitis obliterans is a type of obstructive lung disease affecting the small airways [1]. It is a rare condition characterized by fibrosis of terminal and respiratory bronchioles; and spirometry showing predominant small airway dysfunction. It usually leads to progressive decline in lung function and has variable outcomes. Etiology includes lung transplant and hematopoietic stem cell transplantation, exposure to inhaled toxins and gases including mustard gas, nitrogen oxides, diacetyl and fiberglass. Bronchiolitis obliterans is also associated with autoimmune disorders. Diacetyl is a natural byproduct during coffee processing and it is established that this chemical induces air way injury consistent with animal inhalation studies [2]. Diagnosis is usually delayed due to variable presentation and is mostly treated as asthma or COPD. Treatment is mainly supportive and measures are to be taken to avoid exposure from work place. This is a case report of bronchiolitis obliterans in a coffee processing unit worker in Wayanad district of Kerala.

Case Report

A 51-year-old male patient, doing manual work, presented with history of progressive breathlessness for the last 10 years. He also had cough with scanty expectoration. No history of haemoptysis or chest pain. He had few hospital admissions during the past 10 years mostly with febrile illness. He was being treated as chronic obstructive pulmonary disease. There was no history of childhood asthma or

family history of asthma. He was a smoker for 25 years and stopped 5 years back. During this admission, X–ray chest was taken which showed hyperinflation and diffuse reticular shadows both sides with coalescence of shadows on both lower zones (Figure 1A). These lesions appeared different from the increased broncho-vascular markings that are usually seen in chronic bronchitis. Spirometry showed obstructive lung function with predominant small airway dysfunction (21% predicted).

High resolution computed tomography (HRCT) thorax revealed diffuse reticular shadows, few nodular shadows, thin walled cysts and air trapping (Figure 1B). This type of HRCT appearance is seen in Popcorn lung and Marijuana smoking. He denied use of Marijuana at any point of time. He was working in a coffee processing unit for 20 years and quit the job once he developed respiratory symptoms. Coffee processing unit was a medium sized facility where coffee roasting and grinding were done. He used to work in both sections. Confirmation of diagnosis with a lung biopsy was suggested, but the patient did not favor any invasive procedure. He was put on bronchodilators, oral corticosteroid and immunosuppressant (azathioprine) and is being advised regular follow up.

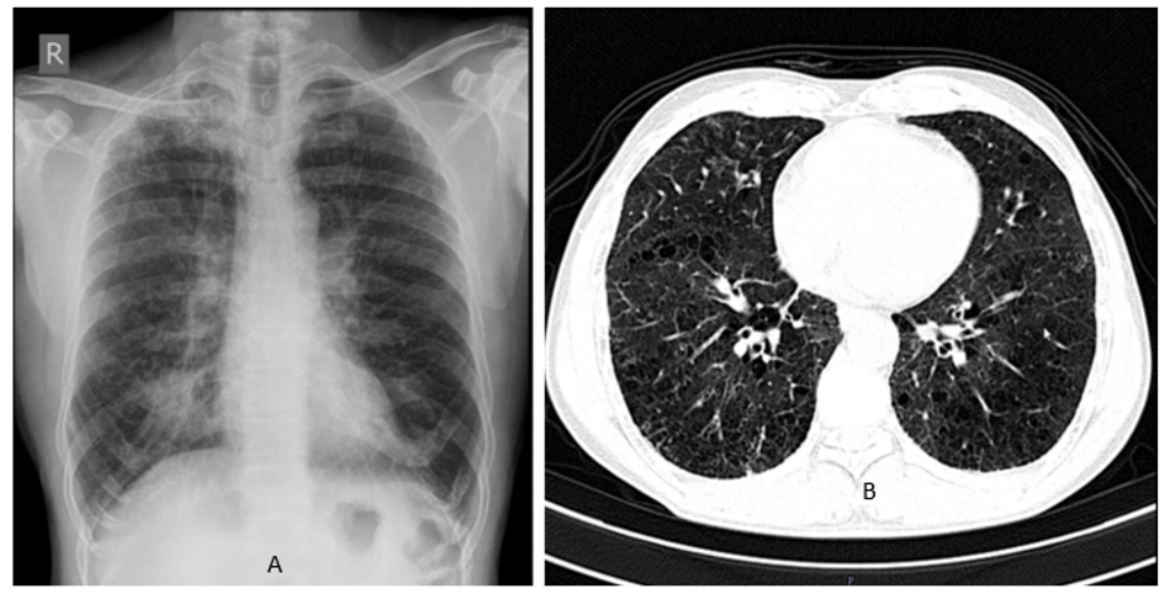

Figure 1: *X-Ray Chest PA view showing hyperinflation and bilateral diffuse reticular shadows. 1B: HRCT Thorax axial section from the lower lobes shows extensive reticulation, bronchiolar wall thickening, air trapping and cyst formation.*

Discussion

Bronchiolitis generically refers to inflammation and/or fibrosis involving (a) airways smaller than 2 mm in diameter [1], which often lack a cartilaginous wall, and/or (b) the alveolar ducts. Bronchiolitis often exhibits nonspecific clinical manifestations that range from an insidious onset of cough and shortness of breath to an acute fulminant illness. Bronchiolitis is divided in to either inflammatory or constrictive. Inflammatory bronchiolitis is due to infection with viral or atypical organism, exposure to organic antigens or noxious particles and aspiration. Constrictive bronchiolitis is seen in transplant recipients.

There will be inflammation of subepithelial structures and dysregulated repair, leading to fibroproliferation and abnormal regeneration of epithelium of the small airways in response to injury from inhalational toxins or autoimmune responses [3]. Histopathology demonstrates the involvement of terminal and respiratory bronchioles (small airways). There will not be any significant changes in alveolar

spaces and distal lung parenchyma. Hypertrophy of the smooth muscles of the bronchioles, peribronchiolar inflammatory infiltrates, accumulation of mucus in the bronchiolar lumen, and bronchiolar scarring are seen in bronchiolitis obliterans [3]. There is a concentric narrowing of the lumen of the bronchioles by the inflammatory fibrosis leading to obstruction and air trapping.

The clinical and radiological presentation resembled that of "Popcorn Lung" reported in workers of a microwave popcorn plant in Missouri in 2002 [4]. This was caused by a flavoring agent termed diacetyl (2,3-butanedione) which is used to give the popcorn a buttery taste [5]. Later, it was reported that this flavoring agent is extensively used in e-cigarettes, favoring the development of this condition among those who use e-cigarettes. Further, it is proved that this chemical is a natural byproduct in coffee-roasting and coffee-grinding processes [6,7]. Hence, unacceptable levels of diacetyl in these units may cause popcorn lung. In their report, the Centers for Disease Control and Prevention (CDC) confirmed that occupational exposure to diacetyl and a related compound, 2,3-pentanedione, can cause bronchiolitis obliterans and loss of lung function. The CDC also reported that these potentially harmful chemicals were found at higher-than-expected levels at some coffee-processing facilities [8]. Popcorn lung (bronchiolitis obliterans) often is associated with symptoms of cough and shortness of breath, similar to that seen in patients with COPD and asthma. This pathology is irreversible and progressive, and there is no definite treatment. Diagnosis is often delayed due to nonspecific clinical features and is initially treated as COPD or asthma. Spirometry demonstrates airflow obstruction that does not reverse with inhaled bronchodilator challenge. Forced expiratory volume in one second (FEV1) will be reduced and the ratio of FEV1 to Forced vital capacity (FEV1/FVC) is also reduced. FEF25-75% will be considerably reduced [9].

Lung tissue biopsy is necessary to confirm the diagnosis of bronchiolitis obliterans. Radiological feature may help in differentiating from COPD and asthma, as these patients will show subtle features of bronchiolar wall thickening and fibrosis. Chest radiographic can be normal or non-specific in early stages. Later it may show features like hyperinflation, attenuation of vascular markings or reticular/reticulonodular markings. These fine reticulations represent bronchiolar wall inflammation. On HRCT chest, there are often sharply defined, areas of decreased lung attenuation associated with vessels of reduced caliber. If there are dynamic images with inspiratory and expiratory films, a mosaic pattern of perfusion may be noted due to air trapping from small airway disease. These changes represent a combination of air trapping and oligemia (mosaic attenuation pattern) [10]. Other features include centrilobular micronodules (often seen as tree-in-bud opacities), bronchiolectasis, bronchial wall thickening and ground glass opacities [11]. In later stage of disease dense network of reticulation with thin walled cysts due lung destruction are seen. These finding are seen also in lung disease secondary to Marijuana exposure.

Treatment of bronchiolitis obliterans syndrome after lung transplant involves augmenting immunosuppression since it is thought to be a form of chronic rejection. Agents like tacrolimus, cyclosporine, mycophenolate mofetil, and prednisone has been used to treat bronchiolitis obliterans syndrome after transplant. Azithromycin has also been shown to decrease the incidence of bronchiolitis obliterans syndrome and improvement in lung function. In non-transplant related bronchiolitis obliterans, removal from offending agents is essential. Immunosuppression with corticosteroids and cytotoxic agents like cyclophosphamide have been used for bronchiolitis obliterans related to rheumatoid arthritis, but have not been beneficial for bronchiolitis obliterans from toxic inhalation or post infectious etiology. They are treated symptomatically.

Wayanad district in Kerala is predominantly a tea and coffee growing farmland at a moderate high altitude. Hence there is lots of small and large scale coffee processing units. It is highly possible that workers in these processing units (Roasting or grinding) may be exposed to diacetyl leading to development of bronchiolitis obliterans. These patients due to their nonspecific symptoms and poor awareness of this entity are treated as COPD [12].

Conclusion

A rare case of bronchiolitis obliterans similar to "popcorn lung" is reported here. The clinical significance is that workers exposed to diacetyl, which is a natural byproduct in coffee processing, develop this disease, and a clinical suspicion among coffee plant workers

presenting with symptoms of obstructive airway disease will help in early diagnosis. Wayanad district of Kerala state is a moderately high-altitude farmland with coffee plantations and coffee-processing units. An epidemiological research to detect the level of these chemicals in coffee-processing units and to assess its health hazard among workers may help establish a causative relationship.

Conflicts of Interest

No.

Funding

No.

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