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Idiopathic bronchiolitis with features of diffuse panbronchiolitis in an African-American patient with hepatitis C virus infection

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Abstract Diffuse panbronchiolitis (DPB) is an idiopathic inflammatory process involving respiratory bronchioles, largely restricted to Japanese people and associated with HLA Bw54. We report a case of idiopathic bronchiolitis with DPB features in an African American with hepatitis C virus infection, correlated with postmortem anatomic findings. The 53-year-old patient presented with shortness of breath and productive cough. Examination revealed hypercapnic respiratory failure. Lung computed tomography showed diffuse centrilobular nodules and branching linear opacities, whereas lung biopsy demonstrated diffuse peribronchiolar fibrosis and chronic inflammation with bronchiolectasis. He died 37 days postadmission. Autopsy revealed numerous bronchiolocentric nodules with bronchiolectasis and sheets of foamy macrophages in alveolar septa and spaces. This is a rare example of idiopathic bronchiolitis with features of DPB in an hepatitis C virus–infected African-American patient. Hepatitis C virus infection is known to be associated with extrahepatic pulmonary manifestations, and DPB may be one of these. Early diagnosis will allow appropriate treatment and may slow the disease progression.
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Keywords: Diffuse panbronchiolitis; Idiopathic bronchiolitis; Bronchiolitis; Hepatitis C virus infection; African American

1. Introduction

Diffuse panbronchiolitis (DPB) is a distinct clinicopathologic entity characterized histologically by chronic inflammation of the respiratory bronchioles. This entity was initially described by Yamanaka et al in 1969 [1] and is largely restricted to the Japanese people. After its initial description in Japan, cases were recognized in other Asian countries, and subsequently, there have been case reports from Europe and North America [2,3]. We report a case of idiopathic bronchiolitis with features of DPB in an African-American patient with hepatitis C virus (HCV) infection.

2. Case report

This 53-year-old African-American male with history of chronic obstructive pulmonary disease, HCV infection, and 30 pack-years of smoking presented with shortness of breath and cough productive of copious sputum. Examination revealed respiratory distress, basal coarse crackles bilaterally, and hypercapnic respiratory failure on controlled oxygen therapy (pH 7.34, p_{O_2} 55 mm Hg, p_{CO_2} 59 mm Hg). Laboratory results included a total immunoglobulin E of 1028 IU/L, negative *Aspergillus fumigatus* precipitans, mildly elevated rheumatoid factor (24 IU/mL, normal <20 IU/mL), and normal α1-antitrypsin level. Sputum culture grew *Pseudomonas aeruginosa*. Liver enzymes were normal, with a high HCV antibody titer. Computed tomography scan showed mucosal thickening in the

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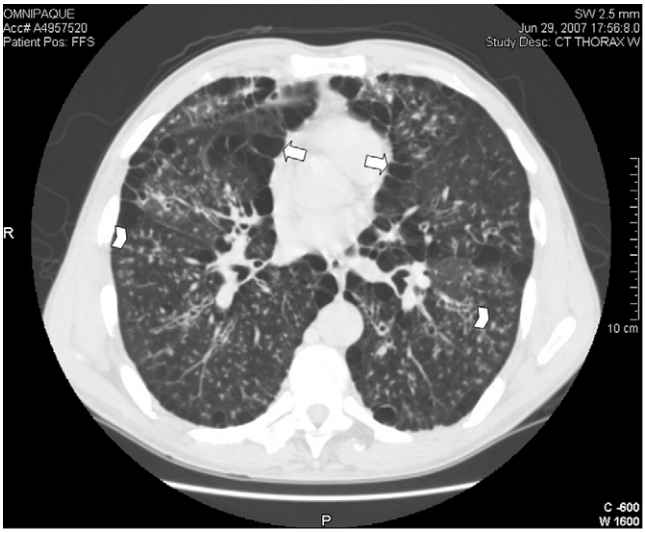

Fig. 1. Chest CT at last admission demonstrating diffuse "tree in bud" opacities (arrowheads), bronchial wall thickening, and mild bronchiectasis. Paraseptal emphysematous changes were also noted (arrows).

maxillary and ethmoid sinuses. The patient did not have a history of exposure to toxic fumes or any significant disease in other organs.

Medications included inhaled triamcinolone and ipratropium/salbutamol. He had been intubated on 3 previous occasions. Computed tomographic scan of the lung performed at his last admission showed diffuse small centrilobular nodules and branching linear opacities bilaterally (Fig. 1). Peribronchial nodularity was also noted along with significant diffuse bronchial wall thickening. Mild to moderate cylindric bronchiectasis was present. These findings suggested bronchiolocentric inflammatory changes and some degree of scarring. Significant paraseptal emphysematous changes were noted. Comparison with computed tomography from 8 months previous showed similar and stable findings. Wedge biopsy of the lung 7 days postadmission demonstrated diffuse and widespread peribronchiolar dense fibrosis and chronic inflammation with extension to the adjacent alveolar septa, without involvement of the alveolar spaces (Figs. 2 and 3). Lymphocytes and plasma cells were identified, with focal lymphoid follicle formation. Acute inflammatory exudates were present in the bronchiolar lumen and epithelium with bronchiolectasis. A small number of foamy macrophages were identified in the alveolar septa adjacent to the bronchioles. Clinical findings raised the differential diagnosis of DPB, cystic fibrosis, primary ciliary dyskinesia, bronchiectasis, and idiopathic bronchiolitis.

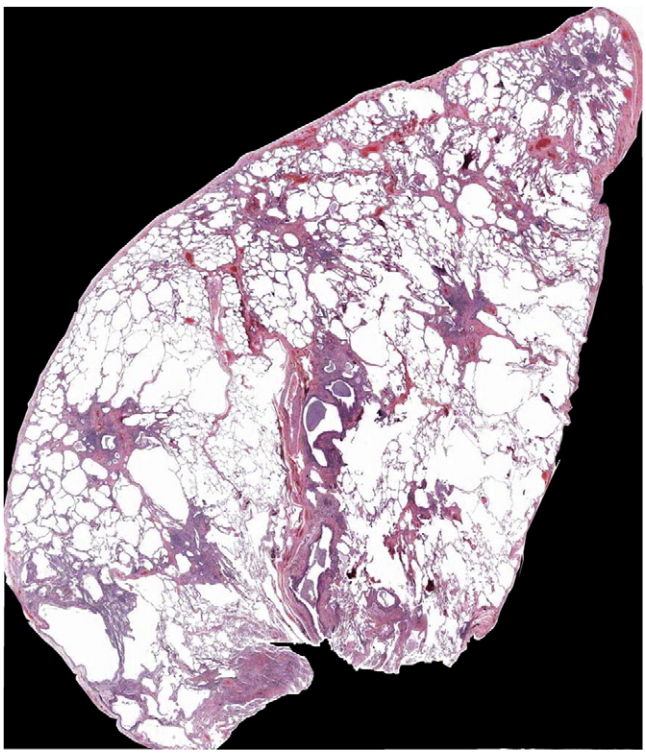

Fig. 2. Scanning power microscopy shows the bronchiolocentric distribution of the inflammatory process.

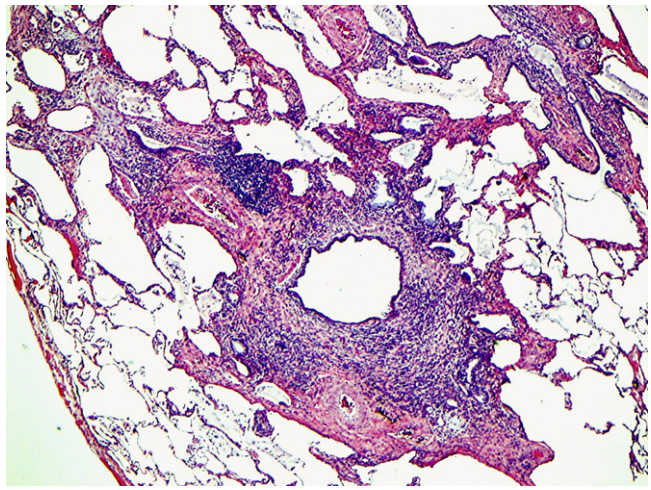

Fig. 3. Higher magnification shows peribronchiolar fibrosis and chronic inflammation with bronchiolectasis.

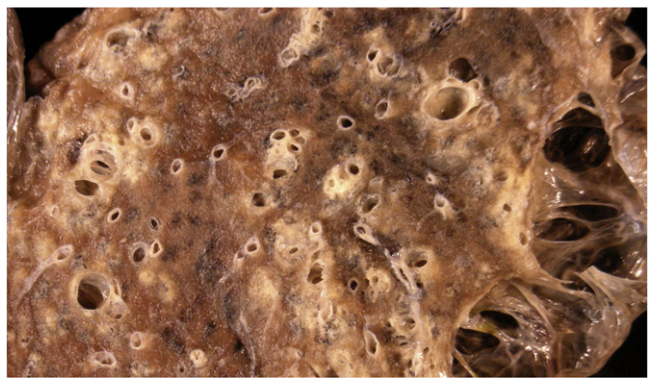

Fig. 4. Gross examination of the lung shows small tan-yellow nodules centered around bronchioles. Paraseptal emphysematous changes are also seen.