Case Report

Bagassosis: A case report on rare hypersensitivity pneumonitis

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ABSTRACT

Bagassosis is a rare form of occupational hypersensitivity pneumonitis (HP) caused by inhalation of bagasse. The incidence is significantly low after 1970 due to public awareness and an increase in organized sector industries. Being one of the largest sugar manufacturing country and still the presence of industries in the unorganized sector, it is one of unnoticed diseases in some part of our country. In the present case, we describe the case of a 60-year-old male who presented with signs of respiratory failure. There was a history of occupational exposure to residuals of the sugarcane industry (bagasse) dust for the past 25 years with recent exposure before admission. The chest X-ray depicted bilateral opacity and computed tomography showed bilateral patchy opacities, septal thickening, and ground-glass opacities. His bronchoalveolar lavage revealed lymphocytosis. He was diagnosed as a case of bagassosis (occupation-related HP). He was admitted and treated with systemic corticosteroid and other supportive treatment and recovered gradually from respiratory failure. The treatment continued with a tapering dose of corticosteroid for the next 6 months, which improved him clinically, radiologically, and physiologically.

Key words: Antigen, Bagassosis, Hypersensitivity pneumonitis

Hypersensitivity pneumonitis (HP) is a broader term encompassing a spectrum of disorders caused by the inhalation of antigens of organic dust or low-molecular-weight chemicals [1]. Bagassosis is an entity caused by an allergic reaction to bagasse dust which is a major remaining product of the sugarcane industry. This product is used in the manufacture of pulp, paper industries, as biofuel or in lacquerware crafts [1,2]. The size of the bagasse dust particle ranges from 0.08 to 9 μm. This is significant because the dust particles of size 0.4 μm can reach the respiratory bronchioles and cause either sensitization, alveolitis, or subsequent fibrosis depending on the duration of exposure, with the influence of individual's genetic and environmental factors [1,3,4].

Here, we are reporting a rare case of bagassosis in a 60-year-old farmer, who used to carry sugarcane to the sugar industry with loading and unloading of bagasse during the entire harvesting season, for a span of 25 years and presented to us with signs of respiratory failure. After the treatment with corticosteroid and supportive therapy, the patient recovered significantly.

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CASE REPORT

A 60-year-old male, with no history of addiction and comorbidities, presented to the emergency department with complaints of progressive breathlessness for 10 days which was associated with cough and mild expectoration, fever for 3 days with generalized weakness, and loss of weight for the past 1 month. There was no history of atopy. The family history was also insignificant.

On examination, his blood pressure was 138/80 mmHg, respiratory rate was 32/min, pulse rate was 140/min, and oxygen saturation level was 90% at 10 L/min with O_2 inhalation. He was admitted with signs of respiratory failure. His chest examination revealed bilateral crepitation.

On blood investigation, hemoglobin was 12.6 g/dl, total leukocytes count was 6400/mm^3 with a platelet count of 3.68 lakhs. The kidney function tests, liver function tests, and fasting blood glucose were within normal limits. The chest radiograph (Fig. 1a) depicted bilateral non-homogenous opacities predominantly in the lower zone. The echocardiography was normal. Arterial blood gas analysis at room air showed pH – 7.40, PaO_2 – 52 mmHg, $PaCO_2$ – 34 mmHg, and HCO_3 – 27 nmol/L.

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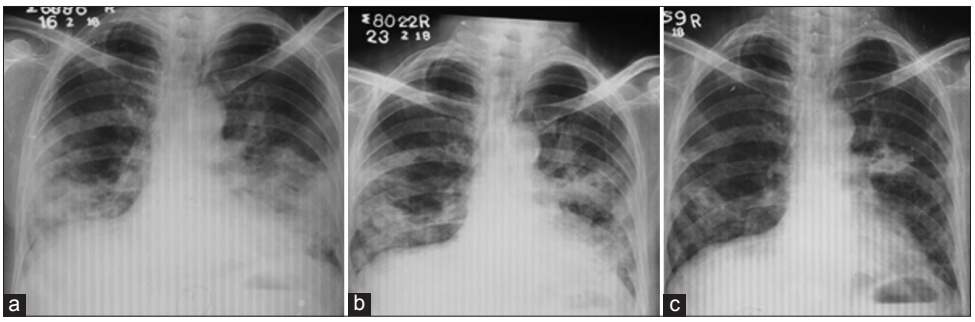

Figure 1: Chest X-ray (PA view) shown bilateral lower and mid zones opacities at the time of presentation (a), improvement in X-ray lesion after 5 days of treatment, (b) resolution of X-ray at the time of discharge (c)

The human immunodeficiency virus serology was negative. In view of hypoxic respiratory failure and occupational history of contact with suspected organic dust for 25 years with recent insult, computed tomography (CT) was done. The CT showed patchy opacities, septal thickening, and diffuse ground-glass opacities (Fig. 2).

After the clinical stabilization of the patient, fiber-optic bronchoscopy was done. No pathology was detected in the bronchial segments. Bronchoalveolar lavage (BAL) fluid showed 45% lymphocytes and was negative for acid-fast bacilli, GeneXpert, fungal stain, and malignant cells. BAL fluid culture also showed no growth of pyogenic, fungus, and *Mycobacterium tuberculosis*. The transbronchial lung biopsy (TBLB) showed non-specific inflammation. The connective tissue serology was normal (Table 1). Based on the relevant occupational history of the patient and the investigations done, he was diagnosed as a case of HP (Bagassosis). The diagnosis was also confirmed on the basis of criteria given by Schuyler and Cormier and Vasakova *et al*. document on HP diagnosis [5,6].

The patient was managed with glucocorticoids and supportive therapy in the form of O_2 inhalation, bronchodilators, and non-invasive ventilation. He improved clinicoradiologically and the requirement of O_2 inhalation was substantially decreased in 10 days. He was discharged on oral steroids (0.5 mg/kg) for the next 6 months, with gradual tapering doses, and an advisory to prevent further exposure to the sugarcane dust. Follow-up chest X-ray and spirometry at the 1st, 2nd, and 6th months also showed progressively significant improvement (Table 2 and Fig. 1b and c).

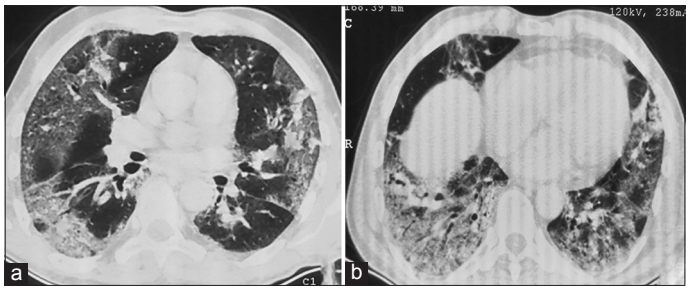

Figure 2: (a and b) Computed tomography showing bilateral diffuse patchy opacity with ground-glass opacities and septal thickening

Table 1: Blood investigations and results of the patient

Investigations	Results
ANA/dsDNA	Negative
Rheumatoid factor	Negative
HbsAg and anti HCV	Negative
C-reactive protein	Negative
Blood culture	Negative
Sputum for AFB	Negative
Urine microscopy	Normal
Urine culture	Sterile

ANA: Antinuclear antibodies, dsDNA: Anti-double-stranded DNA, HbsAg: Hepatitis B surface antigen, Anti-HCV: Anti-hepatitis *C* virus, AFB: Acid-fast bacillus

Table 2: Serial pulmonary function tests of the patient

Spirometry	At discharge (%)	1 month (%)	2 months (%)	6 months (%)
FEV_1	1.91 (65)	2.14 (73)	2.29 (79)	2.24 (78)
FVC	2.48 (67)	2.76 (75)	3.05 (83)	2.92 (80)
FEV1/FVC	77.11 (90)	77.38 (90)	74.99 (97)	76.56 (89)

FEV_1: Forced expiratory volume in 1st second, FVC: Forced vital capacity

DISCUSSION

Bagassosis is a type of HP caused by inhalation of air-borne antigens from bagasse involving complex immunological reactions, which may lead to interstitial inflammation and alveolar destruction causing significant loss of pulmonary function and disability [7]. It is occupation-related HP seen commonly before the 1970s. It is presently quite rare in the developed world due to the development of sugarcane industries into an organized sector. The first case of bagassosis in India was reported from a paper mill of Kolkata in 1955 [8]. Since then, only a few cases have been reported from India, nonetheless, one of the largest sugar manufacturing countries in the world [9,10].

The largest interstitial lung disease (ILD) study from India showed HP as the most common type of ILD, contributing nearly 48% of ILD cases. Out of these, only about 5.8% of cases were occupational-related HP and, moreover, without a single case of bagassosis [11]. Another major study by Dhooria *et al*. [12] also did not report a single case of bagassosis. It is opined that the low incidence may be due to the lack of awareness in unorganized sector of these industries, different prodromal periods with varying duration of exposure, milder form of the disease, and

no regular screening policy of the workers even in the organized sector.

The pathogenesis of this disease starts from the inhalation of respirable dust from the sugarcane residue. On an inhalation, the dust particles containing the antigen enter the respiratory bronchioles. The further interaction between the respiratory bronchiole and the antigen depends on the genetic predisposition and environmental factors, explaining why only a few people get affected among all equally exposed individuals [1,4,13]. On exposure to antigens, sensitization takes place in some individuals while repeated insult activates a cascade of immune-mediated reactions involving cytokines to cause alveolitis [14]. The chronic exposure to the antigen results in a fibrotic stage, indistinguishable from the usual interstitial pneumonia pattern, which manifests on CT as a mosaic attenuation, signifying the role of antigen in affecting small airways as well, apart from reticular opacities, traction bronchiectasis, and honeycombing [15]. The diagrammatic presentation of etiopathogenesis is shown in Fig. 3.

The emphasis should be given on the significance of occupational history in the patient with respiratory symptoms to arrive at the diagnosis. Early recognition is necessary if the history of exposure to sugarcane dust is present, irrespective of the occupation. This is the scenario in rural India where people are exposed to it even without occupation in the sugar industry. The radiological findings of HP are upper and middle lobe predominant ground-glass opacities, centrilobular nodules, mosaic attenuation with fibrosis, and honeycombing in chronic disease [5,15]. The specific antigen detection of the offending agents should be done in all suspected cases with serum specific IgG test. Bronchoscopy, TBLB, and BAL are also needed for the confirmation of the disease. Lymphocytosis on BAL and loose form granuloma with bronchocentric lymphocytic infiltrate is highly suggestive of HP [5]. Our case fulfills the diagnosis criteria of HP diagnosis given by Schuyler and Cormier and Vasakova *et al*. [5,6]. Schuyler and Cormier diagnostic criteria consist of six major and three minor criteria and our case satisfies four major and two minor diagnostic criteria, while as per Vasakova *et al*., HP diagnostic document of 2017, our case also fulfills the probable diagnosis of HP by clinical and radiological evidence of ILD. Our patient had a history of exposure for 25 years with BAL lymphocytosis.

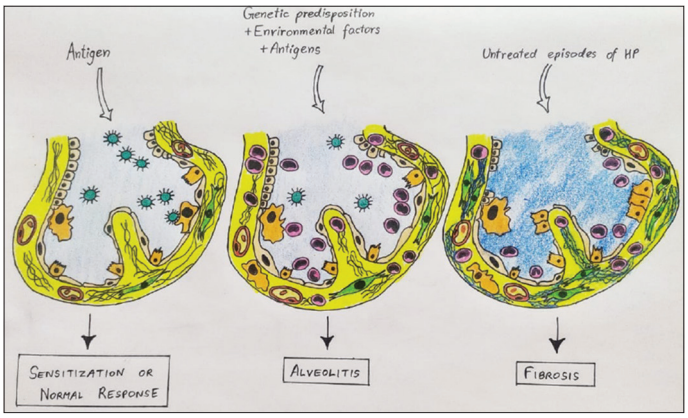

Figure 3: Schematic diagram of pathogenesis of hypersensitivity pneumonitis

Occupational health education in India is the need of the current scenario to avoid work-related hazards. In view of the paucity of reported cases and the number of occupational medicine physicians, occupation-related hazards enter into a vicious cycle. These patients get symptomatically treated for the condition for a short period of time but again land themselves with the same presentation, due to the lack of recognition of the actual cause. The mainstay of the treatment of bagassosis is to avoid the antigen, which in our case was the exposure to the bagasse for a long span of time. Moreover, the patient was not using respirators while working and also was unaware of its application to protect himself from this occupational hazard. HP may lead to subsequent fibrosis and make patients progressively debilitating.

Hence, early diagnosis and treatment is warranted with avoidance of antigen which may improve symptoms and lead to a better quality of life. In our case, the patient improved from the stage of alveolitis and was advised to refrain from further contact with these antigens and use respirators/personal protective equipment in case of unavoidable circumstances. Our case is a rare form of occupational hazard who presented with acute hypoxemic respiratory failure requiring non-invasive mechanical ventilation and later well responded to systemic corticosteroids.

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CONCLUSION

Bagassosis is a rare form of HP commonly seen before the 1970s. However, despite one of the largest producers of sugar, India rarely reported bagassosis which may be due to the lack of awareness and under-reporting in the unorganized sector. Awareness among the people, limiting exposure to antigen and work-related hygiene, may prevent irreversible progression.

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